

The effect of early confirmation of hearing loss on the behaviour in middle childhood of children with bilateral hearing impairment

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ABBREVIATIONS

PCHI Permanent childhood hearing impairment
SDQ Strengths and Difficulties Questionnaire
VABS Vineland adaptive behaviour scales

AIM To determine if the benefit of early confirmation of permanent childhood hearing impairment (PCHI) on children's receptive language development is associated with fewer behavioural problems.

METHOD Follow-up of a total population cohort of 120 children with PCHI of moderate or greater severity (≥ 40 decibels relative to hearing threshold level) (67 males, 53 females; mean age 7y 11mo, range 5y 5mo–11y 8mo) and 63 hearing children (37 males, 26 females; mean age 8y 1mo, range 6y 4mo–9y 10mo). The main outcome measures were the Strengths and Difficulties Questionnaire (SDQ) completed by teachers and parents and the Vineland Adaptive Behaviour Scales (VABS) which are completed on the basis of a parental interview.

RESULTS Children with PCHI had lower standard scores than hearing children on the Daily Living Skills ($p=0.001$) and the Socialisation ($p=0.001$) scales of the VABS. They had significantly higher Total Behaviour Problem scores on the parent-rated ($p=0.002$) and teacher-rated SDQ ($p=0.03$). Children for whom PCHI was confirmed by 9 months did not have significantly fewer problems on the behavioural measures than those confirmed after that age ($p=0.635$ and $p=0.196$).

INTERPRETATION Early confirmation has a beneficial effect on receptive language development but no significant impact in reducing behavioural problems in children with PCHI.

Children with hearing impairment frequently show behavioural problems.^{1–3} In terms of the type of behavioural difficulty associated with hearing impairment, difficulties related to attention have been specifically noted.⁴ For example, between one-third and one-half of children with hearing impairment have been reported as showing externalizing behavioural problems including difficulties with attention.⁵ Emotional problems are also a feature of children with hearing impairment.^{6,7}

It has been suggested that speech and signing were a mediator of the effect of hearing loss on behaviour.¹ The effect of hearing impairment on behaviour was less marked for children with better communication skills. These results parallel those from hearing children. There are well-replicated findings that, for a range of language related skills, poor development is linked with increased behavioural problems.⁸ As with hearing children, behavioural problems in young children with hearing impairment may be associated with a wide range of biological (e.g. preterm birth) and social (e.g. unresponsive parenting) factors.⁹ Moreover it is well established that children with physical disabilities and chronic illnesses have an elevated rate of behavioural and emotional difficulties.¹⁰

The factor of central interest in this paper is whether differences in the age at which permanent childhood hearing impairment (PCHI) is identified in children are related to the risk of behavioural problems. It has been suggested that enrolment in an intervention programme by the age of 9 months lessens the deficit of verbal ability compared with later intervention by as much as 19 points in the verbal ability quotient.^{11,12} We have shown previously, in the sample discussed in this paper, that behavioural problems are found more frequently in children with PCHI than in hearing children.² We have also shown, in the same sample, that early confirmation of PCHI is followed by improved language (particularly receptive language) abilities at primary school age.^{13,14} It would therefore be expected that early confirmation would decrease the rate of behavioural problems in children with PCHI because language ability mediated the impact of PCHI on behaviour. This paper tests this hypothesis in children with hearing loss taking part in a follow-up of a cohort study of the outcome of universal newborn screening for hearing impairment.

METHOD

Measures

The severity of hearing loss was categorized from recent audiological records as moderate (40–69dB hearing level), severe (70–94dB hearing level), or profound (≥ 95 dB hearing level) (Table I) according to four-frequency averaging of the pure-tone thresholds from 500 to 2000Hz (or, if pure-tone thresholds were unavailable, sound fields and electrophysiological test results).

