

Universal Newborn Hearing Screening

Summary of Evidence

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EACH YEAR APPROXIMATELY 5000 infants are born in the United States with moderate-to-profound, bilateral permanent hearing loss (PHL). Estimates of the prevalence of moderate, severe, and profound congenital PHL among newborns range from 1 in 900 to 1 in 2500.¹⁻⁸ Moderate, severe, and profound congenital PHL is associated with delayed language, learning, and speech development.⁹⁻¹³ This delay is measurable before age 3 years^{14,15} and has consequences throughout life. On average, deaf students graduate from high school with language and academic achievement levels below those of fourth-grade students with normal hearing.^{16,17}

Diagnosis and treatment are often delayed until ages 1 or 2 years in children with congenital PHL, particularly among those at low risk for PHL.¹⁸⁻²² Current theory holds that auditory stimuli during the first 6 months of life are critical to the development of speech and language skills.²³⁻²⁵ Advocates of universal newborn hearing screening (UNHS) believe that earlier application of available therapies, such as speech and language therapy, amplification, and family support, could reduce or eliminate the gap in language skills between deaf and hearing children.^{26,27} Screening also identifies infants who have mild and unilateral hearing impairment, but the consequences of delay are not well established

Context Each year, approximately 5000 infants are born in the United States with moderate-to-profound, bilateral permanent hearing loss (PHL). Universal newborn hearing screening (UNHS) has been proposed as a means to speed diagnosis and treatment and thereby improve language outcomes in these children.

Objectives To identify strengths, weaknesses, and gaps in the evidence supporting UNHS and to compare the additional benefits and harms of UNHS with those of selective screening of high-risk newborns.

Data Sources We searched the MEDLINE, CINAHL, and PsychINFO databases for relevant articles published from 1994 to August 2001, using terms for hearing disorders, infants or newborns, screening, and relevant treatments. We contacted experts and reviewed reference lists to identify additional articles, including those published before 1994.

Study Selection We included controlled and observational studies of (1) the accuracy, yield, and harms of screening using otoacoustic emissions (OAEs), auditory brainstem response (ABR), or both in the general newborn population and (2) the effects of screening or early identification and treatment on language outcomes. Of an original 340 articles identified, 19 articles, including 1 controlled trial, met these inclusion criteria.

Data Extraction Data on population, test performance, outcomes, and methodological quality were extracted by 2 authors (D.C.T., H.M.) using prespecified criteria developed by the US Preventive Services Task Force. We queried authors when information needed to assess study quality was missing.

Data Synthesis Good-quality studies show that from 2041 to 2794 low-risk and 86 to 208 high-risk newborns were screened to find 1 case of moderate-to-profound PHL. The best estimate of positive predictive value was 6.7%. Six percent to 15% of infants who are missed by the screening tests are subsequently diagnosed with bilateral PHL. In a trial of UNHS vs clinical screening at age 8 months, UNHS increased the proportion of infants with moderate-to-severe hearing loss diagnosed by age 10 months (57% vs 14%) but did not reduce the rate of diagnosis after age 18 months. No good-quality controlled study has compared UNHS with selective screening of high-risk newborns. In fair- to poor-quality cohort studies, intervention before age 6 months was associated with improved language and communication skills by ages 2 to 5 years. These studies had unclear criteria for selecting subjects, and none compared an inception cohort of low-risk newborns identified by screening with those identified in usual care, making it impossible to exclude selection bias as an explanation for the results. In a mathematical model based on the literature review, we estimated that extending screening to low-risk infants would detect 1 additional case before age 10 months for every 1441 low-risk infants screened, and result in treatment before 10 months of 1 additional case for every 2401 low-risk infants screened. With UNHS, 254 newborns would be referred for audiological evaluation because of false-positive second-stage screening test results vs 48 for selective screening.

Conclusions Modern screening tests for hearing impairment can improve identification of newborns with PHL, but the efficacy of UNHS to improve long-term language outcomes remains uncertain.

JAMA. 2001;286:2000-2010

www.jama.com

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in these infants, and most are not candidates for hearing aids and other therapies associated with early identification.^{7,20,28,29}

Selective screening of high-risk newborns is an alternative to UNHS. Among infants in a neonatal intensive care unit (NICU), the risk of moderate-to-severe PHL is 10 to 20 times higher than in the general population.³⁰ In addition to NICU admission, the Joint Committee on Infant Hearing high risk guidelines specify 4 other risk factors (BOX).^{31,32} From 10% to 30% of newborns meet these criteria, which can identify 50% to 75% of all cases of moderate-to-profound bilateral hearing loss.²

In 1995 the US Preventive Services Task Force (USPSTF) found insufficient evidence to recommend UNHS,³³ based on the low prevalence of PHL and the risk for substantial misclassification. While the evidence for the efficacy of early intervention for patients diagnosed by screening was incomplete, the Task Force endorsed selective screening of high-risk newborns because of their higher prevalence of PHL.

Since 1995, many health care professionals and federal health care agencies have advocated for UNHS, which is now mandated by law in 32 states.^{18,31,34} Is this widespread support for UNHS now justified? To update the USPSTF recommendations, we critically reviewed recent evidence to identify strengths, weaknesses, and gaps in the evidence supporting UNHS.

