

Universal Newborn Hearing Screening Issues and Evidence*

by

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PREFACE

The following is the text of a presentation made at a workshop on Early Hearing Detection and Intervention in Atlanta, Georgia on October 22, 1997. The workshop was sponsored by the Centers for Disease Control and Prevention (CDC). Participants represented various academic disciplines and areas of responsibility and experience related to identifying and providing intervention to very young children with congenital hearing loss. This presentation was invited to set the context for the remainder of the workshop. The text of the presentation, along with the slides, are included here. Slides can be used in other presentations as long as credit is given to the National Center for Hearing Assessment and Management (NCHAM) at Utah State University. Information from this presentation should not be used in publications without permission of the author.

INTRODUCTION

There are two important things about the way in which this meeting has been organized. First is the diversity of the people who are participating and will have an opportunity to comment about issues related to early identification of hearing loss. In spite of the fact that most of us spend a good deal of our time thinking about issues related to newborn hearing screening, I am sure we will hear different points of view today than what we typically hear. The second point is that the meeting has been structured to encourage a free-flowing discussion of the important issues related to early identification of hearing loss.

To help you follow the large amount of material I will be presenting very rapidly, I have included in your packet copies of all the slides I will use, plus some additional slides, notes, and references. Some of the work I will cite is unpublished and should not be cited further without permission from the authors.

I have been asked to summarize the scientific evidence related to whether we should be doing universal newborn hearing screening as a way of identifying congenital hearing loss. To set the context for that discussion, [slide #2](#) is a picture of two babies identified with congenital sensorineural hearing loss at Woman's Hospital in Baton Rouge, Louisiana when they were less than six weeks old. They were fitted with hearing aids by the time they were three months old. Virtually everyone agrees that this is the kind of picture we should see more frequently.

[Slide #3](#) shows a baby being screened for hearing loss prior to being discharged from the hospital. Some people are very enthusiastic about the fact that we are seeing such pictures more frequently, but other people have deep concerns about it. In my presentation, I will summarize some of the most pertinent data on both sides of that issue. My purpose is not to convince you of a particular point of view, but rather to present evidence in a way that will lead to further discussion and analysis.

As shown in [slide #4](#), there has been a rapid increase in the number of universal newborn hearing screening (UNHS) programs which have been implemented in the U.S. since the NIH Consensus Development Conference on Early Identification of Hearing Loss held in 1993. At that time, there were only 11 hospitals screening more than 90 percent of their babies. Now, based on a survey which our Center does each year, there are almost 400 such programs. In other words, there has been an almost forty-fold increase during the last four and a half years.

That increase in the number of operational programs has been supported by a variety of different governmental and professional groups which have endorsed the concept of early detection of hearing loss and have recommended hospital-based universal newborn hearing screening as the most feasible method to identify hearing loss early (see [slide #5](#)). As shown in [slide #6](#), more and more states are also using legislative actions to address this issue. Five states now have legislative mandates requiring all babies to be screened for hearing before they are discharged from the

hospital. Three of those mandates were passed in 1997. At least eight other states will be considering similar legislation during 1998.

The focus of this presentation as shown in [slide #7](#) is whether the picture on the left (hospital-based universal newborn hearing screening) is the best way to achieve the picture on the right (detection of and intervention for hearing loss in babies before six months of age). Even though the focus is on the scientific evidence related to whether hospital-based newborn hearing screening should be the method of choice for early detection of hearing loss, it is useful to consider that evidence in the context of recent legislative activity and the rapid expansion of hospital-based newborn hearing screening programs.

Although there are obviously many people who believe that hospital-based universal newborn hearing screening is the solution, there are other very important groups who have considered the evidence and concluded that more work is needed ([slide #8](#)). For example, the U.S. Preventive Services Task Force Guide to Clinical Preventive Services, published in 1996 (see [slide #9](#)), concluded that even though "congenital hearing loss is a serious health problem associated with developmental delay in speech and language function, there is little evidence to support the use of routine, universal screening for all neonates." The Task Force Guide went on to say that universal newborn hearing screening programs would result in a substantial number of infants being misclassified, the costs and feasibility for UNHS programs are not fully known, and the evidence for the efficacy of early intervention with infants identified in such programs is incomplete.

To address the issue of whether we should be implementing UNHS programs or whether we should be considering other methods for early detection of hearing loss, information will be presented and organized according to six issues ([slide #10](#)). The purpose is to provide a common foundation for those who come from different perspectives to further discuss some of these important issues. After presenting the most relevant evidence related to each of those issues, the remainder of this meeting will be devoted to discussing where the evidence is inadequate and how CDC or other groups might proceed to gather evidence to address the existing gaps. The six issues to be discussed ([slide #11](#)) include:

- [Prevalence of congenital hearing loss](#)
- [Accuracy of newborn hearing screening methods](#)
- [Efficiency of existing UNHS programs](#)
- [Consequences of neonatal hearing loss](#)
- [Benefits resulting from earlier identification and intervention](#)
- [Cost-efficiency of hospital-based UNHS programs](#)
- [Summary](#)

PREVALENCE OF CONGENITAL HEARING LOSS

In discussing the prevalence of permanent congenital hearing loss (PCHL), it is important to consider definitional issues related to severity and type of hearing loss, the age of onset of that hearing loss, and whether the hearing loss is unilateral or bilateral loss (see [slide #12](#)). Consider first the results of 11 different population-based studies designed to determine the number of children with bilateral PCHL in population-based cohorts ranging in size from 10,000 to over 4 million children. Not surprisingly, as shown in [slide #13](#), the prevalence of bilateral PCHL is substantially higher when more lenient definitions are used. When children are required to have a 50 dB bilateral loss before they are considered to be hearing impaired, the prevalence is about one per thousand. However, when children with 30 dB bilateral losses are included, the prevalence increases to about 2.5 per thousand.

