

PEDIATRICS®

OFFICIAL JOURNAL OF THE AMERICAN ACADEMY OF PEDIATRICS

Language Ability in Children With Permanent Hearing Impairment: The Influence of Early Management and Family Participation

Peter Watkin, Donna McCann, Catherine Law, Mark Mullee, Stavros Petrou, Jim Stevenson, Sarah Worsfold, Ho Ming Yuen and Colin Kennedy

Pediatrics 2007;120:e694-e701

DOI: 10.1542/peds.2006-2116

The online version of this article, along with updated information and services, is located on the World Wide Web at:

<http://www.pediatrics.org/cgi/content/full/120/3/e694>

PEDIATRICS is the official journal of the American Academy of Pediatrics. A monthly publication, it has been published continuously since 1948. PEDIATRICS is owned, published, and trademarked by the American Academy of Pediatrics, 141 Northwest Point Boulevard, Elk Grove Village, Illinois, 60007. Copyright © 2007 by the American Academy of Pediatrics. All rights reserved. Print ISSN: 0031-4005. Online ISSN: 1098-4275.

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™



Language Ability in Children With Permanent Hearing Impairment: The Influence of Early Management and Family Participation

Peter Watkin, MBBS, MSc^a, Donna McCann, PhD^b, Catherine Law, MBBS, MD^c, Mark Mullee, MSc^d, Stavros Petrou, PhD^e, Jim Stevenson, PhD^f, Sarah Worsfold, BSocSc^b, Ho Ming Yuen, MSc^d, Colin Kennedy, MBBS, MD^b

^aAudiology Department, Whipps Cross University Hospital, London, England; ^bDepartment of Child Health, ^cPublic Health Sciences and Medical Statistics/Research and Development Support Unit, and ^dDepartment of Psychology, University of Southampton, Southampton, England; ^eInstitute of Child Health, University College, London, England; ^fNational Perinatal Epidemiology Unit, Oxford, England

The authors have indicated they have no financial relationships relevant to this article to disclose.

ABSTRACT

OBJECTIVE. The goal was to examine the relationships between management after confirmation, family participation, and speech and language outcomes in the same group of children with permanent childhood hearing impairment.

METHODS. Speech, oral language, and nonverbal abilities, expressed as *z* scores and adjusted in a regression model, and Family Participation Rating Scale scores were assessed at a mean age of 7.9 years for 120 children with bilateral permanent childhood hearing impairment from a 1992–1997 United Kingdom birth cohort. Ages at institution of management and hearing aid fitting were obtained retrospectively from case notes.

RESULTS. Compared with children managed later (>9 months), those managed early (≤9 months) had higher adjusted mean *z* scores for both receptive and expressive language, relative to nonverbal ability, but not for speech. Compared with children aided later, a smaller group of more-impaired children aided early did not have significantly higher scores for these outcomes. Family Participation Rating Scale scores showed significant positive correlations with language and speech intelligibility scores only for those with confirmation after 9 months and were highest for those with late confirmed, severe/profound, permanent childhood hearing impairment.

CONCLUSIONS. Early management of permanent childhood hearing impairment results in improved language. Family participation is also an important factor in cases that are confirmed late, especially for children with severe or profound permanent childhood hearing impairment.

www.pediatrics.org/cgi/doi/10.1542/peds.2006-2116

doi:10.1542/peds.2006-2116

Key Words

universal newborn screening, congenital deafness, confirmation, management, hearing aids, outcomes, family participation

Abbreviations

PCHI—permanent childhood hearing impairment
 UNS—universal newborn screening
 TROG—Test for Reception of Grammar
 BPVS—British Picture Vocabulary Scales
 CCC—Children's Communication Checklist
 SIR—Speech Intelligibility Rating Scale
 FPRS—Family Participation Rating Scale
 IQR—interquartile range

Accepted for publication Feb 1, 2007

Address correspondence to Peter Watkin, MBBS, MSc, Audiology Department, Whipps Cross University Hospital NHS Trust, Whipps Cross Road, Leytonstone, London E11 1NR, England. E-mail: peter.watkin@whippsx.nhs.uk

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275). Copyright © 2007 by the American Academy of Pediatrics

PERMANENT CHILDHOOD HEARING impairment (PCHI) is relatively common, with significant bilateral impairment of ≥ 40 dB hearing level affecting 1.2 infants per 1000 live births. The inability to hear disrupts communication and, when present from birth, affects social, emotional, and linguistic development. Educational achievement is reduced, and the costs to the child, family, and society are substantial. It is now possible to screen the hearing of newborn infants, and several studies have addressed the issue of whether early detection and management reduce the impact of the condition.¹⁻⁴ However, a 2001 systematic review by the US Preventive Services Task Force concluded that it remained unclear whether universal newborn screening (UNS) and early identification of PCHI were associated with improved language abilities.⁵ More recently, we reported a benefit of UNS and confirmation of PCHI by 9 months of age in the adjusted language scores of children with PCHI in a 5-year (1992-1997) birth cohort.⁶ We also reported that early confirmation was cost-effective.⁷ The present report on the same birth cohort examines the role of events subsequent to confirmation and of family participation on speech and language. In contrast to studies confined to children identified by UNS and enrolled in a single intervention program, one half of the children described here were not born during periods with UNS and there was considerable variation in the ages of confirmation and management.

