

# Newborn Hearing Screening vs Later Hearing Screening and Developmental Outcomes in Children With Permanent Childhood Hearing Impairment

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**P**ERMANENT CHILDHOOD HEARING impairment is a serious, relatively common condition.<sup>1-3</sup> Auditory input is essential for development and social functioning, so early awareness of a child's hearing ability is important in creating opportunities for early amplification and habilitation when necessary.

Until some years ago, distraction hearing screening (behavioral testing) was used for hearing screening around the age of 9 months. Newborn hearing screening (within 2 weeks of birth) was introduced in many developed countries because it was thought that the earlier permanent childhood hearing impairment was diagnosed, the less developmentally disadvantaged children would become.<sup>4,5</sup>

However, to date no strong evidence exists for universal implementation of newborn hearing screening. Studies on developmental effects of newborn hearing screening have been descriptive in nature and based on convenience samples.<sup>6-12</sup> Conducting a randomized trial, apart from presenting practical difficulties (including expense and duration), was considered

**Context** Newborn hearing screening programs have been implemented in many countries because it was thought that the earlier permanent childhood hearing impairment is detected, the less developmentally disadvantaged children would become. To date, however, no strong evidence exists for universal introduction of newborn hearing screening.

**Objective** To study the effect of newborn hearing screening vs distraction hearing screening, conducted at 9 months of age, on development, spoken communication, and quality of life.

**Design, Setting, and Participants** Between 2002 and 2006, all 65 regions in the Netherlands replaced distraction hearing screening with newborn hearing screening. Consequently, the type of hearing screening offered was based on availability at the place and date of birth and was independent of developmental prognoses of individual children. All children born in the Netherlands between 2003 and 2005 were included. At the age of 3 to 5 years, all children with permanent childhood hearing impairment were identified. Evaluation ended December 2009.

**Main Outcome Measures** Performance (education and spoken and signed communication), development (general and language), and quality of life.

**Results** During the study period, 335 560 children were born in a newborn hearing screening region and 234 826 children in a distraction hearing screening region. At follow-up, 263 children in newborn hearing screening regions (0.78 per 1000 children) and 171 children in distraction hearing screening regions (0.73 per 1000 children) had been diagnosed with permanent childhood hearing impairment. Three hundred one children (69.4%) participated in analysis of general performance measures. There was no difference between groups in the primary mode of communication or type of education. Analysis of extensive developmental outcomes included 80 children born in newborn hearing screening regions and 70 in distraction hearing screening regions. Multivariate analysis of variance showed that overall, children in newborn hearing screening regions had higher developmental outcome scores compared with children in distraction hearing screening regions (Wilks  $\lambda = 0.79$ ;  $F_{12} = 2.705$ ;  $P = .003$ ). For social development, the mean between-group difference in quotient points was 8.8 (95% CI, 0.8 to 16.7) and for gross motor development, 9.1 (95% CI, 1.1 to 17.1). For quality of life, the mean between-group difference was 5.3 (95% CI, 1.7 to 8.9), also in favor of children in newborn hearing screening regions.

**Conclusion** Compared with distraction hearing screening, a newborn hearing screening program was associated with better developmental outcomes at age 3 to 5 years among children with permanent childhood hearing impairment.

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ethically infeasible.<sup>9,11</sup> As an alternative, we used the regional differences in allocation of hearing screening type created by national policy in the Netherlands. Newborn hearing screening was gradually introduced by region in

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all 65 regions beginning in 2002 and totally replaced distraction hearing screening by June 2006. This policy meant that the type of hearing screening offered to children was based solely on location and date of birth. Because hearing screening type was independent of the prognosis of the individual child, the region of birth can be considered an instrumental variable.<sup>13-16</sup>

Using the regional differences in hearing screening, we studied the developmental effects of newborn hearing screening compared with distraction hearing screening in 3- to 5-year-old children with permanent childhood hearing impairment. We hypothesized that newborn hearing screening would be associated with better general development and improved spoken communication and quality of life compared with distraction hearing screening.