Each of these studies only included children with bilateral hearing losses. If children with unilateral losses had been included, the prevalence would increase by one-third to one-half as shown in [slide #14](#). Based on these data, we should expect to find 3 to 4 children per thousand with PCHL from well-run, hospital-based, UNHS programs. As shown in [slide #15](#), this is exactly what is being reported. Hospitals with UNHS programs in Rhode Island, Colorado, New York, Utah, Hawaii, and New Jersey, using a variety of different screening techniques and protocols, are reporting between 1.65 and 4.15 infants per thousand identified with PCHL.

It is important to note that the numbers reported from the retrospective studies shown in [slide #13](#) could have included children with late onset or acquired hearing losses. Many people assume that there are about as many young children with acquired losses as with congenital losses. However, states with long-established UNHS programs, where tens of thousands of infants have now been screened, are finding very few children with acquired losses. For example, in Rhode Island, Hawaii, Colorado, and Utah, hundreds of children have now been identified with PCHL. Many of those programs have been operating long enough that there are thousands of 7, 8, and 9 year olds who were screened as infants. If acquired hearing loss were as frequent as is often assumed, we should now be discovering dozens of children with late onset losses as they enter school. In fact, there are only a couple of such children who have been identified. The emerging hypothesis (see [slide #16](#)) is that many of the children, who were previously thought to have late onset losses, were really children with congenital mild or moderate progressive PCHL. As infants, they had enough hearing that they began to babble, acquire some speech, startle, or turn to loud noises. By the time they were 3, 4, or 5 years old, however, it was clear that their speech was substantially delayed, and as a result, hearing loss was diagnosed. Because they were babbling and turning to noises as an infant, it was assumed that it was an acquired loss, when in fact many of these could have been mild or moderate progressive congenital losses.

ACCURACY OF NEWBORN HEARING SCREENING METHODS

As shown in [slide #17](#), there are at least five different methods that are frequently used for early identification of hearing loss. What do we know about the accuracy of those methods as a tool for universal newborn hearing screening? Even though there are many articles in the literature which use the terms sensitivity and specificity with regard to each of these techniques, there are no studies of universal newborn hearing screening where there are sufficiently large sample sizes and sufficiently good follow up to definitively establish the sensitivity and specificity of any of those techniques. As summarized in [slide #18](#), most studies which refer to sensitivity and specificity have used very small sample sizes, have focused only on high-risk babies, or have not followed all of the babies who passed the screening test to determine their true hearing status. A frequent problem is that studies that have allegedly examined the sensitivity and specificity of a particular screening technique have done that by comparing one screening technique to another screening technique. In other words, instead of comparing otoacoustic emissions in a screening program to behaviorally confirmed hearing loss, a study might compare otoacoustic emissions to conventional ABR. Thus, data are not available to definitively establish the sensitivity and specificity of any of the techniques.

Auditory Brainstem Response (ABR)

We do, however, have other types of information that provide reasonably good evidence about the accuracy of each of these screening techniques. For example, Hyde, Riko, and Malizia evaluated 713 high-risk babies who were screened with auditory brainstem response (ABR) prior to hospital discharge, and then were assessed by uninformed diagnosticians when they were an average of almost four years old. The behaviorally-confirmed hearing status was compared to the results of the hearing screening test. For this high-risk population, the sensitivity and specificity was 98% and 96% when the ABR screening threshold was set at 40 dB HL and 100% and 91% when the ABR screening threshold level was set at 30 dB HL (see [slide #19](#)). Additional data about the accuracy of ABR is shown in [slide #20](#). From a UNHS program at Saint Barnabus Medical Center in New Jersey, over a three-year period with more than 5,000 babies screened each year, an average of 3.3 infants per thousand were identified with a congenital hearing loss and referred into intervention programs. Sedation was not used, the referral rate for further diagnostic testing at the time of hospital discharge was 3.1%, and most screening was done by audiologists within 24 hours of the baby's birth.

High-Risk Indicators

Until just recently, the most frequent method used in the United States to identify hearing loss in very young children was based on the high-risk indicators recommended by the Joint Committee on Infant Hearing (JCIH). About 10% of all children born will exhibit one or more of these

indicators, including family history of congenital hearing loss, very low birth weight, congenital malformations of the head and neck, hyperbilirubinemia, etc. Since 1974, the JCIH has recommended that infants who exhibit one of these risk indicators be screened for hearing loss using auditory brainstem response. The rationale for this approach was that by focusing on a subset of the population which was at higher risk, hospitals would be able to afford to use auditory brainstem response, whereas most people did not believe hospitals could afford to use ABR to screen all infants.

Although such high-risk-based screening programs were the most frequent method used to identify hearing loss in very young children during the late 70's and early 80's, most hospitals are no longer using this approach for two reasons. First, as shown in [slide #21](#), even though children who exhibit one of the risk factors are at higher risk of having a congenital hearing loss, only about half of all children with congenital hearing loss will exhibit one or more of these indicators. Thus, even if a high-risk-based screening program were to work perfectly, about half of all children with PCHL would be missed. Secondly, high-risk-based screening programs found that it was very difficult to get parents to come back for the necessary diagnostic evaluations. For example, in 1987, Mahoney and Eichwald reported the results of the Utah- based high-risk screening program over a seven-year period (1978 to 1984). During that time, the JCIH indicators were incorporated into the legally required birth certificate, so they were reported for every child. The program included computerized mailings and follow up, free diagnostic assessments at regional offices, and/or a mobile van that went to parents' homes. As summarized in [slides #22 and #23](#), only about half the parents whose children had a high-risk indicator ever made an appointment for a diagnostic evaluation, and only about half of those parents completed the evaluation. Only .36 children per thousand were identified with sensorineural hearing loss. Other high-risk indicator-based programs experienced similar or even worse difficulties. Even though complete follow-up data is not available for any of the programs, it is clear, based on the very small number of children identified, that such programs have very low sensitivity; and given the relatively high number of children exhibiting risk factors, such programs also have very low specificity.