METHODS

We studied children with moderate or worse deafness in the better-hearing ear, categorized through 4-frequency averaging of pure-tone thresholds from 500 Hz to 4000 Hz. In cases in which pure-tone threshold data were unavailable, sound field and electrophysiological test results were used. Children with a hearing impairment that was known to have been acquired were not included. The sample was drawn from the Wessex and Greater London regions of southern England. Our study was approved by the South and West Multicenter Research Ethics Committee (Dartington, England). Principal caregivers provided written informed consent.

The Wessex subgroup comprised all such children in the birth cohort enrolled in the Wessex controlled trial in which UNS was or was not provided in alternate 4- to 6-month periods from 1993 to 1996.⁸ The Greater London subgroup comprised all such children in the 5-year (1992-1997) birth cohort born either in the area served by the Whipps Cross and Hillingdon UNS programs⁹⁻¹¹ or in the neighboring districts of Redbridge and Brent and Harrow, respectively, neither of which provided UNS. Therefore, approximately one half of the Wessex cohort and approximately one half of the Greater London cohort, together summing to 157 000 births, were born in cohorts that received UNS. Ascertainment of deafness in this birth cohort was through prospective follow-up

monitoring for both the Wessex and Greater London subgroups.¹²⁻¹⁵

Receptive skills for oral language were assessed with the Test for Reception of Grammar (TROG)¹⁶ and the British Picture Vocabulary Scales (BPVS).¹⁷ Expressive skills for oral language were measured by using the Renfrew Bus Story.¹⁸ The "5 longest sentences" and "sentence information" scores were used to assess both semantic and syntactic skills. The effects of confirmation by 9 months of age and of birth in periods with UNS on *z* scores of both measures of receptive language and both measures of expressive language were similar.⁶ Therefore, we report here their effects on aggregate *z* scores for receptive language and for expressive language, rather than their effects on all 4 scores. These assessments were undertaken in the child's home by researchers who were blind to the audiologic history. A speech and language therapist employed British Sign Language for children who signed. Speech was assessed by using the Children's Communication Checklist (CCC)¹⁹ and the Speech Intelligibility Rating Scale (SIR).²⁰ The SIR is rated on a scale from 1 to 5 (ranging from "not intelligible" to "intelligible to all"). This measure is not suitable for hearing children but is appropriate for use with deaf children with limited speech production, even when their main communication is by sign. Ratings by the classroom teacher were used to reflect the child's speech intelligibility to an adult outside the family. The speech scales of the CCC were used as a parental assessment of speech quality, whereas the SIR provided a rating of the child's everyday speech. Scores for signing ability in British Sign Language are not included in the present report because of the small number of observations ($n = 9$) and the doubtful validity of combining assessments of oral and signed language scores. The nonverbal ability of the child was assessed by using Raven's Progressive Matrices²¹ and was used to calculate verbal/nonverbal difference scores. Reading, pragmatic features of communication, behavior, and other secondary outcomes were also assessed but are not reported here.

Speech and oral language scores for the children with PCHI were expressed relative to the group mean score on the same measure for 63 hearing children, matched according to place of birth and age at assessment. The mean and SD of age-adjusted scores for the hearing children were calculated; from these, the age-adjusted *z* scores for children with PCHI were derived, with the *z* scores being equal to the number of SDs by which the age-adjusted scores differed from the group mean score for the hearing children. Use of *z* scores made possible the derivation of aggregate scores (eg, *z* score for receptive language = [TROG *z* score + BPVS *z* score]/2) and difference scores (eg, *z* score for deficit of receptive language, compared with nonverbal skills = *z* score for receptive language - *z* score for nonverbal ability). Ratings rather than *z* scores were used for the SIR because

this assessment was not suitable for use with the normally hearing group.

The age of confirmation of PCHI was defined as the age of the first measurement of increased threshold with a reliable, age-appropriate, audiologic test, and the age of enrollment was defined as the age when the audiologist, in conjunction with a teacher of the deaf (or a therapist), introduced an individual family plan or equivalent. Confirmation, management, and aiding of PCHI were categorized as early or late according to occurrence by or after 9 completed months of age. This was consistent with the previous reports of the Wessex Trial,⁸ with other reports of substantial benefits to language associated with early identification by 6 months, followed by onset of ongoing intervention after a mean interval of an additional 3 months,¹ and with the US Preventive Services Task Force benchmark of diagnosing or treating before 10 months.⁵ The interval between the age of confirmation of PCHI and the age of hearing aid fitting was obtained from case notes by the researchers.