The receptive language of the children was assessed by the Test of Reception of Grammar (TROG)¹⁵ and the British Picture Vocabulary Scale (BPVS).¹⁶ Severity of hearing loss was also measured and grouped as moderate (40–69dB relative to hearing threshold level [dB HTL]), severe (70–94dB HTL), and profound (≥ 95 dB HTL).

We have previously compared the standardized and age-adjusted scores obtained in this study on assessments of receptive and expressive language and of speech between the groups of children with and without hearing impairment.² For the present study, age-adjusted receptive language scores in the children with PCHI and the hearing children were used to derive *z*-scores for receptive language in children with PCHI, where *z* is the score, expressed in standard deviations of the distribution of scores in hearing children, relative to the mean score in hearing children. The use of *z*-scores made possible the derivation of a composite score for receptive language ($z_{\text{receptive language}} = (z_{\text{TROG}} + z_{\text{BPVS}}) / 2$).

The children's non-verbal abilities were assessed by their scores on Ravens Coloured Progressive Matrices (RCPM).¹⁷

Behavioural problems were measured using the Strengths and Difficulties Questionnaire (SDQ).¹⁸ This is a widely used behavioural screening questionnaire that provides data on

What this paper adds

- It is known that children with hearing loss are at risk of poor language and associated behavioural problems.
- We show that early confirmation of hearing loss by 9 months of age and subsequent aiding improves receptive language outcomes but does not decrease the number of behavioural problems compared with those confirmed later.
- We conclude that the better receptive language abilities associated with early confirmation of hearing loss is not sufficient to eradicate the increased risk of behavioural problems.

children and young people's behaviours, emotions, and relationships. It has been recommended as suitable for use with children with PCHI.¹⁹ Both teacher and parent versions of this questionnaire were used. A 'total difficulties' score reflecting behavioural problems in the child was derived from summing the scores of four SDQ scales (Emotional Symptoms, Conduct Problems, Hyperactivity, and Peer Problems) in the parent and teacher questionnaires.

The children's everyday functioning was assessed on the basis of parental interview using the Vineland Adaptive Behaviour Scale (VABS).²⁰ We report here on two of the scales: Daily Living Skills and Socialisation. In each case a standard score is used comparing children with age-matched norms.

Participants

Language and speech outcomes were assessed in children with bilateral PCHI of at least 40dB HTL born in eight districts of southern England. A comparison group of children with normal hearing born at the same hospitals and of comparable age at assessment was also evaluated. Children with PCHI attributable to a known postnatal cause (such as bacterial meningitis) were not included. The presence of other severe health problems that rendered the present study inappropriate (six

Table I: Child and family characteristics of hearing children and children with permanent childhood hearing impairment (PCHI) by age of confirmation

	Age at confirmation of PCHI				Hearing children	
	By 9mo (<i>n</i> =57)		≥ 9 mo (<i>n</i> =63)		<i>n</i> =63	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
Sex						
Male	34	60	33	52	37	39
Female	23	40	30	48	26	41
Degree of hearing loss						
Moderate	32	56	33	52	–	–
Severe	12	21	17	27	–	–
Profound	13	23	13	21	–	–
Multiple disabilities	9	16	7	11	0	0
Additional medical conditions	15	26	12	10	0	0
Age at assessment (y:mo)						
5:5–6:11	21	37	13	21	13	21
7:0–8:11	27	47	34	54	34	54
9:0–11:8	9	16	16	25	16	25
Mother's education						
No qualifications or <5 O levels ^a	22	39	21	34	25	40
≥ 5 O-levels or some A-levels ^a	30	53	22	53	25	40
University degree and above	5	9	9	4	13	21
English first language at home						
No	7	12	14	22	3	5

^aO-levels (now replaced in the UK by General Certificates of Secondary Education) are usually taken at age 16y; A-levels (now replaced by 'A2' examinations) are taken 2 years later as qualifications for entry to higher education.

children) was the only exclusion criterion. The study was approved by the South and West Multi-centre Research Ethics Committee and participating principal caregivers provided written informed consent.

