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Levels of evidence: universal newborn hearing screening (UNHS) and early hearing detection and intervention systems (EHDI)

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Abstract

Levels of evidence differ according to the audience addressed. Implementation of universal newborn hearing screening requires responses to a complex myriad of diverse groups: the general public, families with children who are deaf or hard of hearing, the deaf and hard of hearing communities, hospital administrators, physicians (pediatricians, general practitioners, ear nose and throat physicians, geneticists), managed care, Medicaid, insurance agencies, and politicians. The level of evidence required by medical/health agencies and task forces may differ from the levels of evidence available in education and intervention. Issues related to the low incidence of the disability, the lack of a normal distribution within the disability study, the obstacles to random assignment to treatment, and designs that include a control group with "no treatment" have implications legally and ethically for the professional providing services to families and children who are deaf or hard of hearing. This session will discuss issues related to "convenience samples," number of subjects included in research studies, and the population required to obtain a large enough sample of children with low-incidence disabilities. The level of evidence required to demonstrate sensitive periods of development, which are a critical element for justification of implementing a universal newborn hearing screening includes both behavioral and neurological information. Sensitive periods may have different duration for different aspects of development, such as social-emotional development, auditory and speech development, or language development. Further complicating the question of sensitive periods of development are the distinct possibility that different sensitive periods exist for development of age-level vocabulary, for establishing English phonology, or for mastering English syntax. Research outcomes provide evidence that age of identification of hearing loss is reduced, that age of intervention initiation is lowered, and that the outcomes of intervention are better because of the establishment of a screening program. Most professionals in communication disorders believe that screening is not the actual cause of better developmental outcomes but that the age when children begin to have access to language and communication and the characteristics of the

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intervention are the primary cause of better outcomes. Screening is the avenue through which access to quality intervention is made available. The research still remains at an infant level of development such that there is very little evidence for the efficacy of specific characteristics of the intervention provided. **Learning outcomes:** (1) The learner will be able to identify the obstacles in conducting research on the effectiveness of intervention of children identified through universal newborn hearing screening programs. (2) The learner will be able to identify the type of research on developmental outcomes of children with early-identified hearing loss.

Keywords: Hearing loss; Early identification

Levels of evidence differ according to the audience addressed. Implementation of universal newborn hearing screening requires responses to a complex myriad of diverse groups: the general public, families with children who are deaf or hard of hearing, the deaf and hard of hearing communities, hospital administrators, physicians (pediatricians, general practitioners, ear nose and throat physicians, geneticists), managed care, Medicaid, insurance agencies, and politicians.

The US Preventative Services Task Force (USPSTF) has the task of analyzing medical procedures and determining the level of evidence for procedures that ultimately influences standards of health care and payers of health services such as Medicaid, Medicare, managed care and health insurance companies. This paper is written to address levels of evidence and will include a response to the USPSTF (2001) paper, ''Recommendations and Rationale Newborn Hearing Screening.'' The reader is referred to the original (http://www.ahrg.gov/clinic/3rduspdtf/newbornscreen/newhearrr.htm). The American Speech-Language-Hearing Association has responded to the USPSTF paper (see http://www.audiologyonline.com/audiology/newroot/news/displaynews.asp?id=342) as has the American Academy of Audiology (see http://www.audiologyonline.com/audiology/newroot/news/displaynews.asp?id=341).

1. Commonly asked questions

1.1. Does universal newborn hearing screening (UNHS) result in earlier identification of the hearing loss?

The USPSTF (2001) stated that, ''Studies of statewide universal newborn hearing screening programs in the United States have found that the mean age of identification of hearing impairment has decreased from 12 to 13 months before screening programs were introduced to 3–6 months since their introduction.''

Prior to UNHS, the average age of diagnosis of hearing loss in Rhode Island was 20 months, not 12–13 months (Vohr, Carty, Moore, & Letourneau, 1998). No public or private program has ever published data indicating that the average age of identification of hearing loss prior to universal newborn hearing screening was 12–13 months.

The level of evidence required by the USPSTF for this statement was a misinterpretation of the Rhode Island data and the Harrison, Roush, and Wallace (2003) study that analyzed

parent surveys from 150 families who were selected because they were enrolled in programs that were on the Alexander Bell Association's list. The authors state that prior to UNHS, the age of identification of hearing loss exceeded 2 years of age. Mace, Wallace, Whan, and Stelmachowicz (1991) reviewed the literature on age of identification of hearing loss prior to UNHS and found that the average age of identification of hearing loss ranged from 19 to 36 months of age.