Teachers of the deaf and audiologists were asked to rate independently family participation in the child's management, from 1 to 5 (limited participation to ideal participation) on the Family Participation Rating Scale (FPRS) devised by Moeller.² That author characterized the quality of family participation by using the average of independent ratings assigned by ≥ 2 early interventionists who had worked directly with the family over a period of several years; the author then related those ratings to language outcomes at the age of 5 years. In the current study, the FPRS was scored independently by audiologists and teachers of the deaf who had provided varying aspects of long-term, multiagency management for the child and who were blinded to the results of the study outcomes. The scale descriptors enabled the audiologists to rate overall participation in clinical aspects of management and the teachers to rate family involvement in the child's education. With these FPRS scores categorized as less than average, average, or above average, there was categorical agreement for 41 (52%) of 79 children for whom scores were available from both audiologists and teachers, but Cohen's κ was 0.22, demonstrating only fair agreement. There was only exact agreement for 28 (35%) of the 79 cases with κ of 0.14. The audiologists' and teachers' scores were therefore used for separate analyses. Audiologists had been responsible for the management from the time of identification, during which period the teacher usually had not worked directly with the families. Therefore, the audiologists' FPRS scores alone were used for examination of the relationship between early confirmation and FPRS scores. Teachers had been directly responsible for delivering the long-term educational program to the family, and their ratings of family participation were used for examination of the relationship between FPRS scores and language outcomes. These relationships were

examined with and without exclusion of those enrolled in either a signed communication or cochlear implant program.

Power calculations were reported previously⁶ (J.S., D.M., P.W., S.W., and C.K., unpublished data, 2005). Statistical analyses tested the effects of the exposure variables (early confirmation, management, and aiding of PCHI) on speech and language outcomes by using Student's *t* test. Multivariate linear regression analysis (Stata 8; Stata Corp, College Station, TX) examined the effects of these variables, adjusted for severity of hearing impairment, maternal education, and nonverbal ability, on speech and language scores. Ordinal regression was also used in the analysis of speech intelligibility. The effect of adding FPRS scores to the regression model was also examined. Basic descriptive statistics, χ^2 analysis, Mann-Whitney *U* test, and analysis of variance were used to examine differences between groups, and Spearman's ρ was used to examine correlations between variables.

RESULTS

Participants

Of the 168 children with PCHI in the study sample, 2 were untraceable and 6 were not approached because of the presence of other severe, active, medical illnesses that made the study inappropriate. Of those approached, 120 (75%) agreed to participate, with 15 (9%) refusing study participation and 25 (16%) not responding. No clinically important differences were found in the age, gender, or severity of hearing loss of the participants and nonparticipants. Of the 120 participants, 61 had been born in cohorts that had received UNS. Their clinical characteristics are compared with those of the 63 children in the normally hearing comparison group in Table 1. These 2 groups were similar with respect to maternal educational level and occupation of the head of the household.

Participants had a mean age of 7.9 years (SD: 1.3 years) at the time of assessment of speech and oral language. Not all participants had scores that could be included in the analyses. Sign language alone or entirely nonverbal communication was used by 23 participants. Of the 23 nonverbal children, 15 were unable to complete an oral assessment of receptive language, and 1 additional child who communicated orally did not complete an assessment of receptive language. A measure of receptive language was therefore available for 104 children. Another 9 children communicated orally but assessments of expressive language were incomplete. Therefore, 88 of the participants had a measure of expressive language. Missing data in any one variable reduced the number of children with *z* scores who could be included in the multivariate linear regression analyses; 101 children with a receptive language score and 87

TABLE 1 Characteristics of the Hearing-Impaired and Normally Hearing Children Enrolled in the Study

	n (%)	
	Hearing-Impaired (N = 120)	Normally Hearing (N = 63)
Male gender	67 (56)	37 (59)
Communication method		
Oral	86 (72)	63 (100)
Exclusive use of sign communication	16 (13)	0 (0)
Oral and sign communication	11 (9)	0 (0)
Nonverbal or by gesture	7 (6)	0 (0)
English first language	99 (82)	60 (95)
Medical disorders		
Cerebral palsy	5 (4)	0 (0)
Visual disability	5 (4)	0 (0)
Learning disability	10 (8)	0 (0)
Chromosomal/syndromic	14 (12)	1 (2)
None	97 (81)	62 (98)
Nonverbal ability ^a		
≥75th percentile	38 (32)	28 (44)
26th to 74th percentile	21 (18)	17 (27)
≤25th percentile	49 (41)	18 (29)
Not known	12 (10)	0 (0)
Degree of hearing loss ^b		
Moderate (40–69 dB HL)	65 (54)	0 (0)
Severe (70–94 dB HL)	29 (24)	0 (0)
Profound (>95 dB HL)	26 (22)	0 (0)
Age at time of study, y		
<7	36 (30)	13 (21)
7–9	62 (52)	37 (58)
>9	22 (18)	13 (21)

HL indicates hearing level.