METHODS

We focused our literature search on key questions underlying the clinical logic of screening for hearing impairment in newborns (FIGURE). The logic assumes that screening tests are accurate; that screening reduces delays in diagnosis and treatment; that earlier treatment results in better language function within the preschool period; and that improvement in early language function will improve educational, occupational, and social function later in life. We reviewed the literature about each assumption, and used the results to construct a math-

ematical model of the benefits and harms of UNHS.

Search Strategy

We searched MEDLINE, CINAHL, and PsychINFO for relevant articles published in English from 1994 to September 2000, using the keywords *hearing disorders* and *infant or newborn* combined with terms for screening and relevant treatments, such as *early intervention*, *amplification*, and *American Sign Language*. The search was updated quarterly through August 2001. We also examined reference lists of review articles^{7,35-42} and queried experts. To identify articles published before 1994, we relied on systematic reviews published in 1996 and 1997.^{20,33}

Study Selection, Data Abstraction, Validity Assessment, and Synthesis

Two authors reviewed titles and abstracts of 864 articles from the original searches and selected 340 articles as possibly relevant to 1 of the key questions. We then selected the following to include in evidence tables: (1) controlled trials; (2) reports on the accuracy, yield, or harms of screening using otoacoustic emissions (OAEs) or auditory brainstem response (ABR); or (3) reports of the effects of screening, or of early identification and treatment, on language outcomes. Ten studies of the yield of uni-

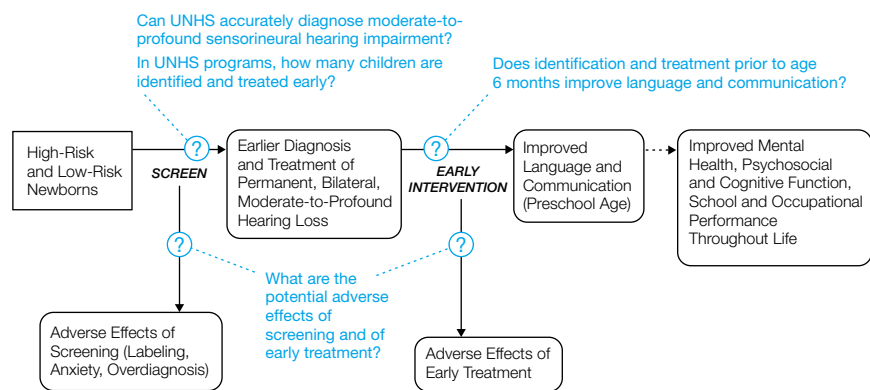
versal screening programs,^{26,43-51} 1 study of the accuracy of OAEs and ABR in high-risk infants,⁵² and 8 studies of language outcomes^{14,15,53-58} met these inclusion criteria. An additional 110 articles provided supplemental data about the included screening studies, results of selective screening programs, and other background information.

Two authors abstracted data for population, test performance, outcomes, and methodological quality from each included study. We classified each study as “good,” “fair,” or “poor” using prespecified criteria developed by

Box. Risk Factors for Sensorineural Hearing Loss in Newborns

1. Neonatal intensive care unit admission for 2 or more days
2. Usher syndrome, Waardenburg syndrome, or findings associated with other syndromes known to include hearing loss
3. Family history of hereditary childhood sensorineural hearing loss
4. Congenital infections such as toxoplasmosis, bacterial meningitis, syphilis, rubella, cytomegalovirus, and herpes
5. Craniofacial anomalies, including morphologic abnormalities of the pinna and ear canal

Figure. Newborn Hearing Screening Analytic Framework



Early intervention indicates hearing aids or other amplification, American Sign Language and/or English instruction, speech and language therapy, and family education and support. UNHS indicates universal newborn hearing screening.

the USPSTF for grading the internal validity of studies.⁵⁹ When necessary, we sought from authors additional information needed to apply the criteria. The entire 13-member Task Force discussed the review, examined and rated the quality of 4 key studies of early intervention,^{54,58,56,57} and provided overall guidance.

We constructed a mathematical model of the likely benefits and harms of UNHS vs selective screening of a hypothetical cohort of 10 000 newborns,

estimating prevalence, sensitivity and specificity, compliance, treatment effect size, and other model parameters from the included studies. Excel 97 (Microsoft Corp, Redmond, Wash) was used for all analyses.

RESULTS

Can UNHS Accurately Diagnose Moderate-to-Profound Sensorineural Hearing Impairment?

Ten publications, including 1 controlled trial, 4 state-based programs, and

5 hospital-based programs, provided information about the yield of UNHS in actual screening programs (TABLE 1). In the 2 good-quality studies, screening detected 1 case of bilateral, moderate-to-profound PHL for every 925 to 1422 newborns screened. The yield was 2041 to 2794 low-risk and 86 to 208 high-risk newborns screened to find 1 case.^{29,43,44,61} Fair-quality and poor-quality studies had higher yields due to inclusion of infants who had mild, unilateral, or unconfirmed hearing loss.