1.2. Does UNHS result in earlier provision of intervention services and treatment?

One important issue to be addressed is the definition of treatment. Does treatment mean medical management or audiologic management (i.e., identification/diagnosis, amplification fitting, and therapeutic intervention for speech, language and social-emotional development)? The treatment after identification of permanent sensorineural hearing loss is typically not medical and does not involve medication or surgical procedures. For almost all children with permanent hearing loss, initiation of intervention begins with hearing aid fitting. Medical intervention for hearing loss occurs as intervention when the hearing loss is medically treatable such as in non-permanent, fluctuating hearing loss caused by otitis media or other fluid in the middle ear cavity, such as amniotic fluid. Medical intervention for hearing loss can also occur if the child is a candidate for cochlear implantation, but even at the earliest, surgical management does not occur until 6 months of age. Most infant implantations occur after the age of 12 months, months after the initiation of educational intervention services.

1.3. Does universal newborn hearing screening reduce the age at which hearing aids are fit?

The USPSTF report (2001) stated, ''The mean age at which hearing aids are fit has been reduced from 13 to 16 months before screening programs were introduced to 5–7 months.'' The studies do not indicate that the mean age at which hearing aids are fit has been reduced from 13 to 16 months before UNHS programs were introduced to 5–7 months after UNHS programs were introduced. Since the average age of identification of hearing loss prior to UNHS was after 2 years of age, the mean age of hearing aid fitting could not have been 13–16 months. The only way to obtain an accurate indication of mean age at hearing aid fitting would be to examine at least 7 years of state data in order to include the many children who are diagnosed later in life. The state surveys conducted by the Marion Downs National Center indicate that age of identification of hearing loss and age at amplification fitting were much more delayed than reported by the USPTF. Again, if the USPSTF uses data that is restricted to certain types of programs rather than epidemiological data, the results can be very different from actual state or national data.

Epidemiological data from the United Kingdom is probably the most comprehensive data on age of identification of hearing loss on a large population of children. Prior to UNHS, the average age of suspicion of hearing loss was 18.8 months, confirmation of hearing loss was 26 months, the average age of prescription of hearing aids was 30 months, and the average age of fitting of hearing aids was 32 months (42.3 months for moderate hearing loss, 23.5 for severe hearing loss and 13.9 months for profound hearing loss; Davis

et al., 1997). The United Kingdom had a universal distraction behavioral screening test at 9 months of age, which did not universally exist in the United States. Thus, their average age of identification of profound hearing loss was younger than would be obtained in the United States during the same period of time.

The average age at hearing aid fitting for the state of Colorado is 5 weeks of age. This is a dramatic reduction from the USPSTF age range of 13–16 months which reflects the average age of identification of those children fortunate enough to be enrolled in early intervention programs but misses the significant number of children identified after 5 years of age, particularly those with milder hearing loss.

1.4. Does universal newborn hearing screening result in improved speech, language or educational development?

The USPSTF (2001) stated, ''There are no prospective, controlled studies that directly examine whether newborn hearing screening and earlier intervention result in improved speech, language or educational development.'' This statement implies that newborn hearing screening itself can result in improved speech, language or education development. This assumption fails to acknowledge the role that educational intervention and counseling may play in improved speech, language or educational developmental. Professionals in the field of hearing loss would argue that early identification by any means with appropriate and immediate early intervention services has the potential to result in improved speech, language or educational development as long as appropriate and immediate educational intervention services are offered. Professionals with experience educating children who are deaf or hard of hearing do not believe that screening or identification by itself result in improved developmental outcomes. Quality intervention services provided immediately upon early diagnosis of the hearing loss is, in all likelihood, the primary cause of better developmental outcomes. Several studies both within the United States and internationally in the United Kingdom indicate that children who were early-identified who did not receive immediate amplification and intervention services did not have better developmental outcomes than those that were later-identified. In the United Kingdom, families with early-identified children who were highly dissatisfied with intervention follow-through indicated a lower quality of family life and a lower quality of life for the child in a retrospective study of children 18 years and older (Hind & Davis, 2000).