## METHODS

### Study Population

The DECIBEL (Developmental Evaluation of Children: Impacts and Benefits of Early hearing screening, Leiden) study included children born in the Netherlands between January 1, 2003, and December 31, 2005. With the assistance of 2 researchers (A.M.H.K. and S.K.), professionals at every audiology center in the Netherlands (n=22) identified all children born in 2003 through 2005 with permanent hearing impairment at age 3 to 5 years. The audiology center is the designated and only organization for diagnostic evaluation and amplification for children with permanent hearing impairment.

Permanent childhood hearing impairment was defined as bilateral permanent conductive or sensorineural hearing loss of 40 dB or greater in the better ear and was classified on the basis of the most recent hearing test (measured unaided and computed using 500, 1000, and 2000 Hz). Hearing loss was categorized as moderate (40-60 dB), severe (61-90 dB), or profound (>90 dB). Because the identification of children at audiology centers was performed cross-sectionally, independent of screening type, the identification was

unbiased with respect to type or result of hearing screening and type or degree of hearing impairment.

Excluded were children who had been in neonatal intensive care units, because they are not a target population for universal hearing screening; children who acquired their hearing impairment after birth; and those who were severely cognitively and physically disabled. Children already participating in other research projects at 1 center or from a center that joined the study at a late stage and children whose parents were not competent in the Dutch language were also excluded from participation.

### Hearing Screening Programs and Study Design

The place and date of birth of the child determined the type of hearing screening offered: distraction hearing screening or newborn hearing screening (eFigure, available at <http://www.jama.com>). The 2 programs differed in the age at screening and the method used; both followed uniform protocols. Distraction hearing screening, offered at the age of 9 months, is a 3-stage hearing screening using sounds to provoke a behavioral reaction. If a child does not react as expected at the first screening, a repeat screening is planned, and this is repeated once more if necessary. A third-stage result positive for hearing impairment is followed by referral to an audiology center for diagnostic investigation and confirmation of hearing impairment. The distraction hearing screening test is subjective and has been shown to be unreliable in children with cognitive and physical handicaps.<sup>17</sup>

Newborn hearing screening for well infants, offered before the age of 2 weeks, is also a 3-stage screening program, but it uses transient evoked oto-acoustic emissions for the first 2 stages and automated auditory brainstem responses in the third stage. A unilateral or bilateral positive result for hearing impairment is followed by repeat screening, and a positive result in the third stage is followed by referral to an audiology center. Screening is performed either

during a home visit, together with newborn blood spot screening, or at a well baby visit.

We verified whether the hearing screening the child was offered on the basis of the regional hearing screening program at the time of birth corresponded with the hearing screening the child had actually received, as reported by parents and noted in the audiology records. Parents were invited to participate in the study by mail. Medical records of all children identified were reviewed for available information on characteristics and performance measures. Characteristics included maternal education level (representing socioeconomic status), parental hearing status, type and result of hearing screening, age at start of amplification (hearing aid, bone-anchored hearing aid, or cochlear implant), degree of hearing impairment, and etiology (if available).

After parents provided written informed consent, they were sent (by mail or e-mail) 3 standardized instruments measuring developmental outcome and a questionnaire to complete the characteristics and performance measures. Parents were asked for their country of birth and the race/ethnicity of their child to determine any cultural differences between the groups. Evaluation ended December 2009.

This study was approved by the medical ethics committee of the Leiden University Medical Center. The privacy committee of the neonatal intensive care unit hearing screening program gave permission for anonymous verification of its patients.

### Assessment of Development

Performance measures included the primary mode of communication (oral language only or oral and sign language) and the type of education (regular education, education for hearing impaired children, or education for children with developmental disabilities) as reported by parents or audiology records. General and language developmental outcomes were measured using the Child Development Inventory, expressive language development using

the MacArthur Communicative Development Inventory, and quality of life using the Pediatric Quality of Life Inventory 4.0, all completed by parents.