Table 1. Studies of Universal Newborn Hearing Screening*

Study	Description (Quality Rating)	Screening Tests	No. Screened/ No. Available (% Screened)	Yield (NNS) to Identify 1 Case of Bilateral PHL	Positive Screen, No. (%)
Wessex UNHS Trial Group, ⁴³ 1998	Controlled, nonrandomized trial at 4 hospitals from 10/93 to 10/96 (good)	TEOAE followed by ABR	21 279/25 609 (83)	23/21 279 (925)	342 (1.6)
Prieve et al, ⁶⁰ 2000	Statewide demonstration project at 7 perinatal centers, 8 hospitals in New York (good)	TEOAE followed by TEOAE or ABR at birth admission; TEOAE, ABR at 4-6 wk (stage 2)	69 766/71 922 (97)	49/69 736 (1422)	4699 (6.5) (stage 1)
Vohr et al, ²⁶ 1998	Cohort from 8 maternity hospitals in Rhode Island from 1/93 to 12/96 (fair)	TEOAE followed by ABR (high-risk infants); TEOAE and ABR in 2-6 wk (low-risk)	52 659/53 121 (99)	79/52 659 (666)‡	5397 (10.2) (stage 1); 677 (1.3) (stage 2)
Finitzo et al, ⁴⁵ 1998	Cohort from 9 Texas hospitals from 1/94 to 6/97 (fair)	ABR or TEOAE at birth admission followed by either ABR or TEOAE at 1-8 wk	52 508/54 228 (97)	20/17 105 (855)‡	1787 (3.4)
Barsky-Firsker and Sun, ⁴⁷ 1997	Hospital-based series at St Barnabas Medical Center, New Jersey, from 1/93 to 12/95 (fair)	ABR by audiologists (stage 1)	15 749/16 229 (97)	NR	485 (3.1)
Watkin, ⁴⁸ 1996	Hospital-based series at Whipps Cross Hospital, England (fair)	TEOAE followed by TEOAE and ABR within 4 wk	11 606/14 353 (81)	19/11 606 (755)	337 (2.9)
Mehl and Thompson, ⁴⁶ 1998	Cohort from 26 Colorado hospitals from 1992 to 1996 (poor)	19 ABR, 1 TEOAE, 6 ABR (follow-up screen not reported)	41 796 (NR)	NR	2709 (6.5)
Aidan et al, ⁴⁹ 1999	Hospital-based series in Paris, France, of infants in normal newborn nursery (poor)	TEOAE in 48 h; TEOAE within 4 wk	1421/1727 (82)	2/1421 (711)	238 (16.7)
Clemens et al, ⁵⁰ 2000	Hospital-based series at Women's Hospital of Greensboro, NC, from 7/98 to 6/99 (poor)	ABR followed by retest for failure (stage 1a) or ABR (stage 1b); outpatient ABR and diagnostic ABR (stage 2)	5010/5034 (99.5)	NR	103/5054
Mason and Herrmann, ⁵¹ 1998	Series of infants born at Kaiser, Honolulu, Hawaii, from 3/92 to 2/97 (poor)	ABR	10 372/10 773 (96)	12/10 372 (864)	415 (4.0)

*NNS indicates number needed to screen; PHL, permanent hearing loss; UNHS, universal newborn hearing screening; TEOAE, transient evoked otoacoustic emissions; ABR, automated auditory brainstem response; NR, not reported; NIH, National Institutes of Health; NICU, neonatal intensive care unit; and FH, family history.

†Reported different rates for misses and fails.

‡Includes mild, bilateral hearing loss.

§Includes unilateral hearing loss.

||Data reported for 1996 only.

Most programs in Table 1 used a 2-stage screening protocol, in which an infant who fails the initial test (OAE or ABR) is retested in the hospital or as an outpatient within 12 weeks of discharge, and is referred for audiological evaluation if he or she fails the second test. Among infants with positive screening tests, the likelihood that the infant has hearing loss (ie, the positive predictive value [PPV] of the screening test) varied by study. In 1 good-quality study, the overall PPV for the second-stage

screening test was 6.7%.⁴³ In the well-baby nursery, the PPV was 2.2%, meaning that 1 of every 45 infants referred for outpatient audiological evaluation eventually proved to have moderate-to-profound bilateral PHL. None of the screening programs measured the sensitivity and specificity of neonatal screening against an independent gold standard, although 3 reported the percentage of cases (6%-15%) missed by screening but eventually diagnosed by other means.^{26,28,43}

A behavioral test is the appropriate gold standard determination for permanent hearing impairment,⁶² but cannot be performed reliably before 8 to 9 months of age.^{63,64} One large, good-quality study of test performance measured the sensitivity and specificity of OAE and ABR using an independent gold standard, visual reinforcement audiometry, performed at ages 8 to 12 months.⁵² The study found that these screening tests are not sensitive enough to rule out significant hearing loss. The sensitivity of OAE ranged from 80% for moderate hearing loss to 98% for profound hearing loss. For ABR, sensitivity and specificity were 84% and 90%, respectively. The 2-stage protocol missed 11% of affected ears. Overall, neonatal testing resulted in a final diagnosis of bilateral moderate-to-profound PHL among 1 in 230 high-risk and 1 in 2348 low-risk infants.