It is important to define the purpose of universal newborn hearing screening, whether it is to insure optimal developmental outcomes for the life of the child, to insure earlier identification of the hearing loss, to insure earlier intervention initiation for the child and family, to insure early access to communication and language development, and/or to insure early access to hearing. Universal newborn hearing screening begins as a part of the medical/health system but outcome is dependent upon the educational system. One could argue that the purpose of medicine and health is to insure that the child has an opportunity for equal access to education, not necessarily that the developmental outcome will be optimal through the entire continuum of childhood.

There is a new population of early-identified children with significant hearing loss, whose language levels are within the low average range from infancy through 6 years of age (Pipp-Siegel, Sedey, & Yoshinaga-Itano, 2001; Yoshinaga-Itano, Coulter, & Thomson,

2000, 2001; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998). There has never before been a ''population'' of children with significant hearing loss who have achieved average age-appropriate language skills in the history of the research literature. These children have done well, and their hearing losses were all identified early. If their language levels are not a result of early identification programs, there is no other variable that can explain the results since the children had the same access to intervention, the same intensity of intervention and the same intervention providers as the later-identified children. These children with early-identified hearing loss, and no secondary disabilities have an 80% probability of language development in the low average level in their first 5 years of life (Yoshinaga-Itano, Coulter, & Thomson, 2001).

Therefore, although not a prospective study from screening or controlled random assignment to screening study, the conclusion is clear; on average, children with hearing loss and no additional disabilities, when identified early and when treated with appropriate intervention have the ability to develop language skills within the normal range of development.

A follow-through study, such as that recommended by the USPSTF would cost millions of dollars and would likely require several generations of children. Statistics on children with significant hearing loss indicate that 500–600 must be screened to find a single child with bilateral permanent hearing loss. Simply, the costs of the recommended study would be prohibitive. Longitudinal follow-through of children past the age of 5 years has never been done with any population of children with significant hearing loss with the exception of very small sample sizes, making generalization to the population at large very difficult. The study the USPSTF proposed would be improbable, but reasonable conclusions can be drawn without waiting for such a study to be funded and completed. If the assumption that the quality of intervention provided within a sensitive period of development is the cause of the improved developmental outcomes is accepted, a study that has no control over the type of intervention, the quality of the intervention, the training of the professionals, the type and education of professionals providing service, or the components of the intervention program would have significant confounding uncontrolled variables. Thus, assessing a few children from many programs throughout the United States would have little control over the quality or the impact of intervention services or intervention providers. It would be difficult to interpret the results of such a study should aggregate data be taken from many state systems in order to obtain a sufficient number of children. Only if this assumption is incorrect, would such a study yield interpretable data. If the improved developmental outcome was a result solely of universal newborn hearing screening and the timeliness of identification of hearing loss, and the type and quality of intervention were not critical, then improved outcomes would be obtained. However, lack of improved developmental outcomes could be the result, not of a failed universal newborn hearing screening program, but of failure of the follow-through system to insure immediate and accurate identification of the hearing loss, immediate and accurate fitting of amplification, and immediate and appropriate intervention services.

Based upon the state of Colorado 2002 statistics, 70 children with bilateral permanent hearing loss were identified in a population that exceeded 63,000 screened newborns. The return-to-follow-through rate was over 80%. Of these 70 children, 21 had a bilateral conductive hearing loss and 49 had a bilateral sensorineural hearing loss. About half of the

bilateral conductive hearing losses were permanent. Fifteen of the 70 children had mild hearing losses. Approximately 40%, 24 of the 60 children with permanent hearing loss, had additional disabilities. Five of the children had bilateral auditory neuropathy. This leaves 31 children as potential research subjects for a longitudinal study. These children were further divided into at least four categories of hearing loss: mild, moderate, severe and profound, with proportionately more children in the moderate and severe categories. They were then further divided by socio-economic variables. In order to be able to use all 31 of the potential research participants, the study would have to accept both conductive and sensorineural hearing loss. Assuming that there are 31 potential research candidates for a longitudinal study, in order to obtain 294 children, it would take 100% capture for 10 years and screening of 630,000 infants in the state of Colorado.

1.5. Has the research identified all the variables that might contribute to successful or unsuccessful language development?

The US Preventative Task Force questions whether there were other factors that might contribute to language development. ''Other factors, such as family involvement (an important contributor to language development), the degree of other disabilities, or the quality of pediatric care, might have influenced the time of identification and the language outcome'' (2001).