Four of the districts (Southampton, Portsmouth, Swindon, and Bath) are in the Wessex region and provided a cohort of 54 000 births in four maternity units over a 3-year period from 1993 to 1996. This group also formed the sample for the Wessex Universal Neonatal Hearing Screening Trial Group, 1998.²¹ The other four districts are in the London region and provided a cohort of 103 000 births in those districts over a 5-year period from 1992 to 1997. In that period, two of these districts (Waltham Forest and Hillingdon) had, and their two neighbouring districts (Redbridge and Brent & Harrow) had not, operated a universal newborn screening programme. Thus, about half of the entire birth cohort of the present study had been in a target population for universal newborn screening for PCHI.

The children with PCHI were divided into those being confirmed by nine completed months of age (early confirmed) and those confirmed later (late confirmed). We prespecified the definition of early confirmation of permanent childhood hearing impairment as confirmation by none completed months of age. This was consistent with the definition in our previous trial of universal newborn screening²¹ and with the US Preventive Services Task Force benchmark for diagnosing or treating infants of nine completed months of age.²²

We have previously reported on measures and procedures used in more detail.^{13,14} We obtained, by review of the case records and outpatient lists, details of the detection and management of all cases of PCHI ascertained in the birth cohort described above. Of the early-confirmed children, nine subsequently received cochlear implants, as did seven of the late-confirmed children. Twenty-nine of the early-confirmed children received hearing aids by nine completed months of age. There were too few children in the groups to allow the joint analysis of the effects of early confirmation and of type of intervention on subsequent behaviour. Details of the interventions received by these children and their relation to language development have been summarized in an earlier paper.²³

Information on these cases was also obtained in this way from other paediatric audiologists and audiology scientists, current medical family practitioners, teachers, specialist teachers for the hearing impaired, speech and language therapists, and other professionals involved. Four researchers worked in two teams. A speech and language therapist proficient in British Sign Language assisted both teams and undertook assessments in those children that communicated with British Sign Language. For both the hearing children and children with PCHI, a pair of researchers arranged a time convenient for a visit to the child's home. During this visit, the principal caregiver, usually the mother, was interviewed and completed several questionnaires. The child was assessed, simultaneously and without knowledge of their early history, in a separate space. Questionnaires for completion by the child's teacher and, where relevant, teacher of the deaf were distributed and returned by mail.

Statistical analysis

First, the children with PCHI confirmed by nine completed months of age were compared with children with later-confirmed PCHI and with the normally hearing children for family background and child characteristics using χ^2 and one-way analysis of variance (ANOVA). For the ANOVA a preliminary test of homogeneity of variances was conducted. Next, we compared the behaviour of children with PCHI and hearing children using independent sample *t*-tests. Where the variances in the two groups being compared on the *t*-test were unequal, a correction to the degrees of freedom was incorporated which resulted in values that were not whole numbers.²⁴ There then followed a more extensive multivariate analysis of variance (MANOVA) analysis of the differences in behaviour of children with PCHI by age at confirmation. The homogeneity of covariances was tested for the MANOVA analyses. These MANOVA analyses were repeated using the following measures as covariates: sex, non-verbal ability (RCPM), severity of hearing loss, presence of multiple disabilities, additional medical conditions, mother's education, English as first language in home, and socioeconomic status. These were selected to reflect a range of possible influences on behaviour that might act as confounders. In no case were the findings altered by the use of covariates; therefore, for ease of presentation, just the main effects of age at confirmation are reported.

The relation between behaviour and receptive language score was examined further using a Loess curve. In this method, for each point in the data set a low-degree polynomial is fitted to a subset of the data, with explanatory variable values near the point whose response is being estimated. The polynomial is fitted using weighted least squares, giving more weight to points near the point whose response is being estimated and less weight to points further away.²⁵ It was used here to look for evidence of nonlinearity in the relation between the receptive language and behaviour in hearing children and in children with PCHI.