The Child Development Inventory is a standardized instrument designed to assess the development of children from age 15 months to 6 years and is often used in research on this topic.<sup>18</sup> The 1992 version of the Child Development Inventory was translated into Dutch according to rules formulated by Guillemin et al<sup>19</sup> and was also adjusted for use in children whose primary language is sign language. Parents completed the questionnaire by indicating which of the listed 270 behavioral items they observed in their child. The items are grouped to form scales, including social development, motor development, and expressive language and language comprehension (combined in a total language scale). The general development score is a summary score that provides an overall index of development by including 10 of the most age-discriminating items from each of the scales. The scores were recalculated by the use of the original norm data into developmental ages, and these generated developmental quotients when divided by chronological age and multiplied by 100. Higher scores indicate better development. A developmental quotient of 80 or more represents normal development. A quotient between 70 and 80 is regarded as borderline development. A difference of 4 points is generally interpreted as clinically relevant.<sup>18</sup>

The short-form version of the MacArthur Communicative Development Inventory (hereafter referred to as MacArthur) was used to assess 3 aspects of expressive language. Active vocabulary (number of words spoken and signed; maximum score, 100), sentence complexity (ranging from 1 for least complex to 3 for most complex for 9 sentences; maximum score, 27), and mean length of 3 longest utterances (number of words) were evaluated.<sup>20</sup> With regard to active vocabulary, parents were asked to indicate which words of the child's originally only spoken vo-

cabulary inventory were currently spoken, signed, or both. In children not speaking in sentences, sentence complexity and mean length of longest utterance were not applicable and were classified as missing. Crude scores were used in the analysis, eliminating any ceiling or floor effects caused by including children who were chronologically or developmentally older than the population for whom the scale was originally designed.

The Pediatric Quality of Life Inventory<sup>21,22</sup> questionnaire encompasses both physical functioning and psychosocial functioning. Each item is scored on a 5-point Likert scale. To create scale scores, the mean crude score was computed as the sum of the items divided by the number of items answered (which corrects for missing items). The total quality-of-life score is the sum of the mean crude score on all scales. Higher scores indicate better quality of life (maximum score, 100). Two age-specific versions were used: for children aged 2 to 4 years and those aged 5 to 7 years, both with comparable constructs and scoring.

### Statistical Analysis

First, the proportion of children with permanent childhood hearing impairment in both hearing screening programs was compared, along with baseline characteristics. A main analysis comparing performance measures and developmental outcome was performed according to the type of hearing screening children were offered (determined by location and date of birth). We also investigated whether children who agreed to participate in extensive developmental outcome measures were comparable with those not participating. For those participating in the extensive outcome study, variables believed to affect the outcome in children with permanent childhood hearing impairment (maternal education, degree of hearing impairment, parental hearing status, mode of communication, and age at start of amplification) were compared between groups. An independent-samples *t* test

was used for continuous variables and  $\chi^2$  test for categorical variables.

Because of multiple testing with correlated subscales, a multivariate analysis of variance (MANOVA) was used to determine the overall difference in outcome between groups. If the MANOVA was significant, linear regression was used to compare developmental outcome per subscale. The assumptions for this type of analysis were tested and met (independence, normality, and homogeneity of variance of the residuals). Adjustment for residual confounding was done for maternal education and chronological age at developmental evaluation (when applicable). The difference in chronological age at developmental evaluation between the 2 groups was considered a consequence of the gradual introduction of newborn hearing screening, with more children in newborn hearing screening being younger.

It is known that hearing impairment in children with congenital cytomegalovirus infection may be progressive over time and not yet detectable by newborn hearing screening. The presence of this infection can be confirmed prior to hearing screening when appropriate methods are used.<sup>23</sup> For these reasons, a sensitivity analysis was performed excluding the children with known congenital cytomegalovirus infection.

An analysis based on the type of hearing screening children actually received was also performed. Differences between this sensitivity analysis and the main analysis are reported. The significance level was set at  $P < .05$  and 2-sided testing performed. All statistical tests were carried out using SPSS version 17.0 (SPSS Inc, Chicago, Illinois).