^a Raven's Progressive Matrices.²¹

^b Averaged from 500 to 4000 Hz in the better-hearing ear.

with an expressive language score were available for these analyses. The speech scales of the CCC were completed for 101 children, but for similar reasons only 95 could be included in the analysis. Speech intelligibility was rated by the teachers of 113 children. Of the 7 missing children, 2 had multiple disabilities and 4 communicated by sign alone. Hearing aids were fitted for all participants whose oral language was assessed. Cochlear implants were fitted for 16, 9 of whom had their deafness confirmed by 9 months and 5 of whom were born in periods with UNS.

Ages of Enrollment in Management and Hearing Aid Fitting

The median ages of confirmation of PCHI, enrollment in a management program, and fitting of hearing aids were 10 months (interquartile range [IQR]: 3–25.5 months), 13 months (IQR: 8–32 months), and 15 months (IQR: 10–40 months), respectively. Information on timing of educational input was missing for 11 children (9%). Of the remaining 109 children, 65 (60%) and 95 (87%) received educational support within 1 and 3 months after confirmation, respectively. There were no significant differences in the ages of enrollment for management or the ages of hearing aid fitting between Greater

London and Wessex participants (Mann-Whitney test, $P = .61$ and $P = .76$, respectively).

Effect of Severity of Deafness

The 55 children with severe or profound PCHI had their deafness confirmed at a median age of 10 months (IQR: 3–14 months), as did the 65 with moderate hearing impairment (IQR: 2–40.5 months). However, those with severe or profound deafness were enrolled for management and aided significantly earlier than were those with moderate impairment (median ages of 11 months [IQR: 7–15 months] vs 20 months [IQR: 9–43.5 months]; $P = .004$; and 11 months [IQR: 7–19.5 months] vs 28 months [IQR: 12.5–49.5 months]; $P < .001$, respectively). The interval between confirmation and aiding was significantly greater for those with moderate impairment than for those with severe or profound deafness (3 months [IQR: 1–18 months] versus 1 month [IQR: 1–2.25 months]; $P = .003$). The median interval from confirmation to aiding was short (1 month; IQR: 0.5–4 months) for all children with severe or profound deafness, regardless of whether the PCHI was confirmed early or late, but the interval was greater for those with moderate PCHI when the PCHI had been confirmed by 9 months than when it was confirmed later (10.5 months [IQR: 3–30 months] vs 1 month [IQR: 1–2 months]; $P < .005$).

Effect of Birth in Periods With UNS

The median age of confirmation of PCHI was 3 months (IQR: 1–12 months) for children born in periods with UNS, compared with 13 months (IQR: 9–41 months) for those born in periods without UNS ($P < .001$). Compared with those born in periods without UNS, enrollment in management was also achieved earlier (10 months [IQR: 4–24 months] vs 18 months [IQR: 11–42 months]; $P = .001$). The median age of aiding for children born in periods with UNS was one half of that for those born in periods without UNS (12 months [IQR: 6–32 months] vs 24 months [IQR: 12–42 months]; $P = .003$). This lower age at aiding applied both for those with moderate PCHI (20 months [IQR: 9–41 months] vs 39 months [IQR: 20–55 months]; $P = .029$) and for those with severe/profound PCHI (9 months [IQR: 3–12 months] vs 13 months [IQR: 11–27 months]; $P = .002$).

Effect of Early Management and Hearing Aid Fitting

PCHI was confirmed by 9 months for 57 of the children. However, only 41 were enrolled in a management program and 29 were fitted with hearing aids by this age. The ability to examine the effect on outcomes of these exposure variables in this group was reduced by progressive increases in the proportions that had multiple disabilities and/or used sign communication exclusively or were nonverbal at the time of the outcome assessments (Table 2). There were consequent reductions in the

TABLE 2 Comparison of the Groups Confirmed, Managed, and Aided by 9 Months of Age

	n (%)		
	Confirmed at ≤9 mo (N = 57)	Managed at ≤9 mo (N = 41)	Aids Fitted at ≤9 mo (N = 29)
Characteristics			
Profound deafness (>95 dB HL)	13 (23)	11 (27)	9 (33)
Multiple disabilities	15 (26)	13 (32)	12 (41)
Exclusive use of sign communication	8 (14)	7 (17)	6 (21)
Communication nonverbal or by gesture	4 (7)	4 (10)	4 (14)
Outcome measures			
No oral language scores	11 (19)	10 (24)	9 (31)
No CCC (speech) scores	13 (23)	12 (29)	11 (38)
No SIR scores	3 (5)	2 (5)	2 (7)

HL indicates hearing level.

number of oral language and CCC speech scores for these groups (Table 2). Compared with scores for those with later managed losses, enrollment in management of PCHI by 9 months was associated with significantly higher adjusted mean expressive and receptive language and language/nonverbal difference scores but no significant intergroup difference in CCC speech scores (Table 3). This pattern of benefits was similar to that reported previously for this sample in association with confirmation of PCHI by 9 months⁶ (Table 3). The effect of early confirmation (or early management) on the SIR scores (median scores: 4 [IQR: 2.8–5] vs 4 [IQR: 3–5]), adjusted in an ordinal regression analysis, was also not significant

($P = .22$). The size of effect on language outcomes was much smaller for early aiding than for early management (0.15–0.31 vs 0.66–0.74 SDs), and no significant differences in group mean scores for any language or speech measure were seen between those aided by 9 months and those aided later (Table 3).