In educational literature the ability to account for 50–70% of the variance in development is rarely seen, predominantly because there are so many variables that usually contribute to developmental outcome and not all of them can be included in a single research design. Researchers in language development and significant hearing loss are continually searching for variables that will account for additional variance in outcome. However, when non-verbal cognitive development and age of identification of the hearing loss account for over 50% of the variance, and family involvement and family interaction variables account for additional variance, the other variables such as method of communication, degree of hearing loss, and socio-economic variables, either add no additional variance or only a very small amount.

The question of whether children who are not screened at birth would be identified earlier because of family involvement is an interesting one raised by the Task Force. Family involvement, as described in the literature, is measured after diagnosis of hearing loss and when the family is involved in intervention services. Hearing loss is an ''invisible disability'' and is typically not identified by the most caring and involved parents until language and speech fail to develop. Children with hearing loss can fool their parents simply by being very visually aware of their environment. To imply that a child would be identified later because parents are not involved is as inappropriate as implying that a child is identified late because the managing physician is ''unconcerned'' or has not provided appropriate care. Testimony from physicians who are involved parents, whose own children were late identified with hearing loss, can attest to the fact that hearing loss is an ''invisible disability.''

Early-identified children have families who are rated higher in family involvement than families with later-identified children (Moeller, 2000). There are probably a whole host of variables that contribute to this effect. Some researchers have also found a very high

relationship between family involvement and socio-economic status. One of the striking findings of the early-identified children is that, at least in the early years of life, the impact of socio-economic status is either eliminated or compensated for, by early intervention services, such that socio-economic status has not been found to be a primary predictor variable of language outcome in the first 5 years of life.

The degree of other disabilities does factor into the time of identification. Children being treated for other medical conditions are often tested for hearing loss, particularly if the medical conditions are typically associated with hearing loss. Early-identified children with milder hearing losses are only identified early without universal newborn hearing screening (UNHS) programs if they have other disabilities, and are being screened for hearing because they are being followed medically for other developmental needs. The children without additional disabilities may appear to respond to sounds in the home. Speech and language delays would not be evident until the child is 2–3 years of age at the earliest, but most typically after the age of 5 years.

The quality of pediatric care is an interesting question. Since the average age of identification of congenital hearing loss in the United States has been very late, typically after 2 years of age, this question seems to have been answered. Without UNHS programs, even the highest quality of pediatric care appears unable to result in the diagnosis of hearing loss in the newborn period, or even in the first 12 months of life. The US Preventative Task Force fails to identify one study that shows that identification of hearing loss for the average child can occur within the first 6 or 12 months of life through ''usual care.''

1.6. Does universal newborn hearing screening results in better outcomes than targeted screening or typical care without screening?

The US Preventative Task Force has questioned this proposition. In a traditional design, if we were to answer this question, all babies would have to be randomly assigned to one of three groups: (1) Traditional/typical care (no screen); (2) Targeted screen, perhaps based on high-risk factors; or (3) UNHS screen. In traditional medical and other clinical efficacy studies, patients can be randomly assigned to either a control (i.e., no treatment) group or an experimental (i.e., treatment) group.

However, babies assigned to these groups would have to be followed prospectively at least into elementary school to determine whether they would eventually be identified with hearing loss. Because of the low incidence of hearing loss and the exclusion of specific groups of children with hearing loss from consideration, I estimate that 20,000 children in each group would have to be followed to get even a small number of children in each of the three groups.

Importantly, federal law prohibits random assignment to treatment, in the timing of the initiation of treatment and in the type of treatment. Additionally, families have the right to choose whether they desire intervention, and if so, the intervention they wish. Thus, random assignment to one of the three groups is not legally possible. Further, identification of hearing loss with concomitant failure to disclose this information to the parents, in order to randomly assign their child to the ''no treatment'' group, would also be prohibited by law. The USPSTF does acknowledge that such a design could probably not be

accomplished. Further, a control group that naturally emerges in the rare instance that a family chooses not to receive treatment would be characteristically different from the experimental group resulting in unequal comparison groups by characteristics of the family and child and also in number of families.

Further exacerbating the problem is the fact that the characteristics of the family and child may influence the intervention choices. For example, families with children who have mild hearing loss tend to choose more auditory/oral interventions because their children have significantly more benefit from amplification. Many families with children who have multiple disabilities choose a simultaneous communication mode. The impact of commitment of the family and the skill of the family in the intervention of choice are variables that have previously been found to affect developmental outcome. Random assignment to treatment would ignore these variables.