Behavioral Evaluations at Seven to Nine Months of Age

Although it is not used very often in the United States, another alternative for early identification of hearing loss is to do behavioral assessments of children when they are 7-9 months old. Such programs are used extensively in Europe and have sometimes been advocated for implementation in the U.S. As they operate in Europe, the screening is done by home visitors who are already making routine visits as a part of the well-child health care system. Because the implementation of such a program in the U.S. would require the creation of a very expensive infrastructure of home visitors, some people suggest that such behavioral evaluations could be done by physicians as a part of well-baby care when babies are 3-9 months of age using techniques similar to what is used in those European countries.

Although often advocated as an alternative for this country, the data on the success of those home-based behavioral screening programs, often called the Home Visitor Distraction Test, is

very disappointing. For example, Peter Watkin and his colleagues ([slide #24](#)) did a retrospective analysis of over 55,000 children in one geographic district in England. For each of the 171 two- to fifteen-year-old children who had a hearing loss, Watkin and his colleagues determined whether the child was first identified through a home visitor or school-age screening program, a parent, or someone else, such as a doctor or teacher. More than a third of the children were missed by both the nine-month and the school-age screening program.

[Slide #25](#) shows the results for just the home-based screening program. Of the 39 children with severe profound bilateral losses, only 44% were identified from the Home Visitor Distraction Test. For children with mild moderate bilateral losses and children with unilateral losses, only 25% and less than 10% were identified with the Home Visitor Distraction Test, respectively. So even with home visitors who were specifically trained to do that type of behavioral assessment in a home setting and were given a great deal of support and monitoring to do it well, most of the children are being missed. If such home visitors are unable to do behavioral screening at nine months of age, it is very unlikely that it can happen as a part of the routine in a busy doctor's office.

Otoacoustic Emissions (OAE)

A fourth newborn hearing screening approach is the use of otoacoustic emissions, either transient evoked or distortion product otoacoustic emissions. Numerous small-scale studies, such as those summarized in [slides #26](#) and [#27](#), have demonstrated that transient evoked otoacoustic emissions (TEOAE) have a very high rate of agreement with auditory brainstem response. The first large-scale evaluation of otoacoustic emissions in a universal newborn hearing screening program was the Rhode Island Hearing Assessment Program (RIHAP) led by Betty Vohr between 1990 and 1994. As shown in [slide #28](#), of the first cohort of 1,850 infants from well-baby and special-care nurseries, 11 were identified with sensorineural hearing loss. Because ABR screening was also done with each of these babies, the study showed that there was very high agreement between TEOAE screening and auditory brainstem response. Furthermore, four of the babies would have been missed if screening had only been done with high-risk infants or with babies in the NICU. Although it is impossible to determine sensitivity since follow up was not done with all of the children who passed, there was a questionnaire study done later, when children were 2-3 years old, to which about 40% of the "passes" responded, and none of the parents believed that their child had a sensorineural loss. If we assume that all of the original passes had normal hearing, then the sensitivity in this study was 100%, and the specificity was 95% ([slide #29](#)).

Automated Auditory Brainstem Response (AABR)

The fifth and final technique for newborn hearing screening considered here is the use of automated ABR. Although several other units are now available, the data in these slides were collected the most widely used AABR equipment produced by Natus Medical. As can be seen in [slides #30](#) and [#31](#), four different studies in which results of the Algo1 were compared to conventional ABR show high levels of sensitivity and specificity for the two techniques. The

combined data are summarized in [slide #32](#). Even though the results do not provide information on the sensitivity for determining hearing loss since behavioral evaluations of all the children were not done, given what we know about the accuracy of conventional ABR, these data provide convincing evidence about the accuracy of automated ABR.

University of Washington Research

One other study that is now underway should be mentioned. Sponsored by the National Institutes of Health and being coordinated by the University of Washington, this is a multi-centered study which is evaluating the effectiveness of newborn hearing screening using conventional ABR, transient evoked otoacoustic emissions, and distortion product otoacoustic emissions. Over 7,000 infants (4,500 NICU babies and 2,600 normal-care nursery babies) were screened prior to discharge using all three of the techniques in random order. Babies were behaviorally assessed at 8-12 months of age. All of the behavioral assessments were completed in October 1997, and final results from the study are expected to be available by April 1998. As summarized in [slide #33](#), these data will provide us with more definitive information than we have had about the true sensitivity and specificity of ABR and evoked otoacoustic emissions. It's important to note, however, that the screening methods used in this study are now five years old, so even though the study will provide important information about the underlying mechanism, the equipment used with each of these techniques is quite different today than at the time data were collected for this study. Parenthetically, it is important to note that newborn hearing screening equipment continues to evolve quite rapidly. Whether ABR or EOAE is used, the equipment is becoming faster and more accurate with each passing year. The last five years have seen dramatic changes in both ABR and EOAE equipment, and it is safe to predict that similar advances will happen during the next five years.

EFFICIENCY OF EXISTING UNHS PROGRAMS

When the NIH Consensus Development Conference recommended that all babies be screened for hearing loss before discharge from the hospital, many people questioned whether it was practical for hospitals to implement such programs. Although the rapid increase in hospital-based programs in the last five years provides clear evidence about the practicality of such programs, many questions remain about the efficiency and effects of such programs (slide #34). Many such issues could be addressed, but only four are discussed briefly here.