These estimates of accuracy and yield are lower than expected, but are probably more reliable than those from actual screening programs (Table 1). In those programs, decisions about diagnosis and treatment are made on the basis of a diagnostic ABR performed when the infant is 1 to 6 months old. The use of this intermediate diagnostic standard facilitates earlier intervention, but may overestimate the number of cases of PHL. In the Wessex trial, the first audiological examination was done when the infants were between ages 8 and 12 weeks. Of 158 infants who screened positive, 27 were diagnosed as having PHL; in 2 of these cases (7.4%), however, the diagnosis was wrong, and the infants had normal hearing when reexamined at 4 months or 10 months of age.⁶¹ In another study, 5 (29%) of 17 infants initially diagnosed to have moderate PHL were later found to have only mild hearing loss.²⁸ No other studies listed in Table 1 described follow-up procedures to determine how often the intermediate diagnosis of PHL was incorrect.

In UNHS Programs, How Many Children Are Identified and Treated Early?

The most important indicators of the benefit of UNHS are the number of ad-

No. Followed Up (% Lost to Follow-up)	Definition of High Risk	No. Low-Risk Identified/No. Screened (NNS)	No. High-Risk Identified/No. Screened (NNS)
NR (NR)	NIH criteria	7/19 555 (2794)	20/1724 (86)
† (43.4)	NICU infants	33/NR (2041)‡	52/NR (208)‡
4575 (15.2)	NICU infants	61/47 529 (779)§	50/5130 (103)§
1224 (31.5)	NR	NR	NR
NR (NR)	NICU infants	29/14 014 (483)	23/1735 (75)
290 (14)	Risk factors and/or NICU infants	7 (NR)	13 (NR)
1296 (52.2)	NR	NR	NR
123 (48.3)	Hypoxemia, hyperbilirubinemia, FH	2/1421 (711)	NR
85 (17.5)	NICU infants	NR	4/454 (114)
362 (12.8)	NICU infants	5/8971 (1794)	7/1401 (200)

ditional cases of significant hearing impairment that are diagnosed and treated early.^{18,29} In a British trial comparing UNHS with *no* newborn screening, UNHS increased rates of referral to an audiologist by age 6 months for infants with moderate-to-profound PHL (an increase of 51 per 100 000 infants; 95% confidence interval, 7.4-94.0 per 100 000; $P=.03$), but not confirmation of diagnosis ($P=.22$) or initiation of management within 10 months ($P=.08$).⁴³ Among those infants with moderate or severe hearing loss, however, screening led to highly significant increases in confirmation and management by age 10 months. With UNHS, 13 of 23 (57%) children with moderate or severe impairment were diagnosed by 10 months, compared with 2 of 13 (14%) without UNHS.⁴³ Use of UNHS did not reduce the rate of diagnosis after 18 months, either overall (5/27 for UNHS vs 6/26 for the control group) or in the moderate-to-severe subgroup.

In the best-quality US study,²⁹ ages of diagnosis for mild-to-moderate hearing losses were 3.5 months (mean) and 2.5 months (median), and those for severe PHL were 6.3 months (mean) and 3.8 months (median). In about 40% of cases, the diagnosis of PHL was delayed until ages 1 to 2 years because of infant illness, developmental delays, noncompliant parents,²⁹ or transient conductive hearing losses.^{28,52}

How much of the overall effect of UNHS can be attributed to screening low-risk infants? Uncertainty about the proficiency of selective screening makes it difficult to determine how much UNHS can add. The most frequently cited studies of selective screening were performed over 10 years ago,⁶⁵⁻⁶⁸ and they did not report the number of infants diagnosed before ages 6 or 10 months. In recent good-quality and fair-quality studies of UNHS, 19% to 42% of infants with PHL had no risk factors.^{29,43,51,67} Only 1 study reported results in sufficient detail to calculate how often low-risk infants with bilateral moderate-to-profound PHL are diagnosed early by UNHS. For every 7692

low-risk infants screened, 1 additional case of PHL was diagnosed before age 10 months.

Does Identification and Treatment Prior to Age 6 Months Improve Language and Communication?

No prospective, controlled study directly examined whether newborn hearing screening results in improved speech, language, or educational development. None of the state-based programs described in Table 1 reported the outcomes of treatment for infants identified as having hearing impairment.

TABLE 2 summarizes methodological aspects and results of 8 recent cohort studies from 3 intervention programs that compared early- and late-identified children with impaired hearing. All of these studies used standardized receptive and expressive tests to evaluate speech and language skills in preschool children,^{69,70} and all reported statistically significant associations between the age at the time of diagnosis and language development at ages 2 to 5 years.

Six of these 8 studies reported speech and language results for children enrolled in the Colorado Home Intervention Program (CHIP).^{14,15,53-56,71,72} One of these compared language performance of hearing-impaired children born in hospitals with UNHS programs to that for children born in hospitals with no UNHS program (Table 2).⁵⁵ It found that mean scores for expressive, receptive, and total language were within normal ranges for the screened group and 18 to 21 points higher ($P<.001$) than for the unscreened group. A 20-point gap is more than 1 SD lower than normal for age, which would indicate that a child with average intellect would have the language abilities of a child who had an IQ of 80. Children identified as having hearing impairment prior to age 6 months (whether in the screened or unscreened group) had a smaller gap between language development and cognitive ability than children identified after 6 months. Language development was within the normal range for

56% of the screened group compared with 24% of the unscreened group.

While this study used relevant, validated measures of language outcomes and controlled for several important potential confounders, study design issues limited the conclusions that could be drawn. Eligibility for the screened group was determined by the availability of an assessment of language outcomes at ages 2 to 4 years. Because the groups were drawn from different hospitals and time periods, factors other than exposure to UNHS might have influenced outcomes. Selection of subjects and assessment of outcome were unblinded, and neither the number of excluded subjects, nor the reasons for exclusion, are reported.