The USPSTF suggests that an inception cohort of newborns offered UNHS with infants managed by usual care (including selective screening) should be compared. In order for these comparisons to be made in a meaningful manner, a sufficient number of infants must be identified both in the traditional care versus the UNHS group.

The best estimate is that the minimum number of infants required would approach about 120,000 infants, to obtain a cohort of 30–50 eligible participants (i.e., those with no secondary disabilities and with bilateral moderate to profound permanent hearing loss) in each group. These infants would have to be followed for their entire educational career to demonstrate ''better function later in life.'' Of course if the study was initiated this year (2002), and if we accepted the termination of their college BA/BS degrees as the endpoint of the study, in 2024 we can expect to analyze the resultant data.

Importantly, random assignment would in no way guarantee equal groups by socioeconomic status, cognitive quotients, degree of hearing loss, gender, ethnicity, or presence of an additional disability, etc. Random assignment results in equal comparison groups only when the population sampled is normally distributed, and hearing loss is not. The variability of the population would potentially necessitate doubling the number of infants in the study from 120,000 to 240,000 to guarantee sufficient numbers for statistical analysis.

The USPSTF (2001) assertion that previous studies do not establish that ''screening low-risk newborns'' is an important factor because there is not a control group of newborns with usual care. The fact that no cohort of early-identified children (i.e., identified in the first 6 months of life) has existed in the literature and they do, indeed, now exist, addresses this issue and should suffice. Research indicates that 50% of the infants born with hearing loss are missed through high-risk register screenings. In countries that have used a universal ''distraction testing,'' a visual reinforcement head-turning screen, at 9 months of age (see data from the UK, Australia, Scandinavia, Germany, and Holland) report an average age of hearing loss identification between 2 and 2.5 years.

1.7. Do the samples of early-identified children represent biased samples?

The USPSTF asserted that samples of participants in the studies of early-identified children could represent a biased sample. The USPSTF (2001) stated, ''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.'' Since the Colorado Home Intervention Program serves over 90% of the children with hearing loss in the state between the ages of birth and 3 years of age, it is unlikely that selection bias affected the results of the Colorado studies. The Colorado Home Intervention Program outcomes are prospective studies of the language outcomes of the *population*, not a sample of children in Colorado followed from identification of hearing loss. Today in Colorado, the identification of hearing loss is a direct follow-through from universal newborn hearing screening. The data management in the early years of UNHS in Colorado did not include child identification information, but included aggregate data, numbers born, numbers screened, numbers passed and referred. Parents, therefore, reported whether their infants had participated in UNHS. A few articles include longitudinal data analysis although the vast majority of the data published so far are cross-sectional.

In the Colorado studies, the later-identified groups were children identified through typical care or the high-risk register. Since screening began in 1992 in the state of Colorado, all children born before 1992 were identified through either typical care, high-risk register, or a targeted screen in a newborn intensive care unit.

In several studies (Apuzzo & Yoshinaga-Itano, 1995; Yoshinaga-Itano et al., 1998) and subsequent Colorado studies, children with multiple disabilities were included in the research designs. The inclusion of these participants makes the results of the studies generalizable to a larger population, but also makes the probability of finding an effect much less likely.

The Yoshinaga-Itano et al. (2000) study compared (a) children identified through universal newborn hearing screening, (b) children unscreened between 1992 and 1999, and (c) children unscreened through UNHS because they were born prior to the initiation of screening in Colorado. It should be noted that the actual average age of diagnosis of the unscreened children is probably much higher because only children enrolled in the early intervention program, who were identified with hearing loss in the first 3 years of life were included. Children identified through preschool screening or school-screening programs for the first time would not be included in this data. Thus, children with mild and moderate hearing losses who were identified later in life are not represented in these statistics, making the average age of identification for this population lower than the actual state statistics.