The first issue relates to how successful programs have been in screening all babies prior to discharge and what types of referral rates are typically achieved. Slide #35 shows the results of a survey of 120 universal newborn hearing screening programs, all of which used either OAE or ABR as a screening tool. Fifty-five of the 64 OAE-based programs used TEOAE, and 54 of the 56 ABR-based programs used the Natus automated ABR. These data show that the percentage of babies screened prior to discharge is about 95% for both OAE and ABR-based programs. Less than 10% of babies do not pass the screen prior to discharge, with referral rates for ABR about half the referral rates (4% on average) as compared to the referral rate of OAE-based programs (about 8%). These numbers are very different from the percentages published about the original Rhode Island Hearing Assessment Program in which 27% of the infants were referred for additional screening at the time of discharge. With changes in equipment and procedures for doing screening, referral rates have come down dramatically for OAE-based programs. Referral rates for ABR-based programs have also been reduced, even though they were never as high as 25%. With the newest equipment, it is not unusual to have referral rates for ABR-based programs of 2% or 3%.

In spite of the fact that most hospitals are discharging babies at an average of about 24 hours after birth, coupled with frequent reports of referral rates of less than 10% at the time of hospital discharge, there is still a persistent belief that referral rates will be very high for OAE-based programs when babies are less than 24 hours of age. An article by Maxon and her colleagues provides data showing how it is possible to obtain very low referral rates for babies within the first few days of life (see slide #36). The article also makes many concrete suggestions for how to achieve acceptably low referral rates in TEOAE-based newborn hearing screening programs.

Questions have also been raised about whether UNHS programs really do reduce the age at which children with hearing loss are identified. Some people have felt that because of the difficulties in follow up and diagnosis of very young children, we could go to a great deal of work and still not substantially reduce the age of identification. Data addressing this question were reported recently by Parving and Salomon (slides #37 and #38), in which three different five-year cohorts of all births in Copenhagen County beginning in 1970, 1980, and 1990 were analyzed for the age at which a bilateral hearing loss greater than 25 dBHL was identified. For the 1970-74 cohort, no specific procedures were in place for early identification of hearing loss. Beginning in 1975, a

home-based behavioral screening program was used with more than 95% of all the children. From 1990 to 1994, an OAE newborn hearing screening program was implemented for approximately 20% of all of the births. There were approximately 37,000 births in each five-year cohort, and procedures for diagnosis and follow up were essentially the same for all of the cohorts. As shown in slide #38, the average age at which children with hearing loss were identified dropped substantially for each of the cohorts. Even though universal newborn hearing screening had not been implemented county-wide in 1990, more than 60% of the children with hearing loss were identified before 12 months of age. The prevalence of hearing loss in each of the three cohorts was about the same. The data from Parving and Salomon provide good evidence that hospital-based universal newborn hearing screening programs will substantially reduce the age of identification compared to a home-based behavioral screening program.

A third issue related to the efficiency of newborn hearing screening programs is whether such programs create unacceptable levels of parent anxiety or disruption of family functioning. Ellen Clayton has summarized some of the possible negative consequences of different screening outcomes (slide #39). To assess how frequently such problems occur, Tluczek et al. (1992) gave questionnaires to 104 parents of children who had failed a screening test for cystic fibrosis. A similar study was done by Uzcategui (1997) of 171 parents of children who failed the initial newborn hearing screening test (slide #40). In both cases, a Likert-type scale was used to assess parent feelings on a number of dimensions related to anxiety. Slide #41 shows the results from both studies. Although the level of concern/fear, shock, anger, and confusion are substantially less for the newborn hearing screening sample than for the cystic fibrosis sample, it is clear that issues related to parent anxiety need further investigation. Other results from the Tluczek study emphasized the need to make sure parents are appropriately informed about the intent to do screening and the results of the screening process (slide #42).

A study by Barringer and Mauk (slide #43) showed that virtually all parents would give their permission to have their baby screened if they were asked, and the majority would be willing to pay for it out of their own pocket. Almost 90% said that any anxiety caused by the baby not passing the initial screen would be outweighed by the benefits of early detection if hearing loss was found to be present.

Similar results were found by Watkin and his colleagues (1995) when they surveyed 208 parents of children with sensorineural loss (slide #44). The majority wished their child had been identified earlier; 89% preferred having a newborn hearing screening program instead of what they had; and most were dissatisfied with the age at which their child's hearing loss was identified.

A final issue related to the efficiency of newborn hearing screening programs has to do with tracking babies who are referred from the initial screen through the diagnostic process and making sure they receive appropriate early intervention. Most operational programs identify tracking and follow up as the biggest challenge related to early identification of hearing loss. As shown in slide #45, many programs are unable to obtain conclusive diagnostic information on as many as half of the children who failed the screening process. Not surprisingly, programs with the highest prevalence rate are those that are most successful at following children through to a conclusive diagnosis. Clearly, finding ways to keep better track of children until a diagnosis is obtained is one

of the most important challenges that needs to be addressed by programs whose goal is early identification of hearing loss. Screening itself has proven to be relatively easy, but completing the process through diagnosis and appropriate early intervention remains a substantial challenge. It appears that how successful we are at tracking children through the diagnostic process is partly a function of geographical and socio-economic circumstances. Clearly, programs that don't work hard at keeping track of children will not be successful. However, many programs that have been working very hard still lose track of a substantial number of children.

These data emphasized the need for better tracking and follow-up systems and integrating those tracking and follow-up systems with other public health information databases. It is also important to point out, however, that even in those programs which are having difficulty tracking the majority of children through a conclusive diagnosis, the prevalence of children with hearing loss being identified is substantially higher than has historically been the case. Thus, it is clear that universal newborn hearing screening programs are efficient in identifying children with hearing loss at an early age, but improvements are still necessary.

CONSEQUENCES OF NEONATAL HEARING LOSS

Are the consequences of neonatal hearing loss serious enough that we need to be concerned about them in the first place? Fortunately, there is a wealth of data on this question, and only three very brief examples will be given related to children with severe profound bilateral losses, children with mild bilateral and unilateral losses, and children with fluctuating conductive losses ([slide #46](#)).