Effect of Family Participation

The audiologist FPRS scores did not correlate with age at confirmation of PCHI ($r = -0.09$; $P = .376$) but did show negative correlations with age of enrollment into management ($r = -0.24$; $P = .022$), age of hearing aid fitting ($r = -0.24$; $P = .020$), and length of the interval

TABLE 3 Confirmation, Management, and Aiding of PCHI by 9 Months of Age and Language and Speech Scores

Outcome Measure	z Scores for Age						Adjusted Mean Difference (95% CI) Between Groups ^a	P
	≤9 mo			>9 mo				
	n	Mean	SD	n	Mean	SD		
Confirmation of PCHI								
Receptive language								
Aggregate score	45	-1.76	1.47	56	-2.38	1.72	0.76 (0.26–1.27)	.004
Aggregate minus nonverbal	45	-0.82	1.23	56	-1.68	1.44	0.82 (0.31–1.33)	.002
Expressive language								
Aggregate score	39	-0.59	1.31	48	-1.07	1.21	0.50 (0.00–1.01)	.050
Aggregate minus nonverbal	39	0.14	1.29	48	-0.50	1.34	0.70 (0.13–1.26)	.016
CCC speech	44	-1.15	1.42	51	-1.33	1.54	0.29 (-0.28 to 0.87)	.319
Enrollment in management								
Receptive language								
Aggregate score	30	-1.83	1.38	71	-2.13	1.70	0.66 (0.08–1.24)	.027
Aggregate minus nonverbal	30	-0.97	1.32	71	-1.43	1.44	0.68 (0.09–1.28)	.024
Expressive language								
Aggregate score	26	-0.66	1.35	61	-0.93	1.25	0.55 (-0.01 to 1.12)	.055
Aggregate minus nonverbal	26	0.10	1.35	61	-0.35	1.34	0.74 (0.11–1.37)	.023
CCC speech	29	-1.23	1.3	66	-1.26	1.56	0.16 (-0.48 to 0.81)	.618
Hearing aid fitting								
Receptive language								
Aggregate score	19	-2.17	1.40	82	-2.01	1.66	0.15 (-0.54 to 0.83)	.671
Aggregate minus nonverbal	19	-1.29	1.31	82	-1.30	1.45	0.20 (-0.50 to 0.90)	.573
Expressive language								
Aggregate score	16	-0.80	1.36	71	-0.86	1.27	0.14 (-0.52 to 0.81)	.667
Aggregate minus nonverbal	16	-0.15	1.38	71	-0.23	1.36	0.31 (-0.44 to 1.06)	.417
CCC speech	18	-1.37	1.29	77	-1.22	1.5	-0.12 (-0.88 to 0.62)	.733

^a The linear regression model adjusted for maternal educational qualifications, severity of PCHI, and, except in the case of mean difference scores, Raven's Progressive Matrices score.

between confirmation and hearing aid fitting ($r = -0.24$; $P = .023$). This interval was greater for those with below-average participation (rating of 1, 2, or 3) than those with above-average participation (rating of 4 or 5) (medians: 7.0 months [IQR: 1–24 months] vs 2.00 months [IQR: 0.3–4 months]; $P = .018$). There was a significant interaction between these effects and severity of PCHI, so that the negative correlation between FPRS scores and the interval from confirmation to aiding was strong for those with moderate PCHI confirmed before 9 months but was absent for those with moderate impairment confirmed later and for those with more-severe deafness, whenever it was confirmed (Fig 1).

Language and speech outcomes were correlated significantly with the teacher's FPRS scores for all degrees of hearing impairment, but the relationship was stronger for the families of children with severe or profound deafness who had their hearing loss confirmed after 9 months (Table 4). For profoundly deaf children, the correlations with the receptive language and speech intelligibility scores were 0.95 ($n = 9$; $P < .001$) and 0.78 ($P = .008$), respectively. The relationship was also examined by excluding the children who had been enrolled in either a signing or cochlear implant program, but there was no substantial change in the correlation for the children with severe or profound hearing impairment (Table 4).

The FPRS scores were related to severity, and inclusion of both severity and FPRS scores in the regression model therefore probably represented overadjustment. With both of these variables included in the model, however, the association between confirmation by 9 months and higher language scores remained significant and of similar size.