An early-identified child from selective screening (i.e., newborn intensive care screening, distraction testing, or high risk register screening) has the same outcomes as those identified through UNHS (Yoshinaga-Itano et al., 2000; Yoshinaga-Itano, Coulter, & Thomson, 2001). The early-identified children in the Apuzzo and Yoshinaga-Itano (1995) study were children identified through high-risk register. Their outcomes were virtually identical to those from both the 1998 (Yoshinaga-Itano et al., 1998) and 2000 studies (Mayne, Yoshinaga-Itano, & Sedey, 2000; Yoshinaga-Itano et al., 2000). When considering only children with cognitive quotients of at least 80, 31.6% of the children in the ''not screened'' group, 62.1% in the ''probably not screened'' group (parent report of ''no UNHS'' but no state records), and 44.3% of the children born before 1992 had language quotients that were 80 or above. This compares with the 82.4% in the ''screened'' group and 81.0% in the ''probably screened'' group (parent report of UNHS but no state records) who had language quotients that were 80 or above.

1.8. Does attrition of subjects and failure to follow-up bias the samples of the existing literature?

The USPSTF (2001) reported, ''Studies had unclear criteria for selection of subjects. 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.''

In the Colorado cohort, over 90% of the children for whom early intervention is recommended enroll in the Colorado Home Intervention Program. Most of the remaining 10% enroll in other intervention services. Some of these children are children with multiple disabilities and hearing loss is not the primary disability. Others enroll in private intervention programs or school-based programs. However, many of these providers also participate in the outcome data collection. Over the last 20 years of data collection, there has only been a 5% attrition rate. Attrition typically occurs because the family moves out of state or because of a death. Since the program is a publicly funded program, and the program is state-wide, very few children are lost to follow-up. Early identified children who do not continue follow-through in the program are typically those who are no longer receiving special educational services because their developmental levels are so high that they no longer qualify for services. Thus, if we are missing any early-identified children, they are most likely the ones with the highest functioning levels. All children in the state who had been early-identified were included as potential subjects for these studies. Seventy percent of the available participants (i.e., children identified with hearing loss as a result of UNHS) were included in the study.

The USPSTF report states, ''The diagnosis of congenital hearing loss is often delayed. In one survey conducted before hearing screening was common, the median age at diagnosis was 13 months for infants with severe to profound bilateral SNHL and 17 months for those with mild-to-moderate hearing losses'' (Harrison & Roush, 1996).

However, studies using surveys of early intervention programs throughout the United States cannot be used as an index of the average age of identification of hearing loss. While surveys provide valuable information for the profession, they were not designed as an accurate measure of the average age of diagnosis of the hearing loss in the United States. The Harrison and Roush (1996) study analyzed surveys sent to intervention programs throughout the United States. As in most survey studies, a small percentage of those contacted responded to the surveys. Early intervention providers were asked to fill out a response indicating the age at which children in their program were typically diagnosed. No audiogram data was submitted to verify the information provided. Probably the most significant issue is that the programs only enrolled children between the ages of birth and 3 years of age, therefore, any child identified after the age of three would not be included in the average age of identification. The characteristics of the intervention programs (public vs. private) responding may also influence the data obtained. All of the programs that responded were on a mailing list of the Alexander Graham Bell Association, an association that promotes oral education of children with significant hearing loss. Since 80% of the children who are deaf or hard of hearing are educated in programs that use some form of sign language, it is highly probable that many of the programs that responded were private intervention programs. Additionally, the

lack of data verification through initial audiogram reports or interview could result in huge errors in the reporting. The respondents did not report the actual number of children in each program who were identified at specific age ranges. Surveys provide valuable information, but they do not provide the epidemiological data that would be required to use the information as a determination of the average age of identification of hearing loss prior to UNHS. Although the survey was conducted before UNHS was common practice, it is highly possible that many of the children enrolled in these early intervention programs had been identified through UNHS programs as programs did exist in the United States at this time (i.e., 60% of the children in the state of Colorado were being screening for hearing in the newborn period).

Yoshinaga-Itano et al. (2001) reported that 84% of the children in the ''screen'' group were identified by 6 months of age as compared to 8% in the ''no screen'' group. Sixteen percent in the ''screen'' group were identified after 6 months of age, while 92% in the ''no screen'' group were identified after 6 months of age. This information was verified through audiological reports and the data was checked by two certified audiologists.