### RESULTS

In 2003-2005, the number of children born in the Netherlands was 582 214. Of these, 11 828 children (2%) were admitted to a neonatal intensive care unit and therefore excluded. Of all other live newborns, 335 560 children were born in a region where newborn hearing screening was offered and 234 826 in

a region where distraction hearing screening was offered. At follow-up, 263 children in a newborn hearing screening region had been diagnosed with permanent childhood hearing impairment (0.78 per 1000 children) and 171 children in a distraction hearing screen-

ing region (0.73 per 1000 children) (difference, 0.05 per 1000; 95% confidence interval [CI], -0.12 to 0.09) (TABLE 1). Of these 434 children, 133 children were excluded: 17 had an acquired hearing impairment, 53 were severely cognitively and physically dis-

abled, 52 were participating in other research projects, and 11 had parents who were not competent in the Dutch language (FIGURE and Table 1). The remaining 301 children (69.4%) were included in the study on general performance measures. Of these, 150 children (49.8%) agreed to participate in extensive investigations on developmental outcome (Figure). The main reason for refusal to participate was an already extensive schedule of medical evaluations.

In the analysis of general performance measures, the 2 groups (newborn hearing screening, n=183; distraction hearing screening, n=118) were comparable in degree of hearing impairment, primary mode of communication, and type of education (TABLE 2). Children included in analysis of extensive developmental outcomes (n=150) were comparable with those not participating (n=151) in degree of hearing impairment, sex, and type of amplification (eTable 1). In the analysis of developmental outcome measures, the 2 groups were comparable in all baseline characteristics (newborn hearing screening, n=80; distraction hearing screening, n=70). Compared with children in distraction hearing screening, children in newborn hearing screening were screened at a younger age, their hearing amplified 13 months earlier, and their development evaluated 13 months earlier (47.9 mo vs 60.7 mo) (TABLE 3).

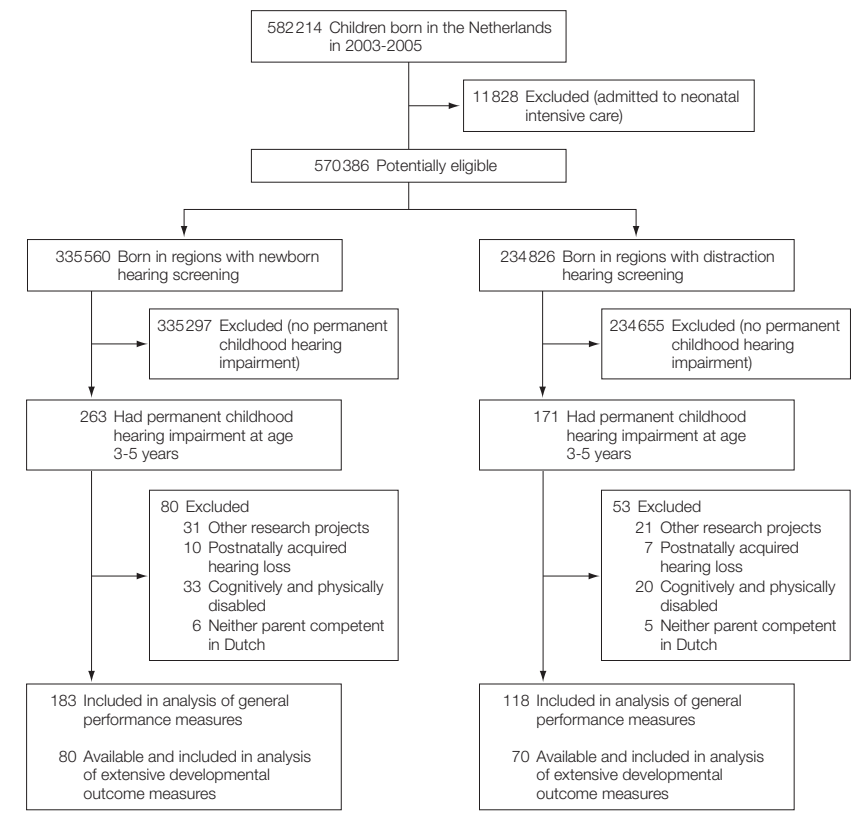
Multivariate analysis of variance (both crude and adjusted for maternal education) showed that children in newborn hearing screening regions had higher developmental outcome scores overall compared with children in distraction hearing screening regions (Wilks  $\lambda$ =0.79;  $F_{12}$ =2.705;  $P$ =.003). On the Child Development Inventory, after adjustment for maternal education, we found a statistically significant difference in social development and gross motor development and a nonsignificant difference in the same direction on all other subscales (TABLE 4). Additional adjustment for the degree of hearing impairment, race/ethnicity, and