[Slide #47](#) shows data from an annual survey done each year by Gallaudet University showing that children with severe profound bilateral losses suffer from substantial deficits in reading comprehension. When these children are 8 years of age, they are already almost 1-1/2 years behind their peers. That gap continues to widen over time, with the average deaf child or youth never exceeding a grade equivalent of 3 years, in spite of the fact that most of them are enrolled in educational programs specifically designed for deaf students. Of course, most of these children did not have the benefit of very early identification.

Although virtually everyone agrees that severe profound bilateral hearing loss has substantial negative consequences for all aspects of academic performance, the consequences of mild bilateral or unilateral hearing loss are less well known. There are, however, a number of studies demonstrating that even mild or unilateral sensorineural hearing loss has substantial negative consequences. [Slide #48](#) summarizes information about five different students in which a sample of children with unilateral hearing loss were compared to similar children with normal hearing, and measures were taken about their performance in math, language, and social functioning. As shown in [slide #49](#), children with unilateral hearing loss lagged substantially behind their peers regardless of the measure used, whether math, language, or social functioning. Even though the sample sizes are quite small in each of the studies, matching was generally done quite well and the results are very consistent. For a 10-year-old child, these results translate into a deficit of approximately 1-1/2 years in math or reading achievement.

Even mild fluctuating conductive hearing loss can have a substantial effect over time. Although virtually all newborn hearing screening programs focus only on identifying children with permanent congenital hearing loss, some children with fluctuating conductive losses are often identified. The reason for presenting this information is to emphasize the point that even mild fluctuating hearing losses have an important negative effect on developmental outcomes. How much more important it is then to identify children with permanent hearing losses.

The data in [slide #50](#) come from the Greater Boston Otitis Media Study Group, where 194 children were followed prospectively for 7 years. Each time the child visited his or her primary-care physician, data were collected about episodes of otitis media (there were an average of 7 visits per year during the birth to 3-year-old period). Data were also collected periodically on a wide variety of measures related to intellectual ability, cognitive functioning, and language competency by trained diagnosticians who were uninformed about the purposes of the study.

Results at 7 years of age for two groups of children are reported here: those with less than 30 days of otitis media over the 3-year period compared to those with 130 or more days of otitis media over the same period. After statistically adjusting the results for a variety of demographic and socio-economic status variables, children with fewer episodes of otitis media did substantially better than their peers on all measures. The difference averaged about one-half standard deviation, which is equivalent to more than a year's worth of development in reading or math achievement.

BENEFITS RESULTING FROM EARLIER IDENTIFICATION AND INTERVENTION

Having established that there are serious negative consequences associated with all types and severities of hearing loss, a natural question is whether there are significant benefits to the child if hearing loss is identified early and appropriate intervention is begun. It is important to note that there have not been prospective randomized clinical trials addressing that issue ([slide #51](#)). Most of the evidence we have can be criticized or is weakened by the fact that there is potential for selection bias, most studies do not include long-term follow up, sample sizes are generally quite small, and in some of the studies the types of outcomes assessed were somewhat subjective.

In spite of those weaknesses, there is a fair amount of information showing that there are benefits associated with earlier identification and intervention. For example, a study reported by Yoshinaga-Itano and her colleagues in 1996 ([slide #52](#)) evaluated the language abilities of 46 children with bilateral hearing loss identified before 6 months of age, with 63 similar children identified after 6 months of age. Language abilities were measured by parent report using a cross-sectional assessment design in which children were categorized into four different age groups. As can be seen in [slide #53](#), the 23 children assessed when they were 13-18 months old already showed an advantage for the earlier identified group. This advantage for the earlier identified group becomes larger for the 28 children assessed when they were 19-24 months of age, larger still for the 31 children assessed when they were 25-30 months of age, and even larger for the 27 children assessed when they were 31-36 months of age. As shown in [slide #54](#) and [slide #55](#), the results are similar for measures of expressive language and vocabulary.

Apuzzo and Yoshinaga-Itano reported a similar study in 1995 ([slide #56](#)) in which 69 children with hearing impairment were categorized into four groups according to the age of identification. Groups were reasonably similar with respect to age at the time of testing, degree of hearing loss, and level of development. All of the children were participating in similar early intervention programs, but had begun at different ages. Outcome data are based on parent report using the Minnesota Child Development Inventory, and results shown in [slide #57](#) are based on covariance adjustments for degree of hearing loss and cognitive ability. As can be seen, the 14 children identified earliest are functioning almost at grade level, while those identified latest (25+ months) are functioning at substantially lower levels.

Another study conducted by Sue Watkins at Utah State University ([slide #59](#)) provides similar information, but the design is stronger because all of the children were assessed at the same time, sample sizes are larger for each group, and there was more extensive matching and statistical adjustment for potential confounding variables. In this study, there were three groups of 23 children who had been matched or the scores were statistically adjusted for a variety of variables, including severity of hearing loss, age, presence of other handicap, age of mother, SES indicators, and number of childhood middle ear infections. The first group had received an average of nine

months of home intervention before 30 months of age, and then received preschool intervention until they were enrolled in public school. The second group of children received no home intervention, but began a preschool intervention program at 36 months of age. The third group received no home intervention or preschool intervention.

A wide variety of measures were collected by trained diagnosticians who were unaware of the group to which the children belonged. As shown in [slide #59](#), it is clear that those who received both home-based intervention and preschool intervention did substantially better than those who received only preschool intervention or did not receive any intervention prior to beginning public school. The effects for reading, arithmetic, vocabulary, articulation, percent of the child's communication understood by non-family members, percent of non-family communication understood by the child, social adjustment, and behavior shows that children who received the most intensive early intervention perform 20-45 percentile points higher than children who do not receive such intervention. The results of this study are particularly convincing because:

- all of the children received the same types of intervention from the same providers, except that they were enrolled in intervention programs at different ages;
- the diagnosticians were unaware of the group to which children belonged;
- matching and/or statistical adjustment was done on a wide variety of variables; and
- measures covered various domains and were collected when children were 10 years old.