RESULTS

Characteristics of the samples

Of the 168 cases of children with PCHI identified, two were untraceable and six were not contacted because of severe active health problems. Twenty-five did not respond and 15 declined to participate. The principal caregivers of the remaining 120 of the 168 children cases identified gave consent for participation in the study. Of the 113 verbal participants, 46 of 53 children with early-confirmed PCHI and 57 of 60 late-confirmed PCHI subsequently completed assessments for oral language ability. Ninety-six of these receptive oral language assessments were in 97 children who used oral expressive language, with or without sign, and the other seven assessments were in 16 children whose only expressive language was sign. The other nine of these 16 users of sign as their only means of language expression completed assessments of receptive signing only (not reported here) and no assessment of oral receptive language.¹³

The background characteristics of the early- and late-confirmed groups and the hearing children are presented in

Table I. There were no clinically important differences between the early- and late-confirmed groups on sex, degree of hearing loss, number with multiple disabilities or additional medical conditions, age at assessment, mother's education, and English as the first language in the home. There were more families of children with PCHI where English was not the first language ($n=21$; 18%) compared with the families of hearing children; this was included as a covariate in the comparisons between these two groups. The mean age-adjusted total scores on the RCPM non-verbal test for early- and late-confirmed groups were not significantly different (early: mean=20.86, SD=6.26; late: mean=21.75, SD=6.12; $t=0.74$, $df=106$, $p=0.460$).

When the hearing children were compared with the children with PCHI there were no significant differences in sex, age at assessment, and mother's education. Consistent with previous research,²⁶ significant differences between the hearing children and the children with PCHI were found in the presence of multiple disabilities and additional medical conditions. The mean age-adjusted total score on the RCPM non-verbal test for the hearing children was significantly higher (mean=25.06, SD=5.20) than that of the children with PCHI (mean=21.35, SD=6.71; $t=4.01$, $df=169$, $p=0.001$). We have shown previously that the language scores of the children with PCHI remain significantly lower than those of hearing children when non-verbal ability is taken into account.¹³

Behaviour in children with PCHI and hearing children

The children with PCHI had significantly lower mean scores than the hearing children on both Daily Living Skills (mean difference=14.51, 95% CI 7.41–21.62; $t=4.93$, $df=179.46$, $p=0.001$) and the Socialisation (mean difference=9.30, 95% CI

4.12–14.12; $t=3.25$, $df=159.27$, $p=0.001$) standard scores on the VABS. Similarly on both the Parent rated (mean difference=2.88, 95% CI 1.27–4.49; $t=3.53$, $df=160.26$, $p=0.002$) and Teacher rated (mean difference=2.17, 95% CI 0.27–4.06; $t=2.67$, $df=172$, $p=0.03$) SDQ total scores, the children with PCHI showed significantly more problems. The mean total difficulties score for the children with PCHI was within the normal range for the SDQ.

These results confirm that children with PCHI show more marked behavioural problems than hearing children. We next consider whether within children with PCHI, age at confirmation of hearing impairment is related to differences in behaviour.

Behaviour in children with PCHI and age at confirmation

The children's scores on the Daily Living Skills and Socialisation scales of the VABS are presented in Table II. For the children with PCHI there were no significant differences between the age of confirmation groups on Daily Living Skills ($t=0.14$, $df=102.43$, $p=0.893$) or Socialisation ($t=0.17$, $df=104.93$, $p=0.865$).

On the Parent rated SDQ, a MANOVA of the four SDQ subscales showed there to be no difference between the early- and late-confirmation groups on the four subscales (multivariate $F=0.64$, Wilk's $\lambda=0.98$, $df=4,133$, $p=0.635$; test of homogeneity of covariance, Box's $M=12.63$, $p=0.275$; Table III). The effect of early confirmation remained non-significant in the MANCOVA with sex, non-verbal ability (RCPM), severity of hearing loss, presence of multiple disabilities, additional medical conditions, mother's education, English as first language in home, and social economic status as covariates (multivariate $F=0.14$, Wilk's $\lambda=0.99$, $df=4,94$, $p=0.968$). For the Teacher rated SDQ there was a consistent tendency for the late-confirmed children to have higher scores, but this was not significant (multivariate $F=1.54$, Wilk's $\lambda=0.95$, $df=4,108$, $p=0.196$; test of homogeneity of covariance, Box's $M=16.59$, $p=0.101$; Table IV). It remained non-significant when the above covariates were included (multivariate $F=1.69$, Wilk's $\lambda=0.93$, $df=4,90$, $p=0.158$).