**Table 1.** Number of Children Allocated to Distraction or Newborn Hearing Screening Programs and the Prevalence of Permanent Childhood Hearing Impairment

	Birth Year			Overall
	2003	2004	2005	
<b>Distraction hearing screening</b>				
Children screened, No.	152 900	76 200	5526	234 626
Children identified with permanent hearing impairment, No.	118	48	5	171
Prevalence of hearing impairment per 1000 children screened <sup>a</sup>	0.77	0.63	0.90	0.73
<b>Newborn hearing screening</b>				
Children screened, No.	42 166	114 374	179 020	335 560
Children identified with permanent hearing impairment, No.	32	94	137	263
Prevalence of hearing impairment per 1000 children screened <sup>a</sup>	0.76	0.82	0.77	0.78

<sup>a</sup>Calculated by dividing the number of children identified with permanent hearing impairment by the number of children screened.

**Figure.** DECIBEL Study Flow Diagram





parental hearing status did not result in materially different results.

Using the MacArthur, children in the newborn hearing screening group were found to use statistically significantly fewer signed words compared with children in the distraction hearing screening group (mean difference, -11.2; 95% CI, -20.6 to -1.9) when adjustment was made for maternal education and age at developmental evaluation. In children in the newborn hearing screening region, a larger (but not statistically significantly different) spoken vocabulary was found (mean difference, 8.7; 95% CI, -3.9 to 21.2). The number of spoken words was inversely associated with the number of signed words, resulting in opposite results, in favor of children in newborn hearing screening. The level of sentence complexity and the mean length of longest spoken utterance were comparable in both groups after adjustment.

Quality of life was statistically significantly higher in children in the newborn hearing screening group on all scales, except for the mean crude score on the emotion scale. Adjustment for maternal education did not influence the results (Table 4).

In a sensitivity analysis, 10 children with congenital cytomegalovirus infection were excluded (5 in each group). In addition to social development, gross motor development, and quality of life, the mean differences on language developmental outcome measures (expressive language, total language, and spoken vocabulary) increased, although they remained not statistically significant, in favor of children who had newborn hearing screening (eTable 2).

Verification of the type of hearing screening showed that 12 children underwent no hearing screening, 9 children underwent direct diagnostic evaluation, 10 children were screened by newborn hearing screening but were in the main analysis in distraction hearing screening, and 1 child was screened by distraction hearing screening instead of newborn hearing screening. This resulted in 129 children who underwent 1 of the 2 types of hearing screening (sen-

**Table 2.** General Performance Measures in Children With Permanent Childhood Hearing Impairment at Age 3 to 5 Years<sup>a</sup>

Characteristic	No. (%)		P Value
	Newborn Hearing Screening (n = 183)	Distraction Hearing Screening (n = 118)	
Degree of hearing loss	(n = 179)	(n = 118)	
Moderate (40-60 dB)	88 (49.2)	58 (49.2)	.90
Severe (61-90 dB)	49 (27.4)	30 (25.4)	
Profound (>90 dB)	42 (23.5)	30 (21.7)	
Primary mode of communication	(n = 97)	(n = 80)	
Oral language only	28 (28.9)	26 (32.5)	.60
Oral and sign language	69 (71.1)	54 (67.5)	
Education type in children aged >48 mo	(n = 71)	(n = 89)	
Regular education	18 (25.4)	23 (25.8)	.13
Regular education with counseling for hearing impaired	1 (1.4)	5 (5.6)	
Education for children with hearing impairment	49 (69.0)	61 (68.5)	
Education for children with developmental disabilities	3 (4.2)	0	

<sup>a</sup>Categorical variables were compared between groups with  $\chi^2$  test.