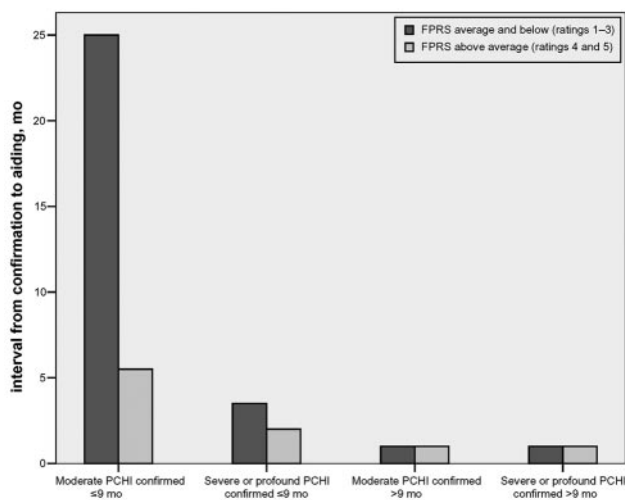


FIGURE 1
Median number of months between confirmation and hearing aid fitting, according to severity, age of confirmation, and FPRS score.

DISCUSSION

The present study indicates that, compared with children with PCHI managed later, children with PCHI managed by 9 months had higher adjusted language scores at 5 to 11 years of age. We reported previously that this was the case for those whose PCHI was confirmed early,⁶ but the present report demonstrates that benefits were also present and of similar size in the early managed group, although it was smaller and had greater proportions of children with other disabilities than the early confirmed group. These trends toward more-adverse baseline characteristics were even more apparent in the group aided by 9 months, for whom a much smaller and nonsignificant benefit in language and speech was demonstrable. For families whose ratings of participation were higher, speech and language outcomes were also higher but only for those with late confirmed losses, especially severe or profound PCHI.

Our observations support 3 earlier cohort studies that reported higher language scores after early intervention. Yoshinaga-Itano et al¹ compared 72 children identified before 6 months and 78 identified after 6 months who were enrolled in the Colorado Home Intervention Program. The children identified early had a significantly higher adjusted mean total language quotient than did those identified later (mean \pm SD: 79 \pm 25.6 vs 63.8 \pm 19.3; $P < .001$).¹ Moeller² reported a negative correlation ($r = -0.464$; $P < .01$) at 5 years between Peabody Picture Vocabulary Test scores of 112 children and the age of enrollment into the Nebraska Diagnostic Early Intervention Program. Similarly, Calderon and Naidu³ reported that age at entry significantly predicted receptive and expressive language scores ($r = 0.438$ and $r = 0.367$, respectively; $P < .005$) for 80 children at the age of 3 years who had completed the Western Washington State Early Childhood Home Instruction Program.

A limitation of those reports is that the improvements may not be generalizable beyond particular, well-developed, preschool intervention programs because, in contrast to the present study, adherence to such a program was a study requirement. The median time that elapsed between identification and hearing aid fitting was 1 to 2 months and 3 months for the Colorado Home Intervention Program and the Diagnostic Early Intervention Program, respectively, whereas identification was simultaneous with provision of amplification in the Early Childhood Home Instruction Program. However, the reported benefits in the largest of these studies,¹ calculated to be ~ 0.5 to 0.6 SDs of the distribution of language scores for normally hearing children,⁶ are similar in magnitude, despite differences in management, to those associated with enrollment in management by 9 months that we report here for both the London and Wessex subgroups of our United Kingdom sample.

It is the experience of many audiologists that not all parents of deaf children immediately engage with inter-

TABLE 4 FPRS Scores and Language and Speech Outcomes According to Degree of Hearing Loss and Timing of Confirmation of Hearing Loss, Excluding Those Who Had Received a Cochlear Implant and Those Who Communicated Through Sign Language

Degree of Hearing Impairment	Sample	Timing of Confirmation of Hearing Loss	Correlation With Receptive Language Scores		Correlation With Expressive Language Scores		Correlation With CCC Speech Scores		Correlation With SIR Rating	
			<i>n</i>	Coefficient	<i>n</i>	Coefficient	<i>n</i>	Coefficient	<i>n</i>	Coefficient
			Moderate	All	≤9 mo	27	0.26	26	0.18	27
		>9 mo	26	0.34	24	0.47 ^a	25	0.31	26	0.52 ^b
		Total	53	0.30 ^a	50	0.29 ^a	52	0.29 ^a	54	0.34 ^a
	Excluding cochlear implantees	Total	49	0.37 ^b	47	0.35 ^a	49	0.31 ^a	50	0.42 ^b
	Excluding sign language users	Total	48	0.27	48	0.28	48	0.23	49	0.31 ^a
Severe/profound	All	≤9 mo	13	0.26	8	0.63	12	0.01	21	0.01
		>9 mo	24	0.48 ^a	17	0.55 ^a	19	0.59 ^b	24	0.57 ^b
		Total	37	0.44 ^b	25	0.52 ^b	31	0.34	45	0.21
	Excluding cochlear implantees	Total	31	0.41 ^a	20	0.50 ^a	24	0.46 ^a	35	0.11
	Excluding sign language users	Total	27	0.47 ^b	22	0.48 ^a	26	0.41 ^a	29	0.40 ^a

^a*P* < .05.