The most widely cited CHIP study compared 72 hearing-impaired children identified prior to age 6 months with 78 hearing-impaired children identified after 6 months.⁵⁴ After adjustment for cognitive function, children identified before age 6 months had language scores at or near their cognitive test scores, whereas children identified after age 6 months performed, on average, 20 points lower on language scores than on cognitive scores. Children with low cognitive abilities (cognitive quotient <80) experienced a smaller improvement in total language, but no statistically significant improvement in receptive and expressive language abilities.

This study had limitations as well. Late-identified children were more likely to be cognitively impaired, to have severe or worse hearing loss, to use sign language, and to have mothers with lower educational achievement. The statistical method used in the analysis did not simultaneously adjust for more than 2 factors and may not have removed the influence of these differences. Additionally, the study did not provide data on dropout rates in the 2 groups, and outcome assessments were not masked.

All of the studies in Table 2 had several important limitations. The study populations were composed of convenience samples. None of the studies had clear criteria for inclusion, none had

Table 2. Cohort Studies Reporting Language Outcomes*

Study (Quality)	Selection of Subjects	Comparability and Maintenance of Early vs Late Groups	Adjustment for Confounders	Results
Studies From Colorado Home Intervention Program (CHIP)				
Apuzzo and Yoshinaga-Itano, ⁵³ 1995 (fair)	Convenience sample of 69 high-risk infants diagnosed between ages 2 and 25 mo. Those with severe cognitive delay were excluded.	Late-identified group was more likely to have severe-to-profound hearing loss (65% vs 50%). No report of attrition or follow-up rates.	One-way ANOVA did not adjust for SES, family involvement, or other potential confounders.	At age 40 mo, infants identified before age 2 mo had higher mean MCDI scores for expressive language ($P < .01$).
Yoshinaga-Itano and Apuzzo, ¹⁴ 1998 (poor)	Convenience sample of 40 high-risk infants, divided into those identified and treated before age 6 mo ($n = 15$) and those treated after age 18 mo ($n = 25$). Those with severe cognitive delay were excluded ($DQ < 60$).	Late-identified group was more likely to have severe-to-profound hearing loss (52% vs 47%). No report of attrition or follow-up rates.	Child's sex, severity of hearing loss, cognitive function, and other disabilities examined in 2-way ANCOVAs, not multiple regression (no simultaneous adjustment for multiple confounders).	At age 40 mo, infants identified before age 6 mo had better adjusted mean MCDI scores for expressive language (81.1 vs 64.3, $P < .05$) and receptive language (84.4 vs 70.1, $P < .05$).
Yoshinaga-Itano and Apuzzo, ¹⁵ 1998 (poor)	Convenience sample of 82 infants, ages 19 to 36 mo, with mild-to-profound PHL, divided into those identified before age 6 mo ($n = 34$) and between ages 7 and 18 mo ($n = 48$). Early group identified by high-risk registry; late group by usual care. Those with severe cognitive delay were excluded ($DQ < 60$).	Late-identified group was more likely to have severe-to-profound hearing loss (77% vs 42%). No report of attrition or follow-up rates.	Child's sex, severity of hearing loss, cognitive function, and other disabilities examined in 2-way ANCOVAs, not multiple regression (no simultaneous adjustment for multiple confounders).	At age 26 mo, infants identified before age 6 mo had better adjusted mean MCDI scores for expressive language (76.2 vs 56.6, $P = .001$), receptive language (82.1 vs 58.3, $P = .002$), MacArthur CDI adjusted mean receptive vocabulary (200 vs 86.4, $P < .001$), and expressive vocabulary (117 vs 54, $P < .03$).
Yoshinaga-Itano et al, ⁵⁴ 1998 (poor)	Convenience sample of 150 children ages 13 to 36 mo with mild-to-profound PHL, divided into those identified before ($n = 72$) or after ($n = 78$) age 6 mo. The number of low-risk infants and the role of UNHS in identifying subjects are not described. Selection bias is likely because the design probably excluded infants who were diagnosed to have hearing loss but did not enter the program, or who entered, but were lost to follow-up.	At baseline, compared groups differed in some demographic characteristics and in the proportion of subjects with cognitive impairment and severe-to-profound hearing loss (CQ < 80 , 29% early group vs 56% late group; severe-to-profound hearing loss, 34% early group vs 46% late group). No report of attrition or follow-up rates.	There was stratification by CQ (< 80 vs ≥ 80). Other covariates (sex, minority status, maternal education level, Medicaid status, severity, mode of communication, other disabilities) were examined singly in 2-way ANCOVAs.	At ages 13 to 36 mo, adjusted mean MCDI receptive LQ was higher for those identified before age 6 mo (79.6 vs 64.6, $P < .001$). Mean MCDI expressive LQ was higher (78.3 vs 63.1, $P < .001$) and total language (79 vs 64, $P < .001$) was higher in early identified group. No differences in LQ among 4 age-of-identification levels in late-identified group.
Yoshinaga-Itano et al, ⁵⁵ 2000 (poor)	Children born in a hospital with a UNHS program in effect at time of birth ($n = 25$) were compared with children born in a hospital without a UNHS program ($n = 25$). All subjects had been enrolled in the CHIP program. Eligibility for the screened group was determined by the availability of an assessment of language outcomes. The creation of the study groups and description of the patients limited the conclusions that could be drawn.	The exposure was birth at a hospital with a UNHS program, not age of identification. Because the groups were drawn from different hospitals and time periods, factors other than exposure to UNHS might have influenced outcomes. Selection of subjects and assessment of outcome were unblinded, and neither the number of excluded subjects nor the reasons for exclusion are reported.	Pairs matched on age of testing (9-59 mo), degree of hearing loss (mild, moderate, moderately severe, profound), and CQ.	Mean (SE) scores for expressive, receptive, and total language were within normal range for the screened group and 18 to 21 points higher ($P < .001$) than the unscreened group (expressive language, 82.9 [3.7] vs 62.1 [4.3]; receptive language, 81.5 [3.7] vs 66.8 [4.0]; total language, 82.2 [3.3] vs 64.4 [3.9]). Language development was within normal range for 56% of the screened group compared with 24% of the unscreened group.
Mayne et al, ⁵⁶ 2000 (poor)	Convenience sample of 113 children ages 24 to 73 mo, divided into those diagnosed before and after age 6 mo. The number of low-risk infants and the role of UNHS in identifying subjects are not described. Overlap of sample with previous CHIP studies was not reported.	Demographic comparisons of the groups were not reported. No report of attrition or follow-up rates.	Regression analysis adjusted for degree of hearing loss, mode of communication, other disabilities, parents' hearing, CQ, mother's education, ethnicity, SES.	At ages 24 to 36 mo, age at diagnosis explained 23% of the variance in expressive language scores.