**Table 3.** Characteristics at Birth and at Extensive Developmental Evaluation: Main Analysis of Children With Permanent Childhood Hearing Impairment

Characteristic	Newborn Hearing Screening (n = 80)	Distraction Hearing Screening (n = 70)	P Value
At baseline			
Male sex, No. (%)	47 (58.8)	40 (57.1)	.84
White, No. (%)	65 (81.3)	61 (87.4)	.23
Mother with higher education, No. (%) <sup>a</sup>	35 (44.3)	33 (49.3)	.82
Both parents normal hearing, No. (%)	66 (83.5)	60 (87.0)	.74
Referral at hearing screening, No. (%)	65 (81.3)	41 (58.6) <sup>b</sup>	.15
Degree of hearing loss at first evaluation, No. (%)			
Lower than moderate (<40 dB)	9 (11.3)	7 (10.0)	.57
Moderate (40-60 dB)	44 (55.0)	32 (45.7)	
Severe (61-90 dB)	13 (16.3)	17 (24.3)	
Profound (>90 dB)	9 (11.3)	14 (20.0)	
At age 3-5 years			
Age at amplification, mean (SD), mo	15.7 (14.0)	29.2 (14.8)	<.001
Type of amplification, No. (%)			
Hearing aid	57 (71.3)	53 (79.7)	.52
Bone-anchored hearing aid	5 (6.3)	2 (2.9)	
Cochlear implant	18 (22.5)	14 (20.0)	
Hearing aid previously but not currently	0	1 (1.4)	
Mode of communication, No. (%)			
Oral language only	27 (35.1)	28 (41.2)	.42
Oral and sign language	53 (66.3)	42 (60.0)	
Additional handicaps, No. (%)	5 (6.3)	8 (12.9)	.17
Age at development evaluation, mean (SD), mo <sup>c</sup>	47.9 (9.9)	60.7 (6.8)	<.001
Age at language and QOL evaluation, mean (SD), mo <sup>d</sup>	45.9 (9.6)	59.1 (7.2)	<.001

Abbreviation: QOL, quality of life.

<sup>a</sup>Mother's education level is used to represent socioeconomic status. "Higher" education indicates that the mother either completed 6 years of secondary education, completed university, or completed another higher education institution.

<sup>b</sup>In the distraction hearing screening group, no result was available for 12 children because they had not been screened.

<sup>c</sup>Refers to chronological age of child when evaluated with Child Development Inventory.

<sup>d</sup>Refers to chronological age of child when evaluated with Pediatric Quality of Life Inventory and MacArthur Communicative Development Inventory.

sitivity analysis based on screening actually received: 85 children in newborn hearing screening and 45 in distraction hearing screening). The results in the sensitivity analysis were largely comparable with the results found in the main analysis (eTable 3).

## COMMENT

We found that newborn hearing screening, compared with distraction hearing screening, was associated with statistically significantly fewer words signed and better overall, social, and gross motor development and quality of life at 3 to 5 years of age among children with permanent hearing impairment.

This study was not a randomized trial. Due to national policy, all regions replaced distraction hearing screening by newborn hearing screening some time during the period of study, which meant that allocation of newborns to one or the other type of hearing screening was based solely on location and date of birth and not on prognosis of developmental outcome. Therefore, we were able to study the effect of regional differences in hearing screening type on developmental outcome, rather than the effect of hearing screening offered to the individual child, which might be based on a specific reason.<sup>16</sup>

Limitations of the study should be considered. First, response bias is a con-

cern because it is unknown whether parents of children with abnormal development were more eager to participate. However, it is not likely that this bias affected children unequally in the 2 hearing screening groups. Second, possible identification bias is important to address. Because allotment to 1 of the 2 hearing screening regions was established earlier (when the child was born), and was independent of the prognoses of hearing and development for individual children, we believe no bias was introduced during identification procedures at the audiology centers. It is not impossible that after closure of data collection, very late-onset hearing loss was missed, especially in children born in later years of the study. However, the proportion of children with permanent hearing impairment of congenital cause who were unidentified and presented later than school age is most likely small. The extensive network of youth health care organizations monitoring development probably would have detected and referred such children. There is also no reason to believe that an identification bias affected the 2 hearing screening groups unequally.