Even though results of randomized clinical trials are not available to address the question of whether earlier intervention is better than later intervention for children with hearing loss, the consistency of findings from a number of quasi-experimental studies provides consistent and convincing evidence about the benefits of earlier intervention. It is important to note that the type of intervention children need is dependent on the type and severity of hearing loss ([slide #60](#)). Because it has only been in recent years that we have identified children with unilateral and mild bilateral losses at less than one year of age, we do not have a great deal of experience about how to provide the most effective intervention to these children. Over the next several years, as we gain more experience in providing intervention to children in these groups, we will learn more about how to deliver intervention most effectively. As more and more children are identified at earlier ages, it is also expected that more data about the benefits of early intervention will become available.

COST-EFFICIENCY OF HOSPITAL-BASED UNHS PROGRAMS

Although everyone talks about the importance of doing cost analyses related to early hearing detection and intervention, terms are frequently misused. It is important to define what we mean by some of the most frequently used terms. As shown in [slide #61](#), three distinctions are important. First, it is important to determine how much early hearing detection and intervention programs cost completely independent of the effects of such programs. Methodologies for estimating costs are well developed and relatively straight forward to apply.

Cost effectiveness is a second type of cost analysis. By definition, the cost effectiveness of a program can only be determined if one program is being compared to another. It is inappropriate to say that program A is cost effective. Instead, we must evaluate whether program A is more cost effective than program B. In other words, if we examine both the costs and effects of two different programs, which program yields the most effects for every unit of cost? In a cost-effectiveness comparison of two programs, the costs of each program are analyzed in the same way as the first type of cost analysis described above. However, the effects of each program are also calculated according to variables like the number of children per thousand identified, the age at which those children are fitted with amplification, scores on various measures of developmental functioning, etc. The results of a cost-effectiveness analysis might conclude that the cost of identifying a child with hearing loss in program A is \$7,294, while the cost of identifying a child with hearing loss in program B is \$8,492. In this hypothetical example, program A would be more cost effective than program B.

A cost-benefit study can be either comparative or applied to a single program. Costs are computed in exactly the same way as in the first two types of cost analyses. However, in this case, the benefits associated with newborn hearing detection and intervention must be translated into monetary values. A program is said to be cost beneficial if the amount of money spent on the program is less than the monetary worth of the benefits resulting from that program. As you can imagine, good cost-benefit studies are very, very difficult to do, because of the difficulty of assigning monetary value to outcomes such as a year and a half worth of reading gain or the monetary value to a parent of being able to communicate more effectively with their child.

A number of different kinds of cost studies related to early hearing detection and intervention have been reported in the literature. Most of these are either sample cost estimates or cost-effectiveness analyses, and most suffer from serious weaknesses. For example, in most studies the estimates of costs are based on hypothetical assumptions or unverified self-reports. Such cost estimates are often incomplete in that they ignore costs, such as fringe benefits, indirect costs, and costs to parents. Other studies only include a part of the detection and intervention process (e.g., costs for follow up and tracking may be excluded), and standard economic analysis concepts, such as discounting, sensitivity analysis, and robustness estimates, are frequently not used.

The only area where we have some credible data is related to the costs of newborn hearing screening. One of the first such studies was done with the Rhode Island Hearing Assessment Program, in which a complete cost analysis was done using actual expenditures, instead of self-report data, for the entire screening and tracking process, but diagnosis and intervention costs were explicitly not included. As shown in [slide #63](#), the cost of a two-stage screening, including scheduling and tracking the babies into a diagnostic evaluation, but not including the cost of the diagnosis itself, was approximately \$26 per baby. This did not include the cost of parents' time for participating in the screening activities.

A similar study was done at Logan Regional Hospital as reported by Weirather and her colleagues and shown in [slide #64](#). Because of the way this program was organized, the cost per baby was substantially cheaper (\$7.42 per baby), even though the analysis included all of the same activities and was just as complete as the RIHAP study.

Recently, CDC, under the direction of Scott Grosse, did a multi-center study in which the costs of newborn hearing screening were estimated in six different centers in six states ([slide #65](#)). Three of those centers were AABR-based sites and three were otoacoustic-emission-based sites. Cost estimates were based on self-report questionnaires, and site visits were made to four of the six sites to gather confirmatory data wherever possible. Although the actual time devoted to various activities was not tracked, as was the case in the studies done in Rhode Island and Utah, the analysis did use standardized techniques for including the costs of fringe benefits, equipment, supplies, and overhead. As shown in [slide #66](#), the cost per baby ranged from about \$18 to \$26, with TEOAE sites being less expensive.

A final study of costs of screening was reported by John Stevens and his colleagues ([slide #67](#)) for ten different hearing screening programs in Great Britain. Five of these programs did targeted screening with high-risk infants, three were universal newborn hearing screening programs, and two were home-visitor programs. Results ranged from an average of about \$8 per baby born for the high-risk programs to \$22 per baby for the universal newborn hearing screening programs to \$32 per baby for the home-visitor programs. Although not as much detail about the procedures used in the cost analysis were included in this report, the results are quite similar to what has been found by studies done in the U.S. It is also important to note that in the ten programs evaluated by Stevens, there was reasonable consistency within the various types of programs, which makes the results more believable.