(continued)

Table 2. Cohort Studies Reporting Language Outcomes (cont)*

Study (Quality)	Selection of Subjects	Comparability and Maintenance of Early vs Late Groups	Adjustment for Confounders	Results
Studies From Other Programs				
Moeller, ⁵⁷ 2000 (fair)	Convenience sample of 5-year-olds (n = 112) who completed the Diagnostic Early Intervention Program in Lincoln, Neb. Children with nonverbal IQ <70 and those who did not participate in program through age 5 y were excluded. The number of low-risk infants and the role of UNHS in identifying subjects are not described. Outcome assessments were made preintervention and postintervention.	Not reported. No report of attrition or follow-up rates. Early identified children may have more opportunity to drop out, although differential dropout may be less of a problem at 5 y than in studies assessing closer to enrollment.	Multiple regression analysis adjusted for family involvement, degree of hearing loss, and nonverbal IQ.	At age 5 y, family involvement accounted for 57% of variance in vocabulary and age of enrollment accounted for 11.5%. Adjusted mean vocabulary and reasoning scores were within normal range among children enrolled prior to age 11 mo but were lower for later-identified children.
Calderon and Naidu, ⁵⁸ 2000 (fair)	Cohort of 80 children with profound hearing loss enrolled in ECHI in Seattle, Wash. Children with developmental delay were excluded. Cohort grouped by 3 levels by age of entry into program: <12 mo (n = 9), 12-24 mo (n = 39), >24 mo (n = 32). The method of sampling is not described, but the design excluded patients who entered the program but did not graduate.	Not reported. Late-diagnosed group had less severe-to-profound loss (36% vs 66%). Overall loss to follow-up not reported. Because the early diagnosed group was in the program longer, they had more opportunity to drop out, so a differential loss to follow-up is likely.	Controlled for degree of hearing loss, degree of outcome impairment that was present upon entry into program (baseline test levels).	At age 3 y, age at entry to program explained 43.5% of the variance in receptive language and 49% of the variance in expressive language. Children treated before age 2 y had better outcomes than those treated after age 2 y. Only 3 children entered the program prior to age 6 mo.

*ANOVA indicates analysis of variance; SES, socioeconomic status; MCDI, Minnesota Child Development Inventory; DQ, developmental quotient; PHL, permanent hearing loss; ANCOVA, analysis of covariance; CDI, child development inventory; UNHS, universal newborn hearing screening; CQ, cognitive quotient; LQ, language quotient; and ECHI, Early Child Hearing Intervention.

blinded assessments, and all selected children for inclusion based on the availability of a language assessment between ages 2 to 5 years. This could introduce bias, because early identified children who remained in the program may have had better results than early identified children who were not available for follow-up. None of the studies provide information on attrition or follow-up rates. All but 1 compared children who were identified early and late by means other than UNHS, rather than children whose age at identification and enrollment was determined primarily by whether or not they were screened. Other factors, such as family involvement (an important contributor to language development),⁵⁷ the degree of other disability, or the quality of pediatric care, might have influenced the time of identification and the language outcome. The task force rated the strength of evidence linking early treatment with improved language func-

tion “inconclusive” and the quality of evidence as “fair/poor.”

What Are the Potential Adverse Effects of Screening and of Early Treatment?

Screening. Potential adverse effects of false-positive screening tests include misdiagnosis, parental misunderstanding and anxiety, and unfavorable labeling. As noted earlier, the intermediate diagnostic standard determining PHL is imperfect; in expert hands, as many as 7% of infants diagnosed as having permanent PHL may eventually prove to have normal hearing. The frequency of misdiagnosis in everyday practice settings has not been studied.