Third, the informant perspective (parental reporting) could have caused information bias. Again, it is not likely that such bias affected children unequally in the 2 hearing screening groups. Fourth, it is important to acknowledge the difference in age at developmental evaluation between the 2 screening groups. Because age-referenced norm data and age-specific questionnaire constructs were used and (when applicable) adjustment for age was made in the analysis, it is not likely that bias caused by differences in age at developmental assessment influenced the results. Selection bias, possibly introduced by 1 center that excluded some children already participating in research, was ruled out in a subset analysis excluding this center. The results were unchanged. Fifth, as a result of the relatively small sample size, we were only able to detect relatively large differences between the 2 groups. Nevertheless, we did find differences be-

**Table 4.** Developmental Outcome in Newborn Hearing Screening and Distraction Hearing Screening: Main Analysis of Children With Permanent Childhood Hearing Impairment

	No. of Assessed Cases	Mean (SD)		Adjusted Between-Group Difference (95% CI) <sup>b</sup>
		Newborn Hearing Screening (n = 80) <sup>a</sup>	Distraction Hearing Screening (n = 70) <sup>a</sup>	
Child Development Inventory quotient <sup>c</sup>		(n = 70)	(n = 60)	
General development	130	81.4 (17.2)	79.3 (16.3)	3.0 (−2.5 to 8.6)
Self help	130	87.1 (24.3)	81.7 (20.7)	5.9 (−1.8 to 13.6)
Fine motor development	130	89.2 (20.0)	85.4 (15.9)	4.6 (−1.3 to 10.4)
Gross motor development	130	86.1 (24.3)	77.6 (21.0)	9.1 (1.1 to 17.1) <sup>d</sup>
Social development	130	79.9 (25.1)	71.5 (22.0)	8.8 (0.8 to 16.7) <sup>d</sup>
Expressive language	130	82.4 (25.7)	76.0 (25.0)	7.2 (−1.3 to 15.8)
Language comprehension	130	75.4 (19.9)	72.7 (19.2)	3.6 (−2.8 to 10.1)
Total language	130	78.9 (21.9)	74.4 (20.6)	5.4 (−1.6 to 12.5)
MacArthur crude score <sup>e</sup>				
Total words spoken	136	54.0 (34.3)	66.0 (32.5)	8.7 (−3.9 to 21.2)
Total words signed	136	11.8 (17.7)	18.9 (25.1)	−11.2 (−20.6 to −1.9) <sup>d</sup>
Spoken sentence structure	126	16.5 (7.9)	20.5 (6.9)	0.6 (−2.4 to 3.7)
Mean length of longest utterance	115	5.4 (2.4)	6.7 (3.1)	0.3 (−1.5 to 0.9)
Pediatric Quality of Life Inventory score <sup>f</sup>		(n = 77)	(n = 66)	
Total	143	85.8 (8.9)	80.5 (12.0)	5.3 (1.7 to 8.9) <sup>d</sup>
Physical	143	91.5 (10.7)	86.2 (17.4)	5.4 (0.6 to 10.2) <sup>d</sup>
Emotional	143	76.2 (15.3)	72.2 (14.8)	3.6 (−1.5 to 8.7)
Social	143	85.6 (15.1)	77.7 (15.4)	8.1 (2.9 to 13.3) <sup>d</sup>
Psychosocial	143	82.2 (11.4)	77.1 (11.4)	5.2 (1.3 to 9.0) <sup>d</sup>

Abbreviations: CI, confidence interval; DHS, distraction hearing screening; NHS, newborn hearing screening.

<sup>a</sup>Multivariate analysis of variance showed that children in NHS regions had higher scores overall on developmental outcomes compared with children in DHS regions (Wilks  $\lambda=0.79$ ;  $F_{12}=2.705$ ;  $P=.003$ ).

<sup>b</sup>Differences in Child Development Inventory and Pediatric Quality of Life Inventory scores were adjusted for maternal education; differences in MacArthur score were adjusted for maternal education and age at developmental evaluation.

<sup>c</sup>A developmental quotient  $\geq 80$  represents normal development. A quotient between 70 and 80 is regarded as borderline development.