It is possible to do some very rough estimates of some of the more obvious monetary benefits associated with newborn hearing detection and intervention programs by using information from other sources about the prevalence and costs of screening for various diseases among newborns. As shown in [slide #68](#), the prevalence of permanent congenital hearing loss at 3 per thousand is substantially higher than the prevalence of PKU, hyperthyroidism, or sickle cell anemia, which are required for screening in every state. All three of those diseases are screened for using the same blood test, which for purposes of this comparison was estimated to cost \$25 per test. Because the prevalence of those other diseases is so low, the cost for identifying a child with any one of those diseases is approximately \$41,000 per child, compared to a cost of \$8,683 to identify a child with permanent congenital hearing loss (PCHL).

Is it worth it to spend almost \$9,000 to identify a child with PCHL during the first few months of life? Based on data presented about the benefits of early identification, it appears that children identified early will have better cognitive skills, social skills, and language skills, which are all the types of things that contribute to the child being capable of being educated in a regular mainstream classroom or a self-contained classroom, instead of a residential program. In other words, the data we have suggests that if children are identified early, they will be capable of being appropriately educated in a less restrictive and less expensive environment. (Note that these data should not be used to suggest that states could save money by inappropriately moving children from more restrictive to less restrictive environments. All educational systems must be available for the child, and the most appropriate setting should be determined based on the capabilities of the child and the preferences of the parent and child.) As shown in [slide #70](#), the U.S. Department of Education estimates that for hearing-impaired children, the annual costs of education in a regular mainstream classroom in 1990 was \$3,383, while the annual costs for a hearing-impaired child in a self-contained classroom or residential placement was \$9,689 and \$35,780, respectively. Thus, over the educational lifetime of a child, substantial amounts of money would be saved if, as a result of early identification and intervention, the most appropriate educational setting for the child is a regular mainstream classroom instead of a self-contained classroom or a self-contained classroom instead of a residential program. In fact, if only 2% of the children identified with a hearing loss were educated in a self-contained classroom instead of a residential program, it would more than pay for the cost of the newborn hearing screening program in which all of the children were identified. Although there have not been empirical studies of the number of children who would be more appropriately educated in less restrictive environments as a result of newborn hearing screening programs, based on the data about the benefits of early intervention and the costs of early identification programs, it is very plausible to expect many early identified children to be educated in less restrictive environments. As a result, at least this much money, and probably much more, would be saved.

SUMMARY

For each of the issues addressed during this presentation, a summary of the available evidence is given below. Following that summary, the limitations of the existing evidence are described, and references cited in the presentation, along with other references relevant to that issue, are listed. Copies of the slides can be downloaded or information from this presentation can be used in making presentations or informing people about issues related to the implementation of early hearing detection and intervention programs. Information included here, which has not previously been published, should not be cited or used in other publications without written permission from the authors of that information.

Summaries:

- Prevalence of congenital hearing loss
 - Accuracy of newborn hearing screening methods
 - Efficiency of existing UNHS programs
 - Consequences of neonatal hearing loss
 - Benefits resulting from earlier identification and intervention
 - Cost-efficiency of hospital-based UNHS programs
-

Prevalence of Congenital Hearing Loss

A key issue in deciding whether universal newborn hearing screening programs should be recommended is how many children have permanent congenital hearing loss (PCHL). Existing data related to this issue must be interpreted with respect to how PCHL was defined (the severity of the loss [ranging from 20 dBHL to 50 dBHL; single frequency loss vs. an average across several frequencies], whether unilateral or only bilateral losses were included, the technique used to make a determination of PCHL [diagnostic ABR vs. behaviorally-confirmed losses], and what efforts were made to account for PCHL acquired after infancy). Obviously, how individual studies address each of these issues will have a dramatic effect on the reported prevalence rate. Retrospective population-based studies with large samples from around the world consistently show bilateral PCHL from about one per thousand (for PTA losses > 50 dBHL) to three per thousand (for PTA bilateral losses > 20 dBHL). Because none of these studies differentiated between acquired losses and congenital losses, they over-estimate congenital PCHL to some unknown degree (as noted below however, this over-estimate is probably very small). There is also substantial evidence that including unilateral PCHL would increase these estimates by about 40%.

Data from universal newborn hearing screening programs which have operated since the early 1990's provides similar information about the prevalence of congenital hearing loss. Data from these programs must be interpreted in conjunction with issues such as how a determination was made that a child had a congenital hearing loss (behavioral diagnosis or diagnostic ABR), whether

screening was designed to find unilateral or bilateral losses, and the number of infants lost to follow up during the screening and diagnosis process (some programs have not been able to follow as many as 60% of the children initially referred from the screening program). Those programs with large numbers of children who have been successful in following most of the refers, and who have screened for unilateral and mild or worse PCHL, report prevalence rates of three to four per thousand. Programs that have been operational for 5+ years and who have monitored the identification of children with PCHL at older ages report finding very few, if any, acquired hearing losses. The absence of children with acquired losses in those places where universal screening programs have been operational for 5+ years suggests that much of what was previously believed to be acquired hearing loss was probably congenital mild or moderate loss that was progressive.

Taken together, the evidence is quite convincing that congenital PCHL > 25 dBHL in the worst ear is present for at least three infants per thousand.

Limitations of Existing Evidence

Evidence concerning prevalence comes from studies around the world and is based on large cohorts of children. It is reasonably consistent after accounting for issues related to definition, diagnostic procedures, and completeness of follow up. Weaknesses with these data include: a) the methods for diagnosing PCHL are often not adequately described; and b) no definitive methods of accounting for how many children in these data sets have acquired hearing loss. Anecdotal evidence suggests that the percentage of PCHL children with acquired loss is much lower than has historically been assumed.