1.9. Can the cost of universal newborn hearing screening be justified according to the yield and the benefit?

The USPSTF concludes that the cost of screening children in the well-baby nursery has too low of a yield to be cost-effective. Current procedures miss 50% of the children with congenital hearing loss. These children have the highest potential for the greatest benefit from early-identification. The USPSTF further states that, ''To use a hypothetical example, if 50% of the newborns with permanent hearing loss (PHL) would have poor language ability if diagnosed after age 10 months, and early intervention reduces this by 50%, then the newborn neonatal screening (NNS) to prevent 1 additional case of delayed language acquisition would be 6771'' (2001).

The babies with congenital hearing loss who are from neonatal intensive care units have a significantly greater probability of secondary disability, approximately 60% of the population. There are very few babies born with hearing loss in the well baby nursery who are missed through universal newborn hearing screening. At the present time, there is no reason to believe that the babies who later are identified with hearing loss had the hearing loss congenitally. It is more probable that the babies were born with normal hearing and progressed to children with hearing loss.

This statistic only includes moderate to profound bilateral hearing loss. The argument here is flawed because there is no research evidence that 50% of the newborns with PHL would have poor language if diagnosed after age 10 months. In fact, the statistic is more likely greater than 65% of the population and may approach 80%. The comparison groups in all of the studies compared children who were enrolled in early intervention programs and therefore, identified by 3 years of age. They excluded children who were identified after 3 years of age. Only a small percentage of the children with significant hearing loss have age appropriate language levels as they enter adulthood. This statistic is further flawed because the population statistics include 40% of the population with additional disabilities, and if children with hearing loss only were separated from these statistics, the impact would be even greater.

Rather than looking at whether the cost is worth 1 case in 6771 screened, the emphasis should be placed upon the majority of children with significant hearing loss having the opportunity for normal language development. In actuality, the majority of children have language development between 50 and 60% of chronological age and these language quotients either remain or worsen as the child ages.

The cost of later-identification of hearing loss goes well beyond the initial birth period. The cost to the individual child and family, the educational system, and the aggregate cost to society is too high, when only about 16–20% of the later-identified children achieve and maintain age-appropriate language levels.

1.10. Does the fact that a significant portion of children with congenital hearing loss are children from the neonatal intensive care units result in the conclusion that the cost of screening universally is unjustified?

The USPTF (2001) found good evidence that the prevalence of hearing loss in infants in the newborn intensive care unit (NICU) and those with other specific risk factors is 10–20 times higher than the prevalence of hearing loss in the general population of newborns. Both the yield of screening and the proportion of true positive results will be substantially higher when screening is targeted towards these high-risk infants, but selective screening programs typically do not identify all infants with risk factors. Evidence that early identification and intervention for hearing loss improves speech, language, or auditory outcomes in high-risk populations is also limited.

Although yield is higher in the NICU, children with risk factors only account for 50% of the population of children with hearing loss. Thus, targeted screening would have a high yield of identification of hearing loss per child screened, but would miss 50% of the children. High-risk screening has existed in the United States for decades. It has failed to identify a large cohort of children in the first year of life. Further, those children identified in the first year of life have a significantly higher incidence of secondary disabilities (approximately 66%) than the children identified through UNHS and the well-baby nurseries (about 30%). Thus, the children with the highest potential for success, children with hearing loss only, are the most likely to be missed.

1.11. Does the potential harm of false-positive tests of low-risk infants outweigh the social-emotional benefits to families with later-identified children with hearing loss?

The USPSTF stated that, ‘‘Although early identification and intervention may improve the quality of life for the infant and family during the first year of life, and prevent regret by the family over delayed diagnosis of hearing loss, the USPTF found few data addressing these benefits. The USPTF could not determine from existing studies whether these potential benefits outweigh the potential harms of false-positive tests that many low-risk infants would experience following universal screening in both high- and low-risk groups’’ (2001).

The literature does not indicate that there are potential harms of false-positive tests. There has been no evidence that newborn hearing screening causes parental harm since parental stress levels of the ‘‘false positive’’ referral group are similar to those present in the

general population and to those families whose babies have not been screened. There is a body of evidence that screening does not result in parental harm when measured by traditional instruments of parental stress and depression. A summary of the literature indicates the following findings: (1) families of children who are screened for hearing and those whose children are not screened do not differ significantly in their stress levels; and (2) families of children who are screened and pass and families of children who are screened and refer do not differ significantly in their stress level. Approximately 6–13% have high levels of stress in both groups. This level of high stress is the level of stress found in the general public. Finally, families of children with hearing loss identified through UNHS and those identified through other means do not differ in their stress levels. No studies have reported that UNHS results in increased family stress when compared to families of children who have never been screened or families of children who have passed neonatal screening.