The relation between receptive language scores and behaviour

We have previously shown that early confirmation is related to improved language ability in children with PCHI.¹³ We have also shown that the high level of behavioural problems in

Table II: Mean score on Vineland Adaptive Behaviour Scales for children with permanent childhood hearing impairment (PCHI) by age at confirmation and for hearing children

	n	Daily living skills		Socialisation	
		Mean	SD	Mean	SD
PCHI confirmation					
By 9mo	57	80.51	31.14	87.96	22.61
≥9mo	62	81.19	22.61	87.33	17.41
Hearing children	61	95.38	12.78	96.94	14.99

Table III: Mean score on Parent-rated Strengths and Difficulties Questionnaire for children with permanent childhood hearing impairment (PCHI) by age at confirmation and for hearing children

	n	Emotion		Conduct		Hyperactivity		Peer problems	
		Mean	SD	Mean	SD	Mean	SD	Mean	SD
PCHI confirmation									
By 9mo	55	1.75	1.83	1.78	1.72	4.67	3.01	1.84	1.78
≥9mo	63	2.00	2.07	1.87	1.84	4.32	2.98	1.59	1.77
Hearing children	63	1.54	1.69	0.95	1.14	3.57	2.63	0.95	1.56

Table IV: Mean score on Teacher-rated Strengths and Difficulties Questionnaire for children with permanent childhood hearing impairment (PCHI) by age at confirmation and for hearing children

	n	Emotion		Conduct		Hyperactivity		Peer problems	
		Mean	SD	Mean	SD	Mean	SD	Mean	SD
PCHI confirmation									
By 9mo	53	1.40	2.05	0.75	1.30	3.64	2.94	1.74	2.10
≥9mo	60	1.60	2.03	1.94	1.71	4.07	2.93	1.90	1.83
Hearing children	61	1.54	1.96	0.74	1.36	2.64	2.94	1.26	1.88