In the only controlled trial,⁴³ parents whose infants were screened had anxiety and attitudes similar to parents in the unscreened group. Three other small studies found false-positive screening results produced significant or lasting anxiety in 3.5% to 14% of parents.^{50,73,74} Ad-

ditionally, 8% of mothers said they treated their child differently (eg, spoke louder or clapped their hands).⁵⁰ No study attempted to assess the effect of parental anxiety or changes in parental behavior on infants' development or on the parent-infant relationship.

Treatment. The harms of early intervention have not been adequately studied, and differing ethical and philosophical attitudes about deaf awareness and culture have led to controversy about the content of early interventions.³³ Treatment strategies for hearing loss in children include hearing aids or other amplification, American Sign Language and/or English instruction, speech and language therapy, and family education and support. Different experts advocate substantially different approaches based on competing theories of language acquisition and communication. Treatment strategies vary widely, even among programs in states or hospitals with established screening programs.⁷⁵ This

variation reflects uncertainty about the efficacy of the interventions, which have not been evaluated in randomized trials or in population-based cohort studies.^{13,20,33}

The argument for early intervention is based on the prevailing theory of language development, which holds that early auditory input is an important precursor of language development. An opposing viewpoint suggests that, during infancy, nonverbal communication, joint attention, shared experiences, and mutual understanding are more important precursors of language development than are hearing speech and forming sounds. Proponents of this view theorize that early intervention could harm infants because it leads parents to focus on “means of communication the child has the least prerequisites for” and on the child’s disability instead of his or her competencies.⁷⁶ Because there are no randomized trials of different management strategies, it is impossible to assess the merits of these concerns.

Summary of Benefits and Harms

TABLE 3 summarizes the benefits and harms of UNHS and selective screening in a hypothetical cohort of 10 000 newborns. We used our literature review to estimate prevalence, sensitivity and specificity, compliance, and the likelihood of being diagnosed and treated before age 10 months. There are no reliable data to estimate how often selective screening misses patients whose risk factors were not detected during hospitalization. We assumed that in a selective screening program, 20% of high-risk infants are never tested in the hospital, vs 10% for UNHS. There are also no reliable data by which to estimate the probability that a low-risk infant will be diagnosed by 10 months without newborn screening; we estimate this to be 35% in our base case.

With UNHS, an additional 7800 screening tests would be done, resulting in the diagnosis of 6 additional cases of moderate-to-profound hearing loss diagnosed before age 10 months. Of these, 3 additional cases would be treated before age 10 months. Thus, the

number needed to screen (NNS) to detect 1 additional case before age 10 months would be 1441 and the NNS to treat 1 additional case before 10 months would be 2401. With UNHS, 254 newborns would be referred for audiological evaluation because of false-positive second-stage screening test results (vs 48 for selective screening), and 1 of these would also be falsely diagnosed to have PHL at the first posthospital visit to an audiologist.

Of the 6 additional early diagnosed, low-risk newborns, how many would actually benefit from early treatment? The data needed to estimate this—the probabilities of a poor language out-

come with and without early treatment—are not known. To use a hypothetical example, if 50% of newborns with PHL would have poor language ability if diagnosed after age 10 months, and early intervention reduced this by 50%, then the NNS to prevent 1 additional case of delayed language acquisition would be 6771.

COMMENT

TABLE 4 summarizes the evidence for each of the major assumptions underlying the case for UNHS. Several gaps in information about UNHS effectiveness remain. Modern screening tests for hearing impairment can improve identifica-

Table 3. Benefits of Screening a Hypothetical Cohort of 10 000 Newborns for Moderate-to-Profound PHL*

Benefit and Relevant Factors	Probability or Effect Size	UNHS	High-Risk Screening
Assumptions, %†			
Proportion at high risk	0.2		
Prevalence			
High-risk group	0.008		
Low-risk group	0.0008		
Miss rate (proportion not screened in hospital) for UNHS			
High-risk group	0.1		
Low-risk group	0.05		
Follow-up rate for misses	0.9		
Miss rate for high-risk screening			
High-risk group	0.2		
Follow-up rate for misses	0.75		
Sensitivity of 2-stage screening	0.85		
Specificity of 2-stage screening	0.97		
Compliance with follow-up	0.9		
Accuracy of diagnostic ABR			
Sensitivity	1.0		
Specificity	0.995		
Proportion of low-risk infants diagnosed before 10 mo without screening	0.35		
Treated before 1 y	0.6		
Results, No.			
Infants screened		9400	1600
Cases diagnosed before 10 mo		17	12
Cases treated before 10 mo		10	7
Out of total		22	22
False-positive screening tests		254	48
Normal infants incorrectly diagnosed to have PHL at first posthospital audiological examination		1	0
NNS to diagnose 1 case		584	173
NNS to diagnose 1 additional case before 10 mo		1441	...
NNS to treat 1 additional case before 10 mo		2401	...

*PHL indicates permanent hearing loss; UNHS, universal newborn hearing screening; ABR, automated auditory brainstem response; NNS, number needed to screen; and ellipses, not applicable.

†Base case assumptions are derived from the studies in Table 2, except for miss rate for high-risk screening (high-risk group), accuracy of diagnostic ABR (sensitivity), and proportion of low-risk infants diagnosed without screening.