^b*P* < .01.

vention, and both the Whipps Cross Hospital UNS program in the United Kingdom and the New York State UNS program⁴ found variation in the age at which children identified through newborn screening were aided. The children from the Whipps Cross Hospital UNS were enrolled as part of the London subgroup in the present sample, and we reported previously the delays in providing amplification to them.²² The challenges to early aiding in both the London and New York programs included audiologic difficulties in achieving diagnostic certainty, delays in gaining parental agreement for early intervention, noncompliant parents, infant illnesses, and milder hearing losses. Despite this, birth in periods with UNS in our present sample was associated with halving of the average age of aiding to a much lower age than had been achieved previously in the United Kingdom.²³

The authors of the Nebraska study, in which the sample had not received UNS and had a mean age of enrollment into the Diagnostic Early Intervention Program of 22 months, reported a strong correlation (*r* = 0.65) between family participation in management and language outcomes for deaf children at the age of 5 years. Our observations were similar for children with severe or profound deafness who were enrolled in management after 9 months of age. Both studies are therefore consistent with the idea that family participation can help to compensate for the deleterious effect of late management on language. We demonstrated a weaker relationship for children who received confirmation early but had moderate impairment. It is likely that the positive effects of the family are partly submerged in the complex factors that influence improved outcomes for children with lesser degrees of hearing loss. There was no evidence from the present study that the family participation was related to the use of signed communication or enrollment in a cochlear implant program. We cannot exclude the possibility that improved outcomes

led to higher ratings of family participation, but the ratings were made in a blinded manner with respect to the speech and language assessments, and management and confirmation data were collected independently from the case records by the researchers.

Only one half of children who had their deafness confirmed by 9 months had their hearing aids fitted by that age, and they tended to be those with the most adverse baseline characteristics. The delays in aiding, the restricted sample size, and the greater severity of impairment for those aided by 9 months reduced our ability to confirm any positive effect of early hearing aid fitting. Although severity of deafness and nonverbal ability were adjusted for in the regression model, the effect size of early aiding on language outcomes was only 23% to 42% of the effect size of early management. This suggests the possibility that failure to show a benefit of early aiding may not be entirely attributable to the smaller numbers and greater severity in the early aided group. The discrepancy between the benefits of early management and early aiding may therefore reflect the importance of other aspects of early management that act together with improved early auditory input. Early parental awareness and modification of communication strategies may have greater effects on outcomes for those with more-severe deafness.

Compared with later confirmation, confirmation of hearing impairment by 9 months of age was associated with scores on individual language measures that were higher by 0.6 to 0.9 SDs (equivalent in effect size to 9–13 verbal IQ points) of the distribution of scores in our normally hearing comparison group for receptive skills (TROG and BPVS) and ~0.5 SDs for expressive skills (Bus Story sentence information and 5 longest sentences). This was equivalent to reductions in the deficits in language skills, relative to the normally hearing comparison group, by 23% to 33% for receptive skills and

35% to 40% for expressive skills. This report shows the complex interaction between age of confirmation, severity of impairment, family participation, and events subsequent to confirmation. With the additional program development and guidance now available for the national United Kingdom program, improvements in management and outcomes can be expected, and we anticipate that delivery of early intervention to a greater proportion of those whose impairment is confirmed early will lead to greater reductions in language deficits in current and future birth cohorts. It seems likely that this mean difference in language abilities would be of disproportionately greater benefit to children with hearing impairment than it would be to normally hearing children, both because it is relative to a lower baseline level, and the benefit is therefore more likely to lift the children above a threshold level of functional importance, and because of the comorbidities in the hearing-impaired population that lead to disadvantages in other ways.

ACKNOWLEDGMENTS

We are grateful for the funding support of the Wellcome Trust (grant 061839).

We gratefully acknowledge the assistance received from children, families, school staff members, specialist teachers of the hearing impaired, speech and language pathologists, and audiology professionals in the local area teams. We particularly thank senior audiology staff members Dr Margaret Baldwin, Dr Joy Bhattacharya, Dr Alyson Bumby, Irene Curtis, Dr Carol Hunter, Dr David Reed, Scott Richards, Sue Robinson, Dr Peter Savundra, Huw Thomas, and Dr Tim Williamson and teacher of the hearing of hearing impaired Jan Nanor. We also thank the team of researchers (Helen Davis, Shirley Golden, Eleanor Lutman, Kristen Paul, and Helen Ryder) for their excellent work. We thank Julie Brinton and Sue Robinson for their advice on assessments of speech and language and on audiological aspects of the study steering group, respectively.