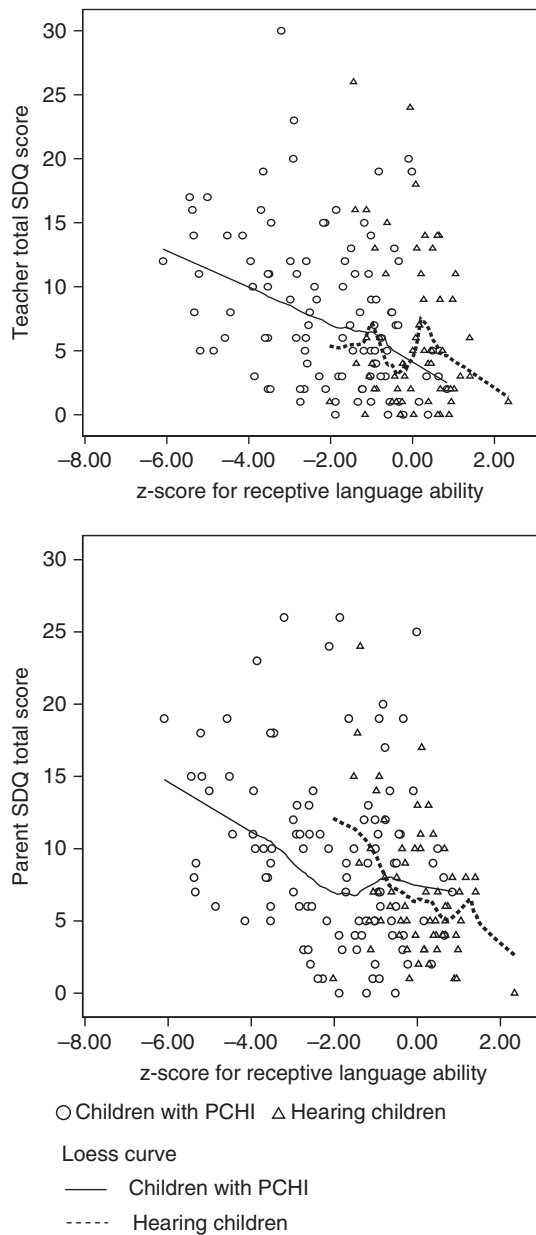


Figure 1: Plots of Teacher and of Parent total Strengths and Difficulties Questionnaire (SDQ) against receptive language score with added Loess curves for hearing children and children with permanent childhood hearing impairment (PCHI).

children with a hearing loss compared with hearing children is accounted for by their poorer communication ability.² However, the results above suggest that the improved language ability achieved by early confirmation is not sufficient to alter the risk of behavioural problems. Children with early-confirmed PCHI had receptive language scores over half a standard deviation higher than those with late-confirmed PCHI (-1.76 compared with -2.37). Nevertheless this leaves the early-confirmed group with a mean receptive language score still over 1.5 standard deviations below their hearing peers. This raises the question of what degree of normalization of receptive language scores would be needed to eradicate the risk of increased behavioural problems in children with PCHI compared with hearing children.

To examine this issue, Figure 1 plots the SDQ total scores (separately for Parent and Teacher rating) against receptive language score of children. The scores for children with PCHI and hearing children are plotted separately. The Loess curves are fitted separately for these two groups. The Loess curves provide a running average best fit polynomial through the data points. It shows both for children with PCHI and hearing children that lower behaviour scores are associated with higher receptive language scores across the full range. The number of behavioural problems continues to decrease across all levels of receptive language attainment. The Loess curves are very similar for children with PCHI and for hearing children. The plots for VABS scores against receptive language show a similar pattern. These findings suggest that normal levels of behavioural problems in children with PCHI could only be reached if their receptive language scores were also close to the distribution seen in hearing children. Early confirmation fails to bring the distribution of receptive language scores close to this point.

DISCUSSION

The present study provided a unique opportunity to examine the possible secondary benefits of early confirmation of hearing loss in a total cohort of hearing-impaired children. The study had the advantage that early confirmation arose in about half of the children in the context of a universal newborn hearing screening programme. For the sample as a whole there were no significant differences in the background characteristics of the early- and late-confirmed children. Therefore, early confirmation was not subject to bias in terms of features of the family, child, or local services determining the age of confirmation.

The main limitation on the study was the need to rely on questionnaires to assess the behavioural outcomes. Resources were not available to allow the use of extensive interview methods which would have had the benefit of providing a diagnostic psychiatric classification for the children at follow-up.

The findings confirm that children with PCHI have more marked behavioural problems than hearing children. We have shown previously that this elevated rate of behavioural problems is accounted for by the poorer language ability in these children.² Having also shown that a later age of confirmation of hearing loss was associated with impaired language ability,^{13,14} we went on here to test whether this may also result in a reduction in behavioural problems.

These findings provide additional support for previous demonstrations of the value of early confirmation in children with hearing impairment.^{11,12} The difference in the level of receptive language ability for those confirmed before 9 months and those confirmed later is approximately 0.6 standard deviations. However, this still leaves the receptive language ability of early-confirmed children on average over 1.5 standard deviations below their hearing peers.

We have previously reported that universal newborn screening for hearing impairment leads to early referral for permanent hearing loss.^{21,27} Although early confirmation of hearing

loss is of benefit to the child in terms of improving receptive language ability, the residual limitations on receptive language development are sufficiently marked that children with PCHI, even if early confirmed, will remain at risk of behavioural problems and less optimal day-by-day living skills compared with hearing children.

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