<sup>d</sup> $P < .05$ .

<sup>e</sup>The category "words signed" was added to the original MacArthur Communicative Development Inventory with permission from the Dutch authors. For active vocabulary (number of words spoken and signed), the maximum score was 100; for spoken sentence structure (ranging from 1 for least complex to 3 for most complex for 9 sentences), the maximum score was 27; and the mean length of 3 longest utterances was measured in number of words. The numbers of children assessed for each MacArthur subscore were as follows: total words spoken and total words signed, 74 NHS, 62 DHS; spoken sentence structure, 68 NHS, 58 DHS; and mean length of longest utterance, 62 NHS, 53 DHS.

<sup>f</sup>Higher scores indicate better quality of life (maximum, 100).

tween the 2 hearing screening groups in many outcome domains that are considered clinically relevant and important. A number of differences in developmental outcomes did not reach statistical significance but did meet the generally accepted level for clinical significance. A larger sample would be necessary to examine more subtle differences.

Our results confirm those of previous studies, most of which were performed in convenience samples, that report that newborn hearing screening leads to advantages in language developmental outcome for children with permanent hearing impairment, when compared with children with no screening or only targeted screening of high-risk infants.<sup>7,9,24-27</sup> Our results on the Pediatric Quality of Life Inventory replicate earlier findings by Moeller<sup>24</sup> that children identified with permanent hearing impairment later are at risk in areas such as behavior, emotion, and quality of life. Moreover, in our study, better outcomes following early hearing screening were demonstrated in the strongest design possible to date, with an instrumental variable facilitating the study.<sup>16</sup>

We found statistically significant differences in overall development, total words signed, social development, gross motor development, and quality of life. Children in newborn hearing screening regions used statistically significantly fewer signed words than children in distraction hearing screening regions. Although not statistically significant, the 8.7-point difference in spoken words could be considered clinically significant. In the hearing world, a larger spoken vocabulary can assist effective communication strategies. It may well be that more effective communication strategies caused by better language development increased social development and thereby quality of life in children who received newborn hearing screening. Quality-of-life outcomes reflect parental awareness, the effect of possible false-negative screening results, and the effect of permanent childhood hearing impairment on daily life. Quality of life is therefore an important outcome mea-

sure reflecting both positive and potentially negative effects of screening programs. It is not clear how motor development is affected by hearing screening programs.

In a previous study, we found that the presence of congenital cytomegalovirus infection influences developmental outcome.<sup>23</sup> Special attention should be paid to the habilitation of these children. Further insight into the developmental consequences of the various causes of hearing impairment will only be achieved if future studies take etiology into account.

It is unlikely that large developmental differences could occur simply by identifying hearing impairment early. Improved outcomes are to be expected only when early identification is followed by early intervention.<sup>9</sup> The Joint Committee on Infant Hearing recommends that intervention (amplification, family support and communication, language and auditory development support) following positive results for hearing impairment and confirmation of permanent childhood hearing impairment should start no later than age 6 months.<sup>4</sup> In our study, however, this recommendation was not always achieved. This was due (at least in part) to the fact that newborn hearing screening was still in its implementation phase. If anything, the delay between identification and amplification might have resulted in a reduction of the developmental differences between newborn and distraction hearing screening groups in this study.

Finally, it is important to realize that despite early hearing screening, the development of children with permanent childhood hearing impairment at age 3 to 5 years following newborn hearing screening is still not comparable with that of normally developing children with normal hearing. Their mean language comprehension is within the borderline range.

The results of the DECIBEL study add evidence to the presumed importance and effectiveness of the implementation of universal newborn hearing screening programs. Because this

study was performed nationwide, among all children born in the Netherlands in 3 subsequent years, we believe our results can be generalized to other countries with universal hearing screening programs, but the feasibility and effectiveness of newborn hearing screening programs in other countries remain to be studied.

## CONCLUSION

We found that a newborn hearing screening program, compared with distraction hearing screening, was associated with better developmental outcomes at age 3 to 5 years among children with permanent childhood hearing impairment.

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**Online-Only Material:** The eFigure and eTables 1 through 3 are available at <http://www.jama.com>.

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