Colorado data in 2002 indicates that of the 66,145 infants screened, 4% were referred for additional testing and 85% returned for follow-through. Of these 4%, only 272 children were referred for full audiological evaluations. Of the 272 children referred for full audiological evaluations, 200 were evaluated by 3 months of age, and 108 children were diagnosed with a significant hearing loss with an average age of 8 weeds at diagnosis. Thus, the positive predictive value at the time of the full audiological evaluation was 50%, or one in every two children evaluated.

Studies demonstrating a better quality of life as a result of UNHS, early identification and early intervention have not yet been conducted. It is a reasonable assumption that as earlier identification and intervention have a positive impact upon the developmental outcomes of children with significant hearing loss, the quality of life of the family should be significantly improved.

Higher language level is related to earlier identification and intervention and higher cognitive levels. Children with higher language levels have better maternal bonding. Children with higher language levels have better personal-social development.

2. Summary

The Colorado studies investigated the relationship of universal newborn hearing screening through logistical regression statistical techniques on a population of 294 children with significant hearing loss. All children meeting the selection criteria were included in the study. The study included children with hearing loss only and excluded those with multiple disabilities. In addition, the Colorado studies investigated the question of early-identification and early intervention for children with hearing loss and developmental outcomes through studies that included matched designs, multi-variance analysis of covariance statistical designs, and multiple regression techniques using both step-wise and block designs. All studies, regardless of the number of participants, resulted in the same results, a robust and repeatable impact of early identification and initiation of intervention in the first 6 months of life on developmental outcomes. Although the USPSTF believes that statistical analysis and experimental group statistical control are not sufficient, the effects were so significant that statistical analysis was unnecessary to demonstrate the impact. UNHS

programs result in earlier identification of the hearing loss, earlier initiation of intervention including fitting of amplification, and better outcomes for the child and the family in language, speech and social-emotional development.

Appendix A. Continuing education

1. Research using experimental control groups of children with identified hearing loss randomly assigned to treatment versus non-treatment are unlikely to be done in the US because:
   a. Federal legislation requires that treatment be offered to parents after the diagnosis of a disability within 48 h after the diagnosis
   b. Parent commitment to the treatment is a confounding variable that cannot be controlled in random assignment
   c. Treatments are complex involving counseling, language therapy, speech and auditory therapy, social-emotional interventions—controlling a single aspect of a treatment is difficult
   d. All of the above
   e. None of the above
2. Which of the following claims was not a conclusion of the USPSTF:
   a. Neonatal hearing screening resulted in earlier identification of hearing loss
   b. There is evidence that neonatal hearing screening results in earlier hearing aid fitting
   c. Neonatal hearing screening results in earlier initiation of intervention
   d. Neonatal hearing screening results in better speech and language outcomes
   e. None of the above
3. Which of the following is NOT TRUE:
   a. Social-emotional harm is likely to result from low referral rates from universal newborn hearing screening programs
   b. Screening of neonatal intensive care units has a 10–20% higher yield of hearing loss than the well-baby nursery
   c. The USPSTF asserts that hearing aid fitting prior to universal newborn hearing screening was between 12 and 16 months of age
   d. Age of identification of hearing loss in the United Kingdom prior to universal newborn hearing screening as 26 months with an average age of hearing aid fitting at 30 months of age
   e. None of the above
4. The average age of identification of congenital hearing loss in the United States prior to the establishment of universal newborn hearing screening was:
   a. The USPSTF asserts that the age of identification prior to universal newborn hearing screening is 12–13 months
   b. Unable to establish precisely because no systematic reporting system for pediatric diagnosis of hearing loss existed
   c. Inaccurate and incomplete if obtaining reports from intervention programs that only enrolled children to age three

d. All of the above
e. None of the above

5. Which of the following is NOT TRUE: Research on language outcomes of early-identified children with earlier intervention:
   a. There is a strong relationship between language outcomes of later-identified children and parent involvement
   b. There is a 30% probability of normal language development for children with hearing loss and no multiple disabilities born in hospitals with a universal neonatal screening program.
   c. Age of identification not universal newborn hearing screening has been shown to be highly related to better language outcomes
   d. 31% of the children in the ''not screened'' group had language development within the normal range.
   e. All of the above

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