Table 4. Strength of Evidence for Universal Newborn Hearing Screening*

Key Question	Evidence	Evidence Type	Quality of Evidence
Can UNHS accurately diagnose moderate-to-profound sensorineural hearing impairment?	OAEs and ABR are highly accurate screening tests for congenital PHL (sensitivity, 84%; specificity, 90%).	Cohort or case-control analytic study	Good. One controlled trial measured the predictive value (6.7%) of a positive test result, and 1 good-quality cohort study measured sensitivity and specificity against an independent gold standard.
In UNHS programs, how many children are identified and treated early?	UNHS increases the chance that diagnosis and treatment will occur before age 6 mo. UNHS increases early identification between 19% and 42% over selective screening in high-risk children.	Controlled trial without randomization; cohort or case-control analytic study	Good. One cohort study in the United States and 1 controlled study in the United Kingdom reported the frequency of treatment before ages 6 and 10 mo, respectively. Other studies did not provide sufficient information, and none included patients who, although screened, were diagnosed and treated late because of loss to follow-up. However, no controlled trials of UNHS vs selective screening have been done.
Does identification and treatment prior to age 6 mo improve language and communication in infants who would not be diagnosed that early in a selective, high-risk screening program?	Evidence is inconclusive.	Cohort or case-control analytic study; multiple time series, dramatic uncontrolled experiments	Fair/poor. Studies have selection bias and baseline differences between compared groups. These studies did not specifically describe outcomes in the subgroup of children who would be identified by UNHS but not by selective screening.
What are the potential adverse effects of screening and early treatment?	Evidence is inconclusive.	Opinions of respected authorities	Poor. Most postulated adverse effects have not been evaluated in studies.

*UNHS indicates universal newborn hearing screening; OAEs, otoacoustic emissions; ABR, automated auditory brainstem response; and PHL, permanent hearing loss.

tion of newborns with PHL, but as many as 10% of newborns with normal hearing will require a second screening test. From 1% to 3% of newborns will be referred for audiological assessment; over 90% of those referred are false-positives. The consequences of false-positives have not been adequately evaluated, nor has the reliability of audiological and behavioral assessment⁶⁴—the standard used to make decisions about treatment—been established. Moreover, the false-negative rate is higher than previously thought, probably 20% to 30% in most programs. This new finding calls into question the assumption that a newborn who passes a screening test has normal hearing. It also suggests that stricter pass criteria, which have been promoted as a way to reduce false-positives, also reduce the effectiveness of screening.

A clearer picture of the consequences of delayed diagnosis in low-risk newborns would strengthen the case for universal screening. While diagnosis of hearing impairment is often delayed in children with congeni-

tal hearing impairment, the risk of delayed language development in otherwise healthy infants diagnosed at ages 1 or 2 years is not known. Because the frequency and severity of poor language outcomes in this group is uncertain, only adequately controlled trials or cohort studies can establish the efficacy of early intervention.

Several cohort studies show that, by ages 2 to 4 years, children with hearing aids and other therapy in the first 6 months of life had better language skills than those treated later. None of these studies compared an inception cohort of newborns offered UNHS with infants managed by usual care (including selective screening). Additionally, these studies had unclear criteria for selecting subjects, making it impossible to exclude baseline differences in the compared groups as a cause for the association of early identification with language development. The hypothesis that early intervention is a predictor of language acquisition is plausible, but the studies do not establish that screening low-risk newborns is the

important factor. None of the studies attempted to link short-term improvements to better function later in life.

As use of UNHS rapidly increases, it is important to conduct longitudinal studies of UNHS to address these knowledge gaps. Further randomized trials of UNHS seem unlikely to be conducted in the United States. Although it would be possible to compare states with and without UNHS, such studies would be prone to uncontrollable confounding due to differences among states. However, better evidence about the effectiveness of UNHS is needed and could be obtained via population-based studies that begin with inception cohorts and carefully report outcomes in all possible patients, as well as rates of loss to follow-up. Speech, language, and scholastic achievement of deaf and hard-of-hearing children should be followed over time. States that have UNHS should conduct such population-based studies to evaluate whether the long-term language outcomes of deaf children improve as the age of identification decreases.

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Obtained funding: Helfand.

Administrative, technical, or material support: McPhillips, Davis, Lieu, Homer.

Study supervision: Thompson, Davis, Helfand.

Funding/Support: The study on which this article is based was conducted by the Oregon Health & Science University Evidence-Based Practice Center, under contract 290-97-0018 to the Agency for Healthcare Research and Quality.

Also Available: A longer and more detailed systematic evidence review of UNHS will be available at <http://www.ahrq.gov/clinic/serfiles.htm>.

Disclaimer: The authors of this article are responsible for its contents, including any clinical or treatment recommendations. No statement in this article should be construed as an official position of the Agency for Healthcare Research and Quality or the US Department of Health and Human Services.

Acknowledgment: We thank Jennifer Sundheim, MLIS, and Michelle Honore, BS, at the University of Washington and Kathryn Pyle Krages, AMLS, MA, Susan Carson, MPH, and Susan Wingenfeld, BA, at Oregon Health & Science University for their assistance on this project.

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Words are chameleons, which reflect the colour of their environment.
—Learned Hand (1872-1961)