REFERENCES

1. Yoshinaga-Itano C, Sedey AL, Coulter DK, Mehl A. Language of early- and later-identified children with hearing loss. *Pediatrics*. 1998;102:1161–1171
2. Moeller M. Early intervention and language development in children who are deaf and hard of hearing. *Pediatrics*. 2000;106(3). Available at: www.pediatrics.org/cgi/content/full/106/3/e43
3. Calderon R, Naidu S. Further support for the benefits of early identification and intervention for children with hearing loss. *Volta Rev*. 2000;100:53–84
4. Dalzell L, Orlando M, McDonald M, et al. The New York State universal newborn hearing screening demonstration project: age of hearing loss identification, hearing aid fitting and enrollment in early intervention. *Ear Hear*. 2000;21:118–130
5. Thompson DC, McPhillips H, Davis RL, Lieu TL, Homer CJ, Helfand M. Universal newborn hearing screening: summary of evidence. *JAMA*. 2001;286:2000–2010
6. Kennedy CR, McCann D, Campbell MJ, et al. Language ability after early detection of permanent childhood hearing impairment. *N Engl J Med*. 2006;353:2131–2141
7. Schroeder L, Petrou S, McCann D, et al. The economic costs of congenital bilateral permanent childhood hearing impairment. *Pediatrics*. 2006;117:1101–1112
8. Wessex Universal Hearing Screening Trial Group. Controlled trial of universal neonatal screening for early identification of permanent childhood hearing impairment. *Lancet*. 1998;352:1957–1964
9. Watkin PM. Neonatal otoacoustic emission screening and the identification of deafness. *Arch Dis Child Fetal Neonatal Ed*. 1996;74:F16–F25
10. Watkin PM. Outcomes of neonatal screening for hearing loss by otoacoustic emission. *Arch Dis Child Fetal Neonatal Ed*. 1996;75:F158–F168
11. Tucker SM, Bhattacharya J. Screening of hearing impairment in the newborn using the auditory response cradle. *Arch Dis Child*. 1992;67:911–919
12. Kennedy C, McCann DM. Universal neonatal hearing screening moving from evidence to practice. *Arch Dis Child Fetal Neonatal Ed*. 2004;89:F378–F383
13. Kennedy C, McCann D, Campbell MJ, Kimm L, Thornton R. Eight year follow-up of a controlled trial of universal newborn screening for permanent childhood hearing impairment. *Lancet*. 2005;366:660–662
14. Baldwin M. *Reducing the Age of Audiological Certainty in Babies Identified by Neonatal Screening* [doctoral thesis]. Manchester, England: University of Manchester; 2005
15. Watkin PM, Hasan J, Baldwin M, Ahmed MAS. 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;3:175–185
16. Bishop DVM. *Test for Reception of Grammar*. Manchester, England: University of Manchester, Age and Cognitive Performance Research Centre; 1983
17. Dunn LM, Whetton C, Burley J. *British Picture Vocabulary Scale*. 2nd ed. Windsor, England: NFER-Nelson; 1997
18. Renfrew C. *Renfrew Bus Story Manual: A Test of Narrative Speech*. 3rd ed. Oxford, England: Renfrew/Winslow; 1995
19. Bishop D. Development of the Children's Communication Checklist (CCC): a method for assessing qualitative aspects of communicative impairment in children. *J Child Psychol Psychiatry*. 1998;39:879–891
20. Allen C, Nikopoulos TP, Dyer D, O'Donoghue GM. Reliability of a rating scale for measuring speech intelligibility after pediatric cochlear implantation. *Otol Neurotol*. 2001;22:631–633
21. Raven C, Raven JC, Court JH. *Manual for Raven's Progressive Matrices and Vocabulary Scales*. Oxford, England: Oxford Psychologists Press; 1998
22. Watkin PM, Baldwin M. Confirmation of deafness in infancy. *Arch Dis Child*. 1999;81:380–389
23. 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;1:1–176

Language Ability in Children With Permanent Hearing Impairment: The Influence of Early Management and Family Participation
Peter Watkin, Donna McCann, Catherine Law, Mark Mullee, Stavros Petrou, Jim Stevenson, Sarah Worsfold, Ho Ming Yuen and Colin Kennedy
Pediatrics 2007;120:e694-e701
DOI: 10.1542/peds.2006-2116

Updated Information & Services	including high-resolution figures, can be found at: http://www.pediatrics.org/cgi/content/full/120/3/e694
References	This article cites 17 articles, 8 of which you can access for free at: http://www.pediatrics.org/cgi/content/full/120/3/e694#BIBL
Subspecialty Collections	This article, along with others on similar topics, appears in the following collection(s): Dentistry & Otolaryngology http://www.pediatrics.org/cgi/collection/dentistry_and_otolaryngology
Permissions & Licensing	Information about reproducing this article in parts (figures, tables) or in its entirety can be found online at: http://www.pediatrics.org/misc/Permissions.shtml
Reprints	Information about ordering reprints can be found online: http://www.pediatrics.org/misc/reprints.shtml

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™