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Accuracy of Newborn Hearing Screening Methods

Five different techniques are used or frequently suggested for early identification of congenital PCHL: 1) auditory brainstem response (ABR), 2) high-risk indicators followed by ABR, 3) behavioral evaluation when children are 7-9 months old, 4) otoacoustic emissions (OAE), and 5) automated ABR (AABR). All except for behavioral evaluation are used at different hospitals in the United States. Behavioral evaluations of 7- to 9-month-old children are used extensively in other parts of the world. Although many articles report figures for the sensitivity and specificity of these various methods, definitive information about the sensitivity compared to behaviorally confirmed congenital PCHL is not available for any of the methods, with the possible exception of ABR. The reason available data is not definitive is that most studies reporting sensitivity figures have either assumed that all children who pass the initial screen had normal hearing, have only followed a small percentage of the children who passed the initial screen, have compared one screening technique to another screening technique to derive sensitivity figures, or were based on unacceptably small samples or samples of only high-risk babies. There is, however, substantial evidence for the accuracy of some of the techniques as described briefly below.

Accuracy of Auditory Brainstem Response for Identification of Congenital PCHL

There is good evidence that Auditory Brainstem Response (ABR) is effective in identifying PCHL in newborns as long as it is used appropriately. Reports of sensitivity for identifying hearing loss are very high (97%-100%), but most are based on small samples of high-risk infants in universal newborn hearing screening programs. Because of the expense and time involved in doing ABR, there are only two known programs in the U.S. who use it as a universal newborn hearing screening technique. There are many programs, however, who screen high-risk infants using ABR.

High-Risk Followed by ABR

Until recently, the most frequent technique for early identification of congenital PCHL was to identify children who exhibited one of the high-risk indicators specified by the Joint Committee on Infant Hearing or to target all children in the neonatal intensive care unit (since a large percentage of these will have one of the JCIH high-risk indicators), and to screen those children with ABR. By targeting screening to this approximately 10% subset of the population, the goal was to keep the cost of screening lower and still identify most of the children. However, because approximately half of children with congenital PCHL do not exhibit any of the risk indicators, many children are missed using this technique. Furthermore, programs have experienced substantial difficulty getting families of children with high-risk indicators to come back for diagnostic evaluations, and consequently, the sensitivity of such programs is very low. If the presence of a high-risk indicator is considered to be a positive screen, the specificity is also very low. With the advent of new screening techniques (OAE and AABR), many high-risk-based early detection programs have been discontinued.

Behavioral Evaluation at 7-9 Months

Although such programs are used primarily in countries where a home health visitor program is already in place, it is sometimes suggested as an alternative here in the U.S. It is also suggested that such evaluations could be done as a part of well-baby care at the doctor's office. Data from well-established programs in other countries which rely on behavioral evaluations at 7-9 months indicate unacceptably low sensitivity. Programs report missing more than half of the children with bilateral sensorineural hearing loss and even higher numbers of children with mild moderate and unilateral losses.

Otoacoustic Emissions

Reported agreement between OAE and ABR testing is high, but there are no studies in which the sensitivity and specificity related to behaviorally confirmed hearing loss can be determined. Early reports of OAE-based newborn hearing screening programs had unacceptably high referral rates for inpatient screening (25%+). Numerous, more recent reports, however, report inpatient referral rates of 5%-8%. There are now dozens of hospital-based universal newborn hearing screening programs using otoacoustic emissions. These programs report identifying 1.5 to 4 PCHL children per thousand (programs unable to track large percentages of referred children are at the lower end of that range, and programs with the most successful follow-up are near the upper range).

Automated ABR

Dozens of hospital-based universal newborn hearing screening programs are using automated ABR to identify children with congenital PCHL. Although there are no data on the sensitivity and specificity of this technique with behaviorally confirmed PCHL, the vast majority of reports which have examined the agreement between AABR and ABR find very high agreement (sensitivity and specificity greater than 95%). AABR-based programs report identifying 2-4 PCHL children per thousand with referral rates of 2%-8% at discharge.

Limitations of Data on Accuracy

Data related to the problems with high-risk indicators and behavioral evaluation of 7- to 9-month-old children are very convincing. The lack of data on sensitivity with behaviorally confirmed PCHL for ABR, OAE, and AABR is concerning, but understandable given the high expense of following all children who pass the initial screening with behavioral evaluations and the fact that other data provide good evidence that most children with congenital PCHL are being identified in these screening programs. Rigorous studies of sensitivity/specificity for universal newborn hearing screening programs using these techniques would certainly be useful, but very expensive to conduct.

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Efficiency of Existing Universal Newborn Hearing Screening (UNHS) Programs

Although hundreds of hospital-based UNHS programs are now operating in the U.S., there are a number of issues related to the operation of those programs which should be considered in deciding whether to recommend the implementation of such programs for all children born in the U.S.

Coverage and Referral Rates

Questions are often raised about whether it is practicable to conduct a universal newborn hearing screening program in a busy hospital environment with relatively short stays. In addition, the relatively high referral rates reported for some early UNHS programs raised concerns about whether universal screening was feasible. More recent data demonstrates that most hospitals with UNHS programs (regardless of the technology) are able to screen 95%+ of all newborns prior to discharge. Referral rates are under 10% in most cases, with the lowest rates coming from hospitals using both OAE and AABR in a two-stage, inpatient screening protocol. The fact that so many different protocols is being used is significant (see Table 1).

Effects of UNHS on Age of Identification

There is clear evidence that the implementation of UNHS substantially lowers the age at which children with congenital PCHL are identified.

Table 1
SCREENING PROTOCOLS
Protocols Used in
Universal Newborn Hearing Screening Programs^a

Screening Procedure	Before Hospital Discharge	After Hospital Discharge ^b	Number of Programs
	TEOAE	TEOAE and ABR ^c	44
	DPOAE	DPOAE	5
	AABR	AABR	24
	AABR	TEOAE	2
	TEOAE and ABR ^c	_____	5
	TEOAE and AABR ^c	_____	2
	DPOAE and ABR ^c	_____	1
	TEOAE	_____	4
	DPOAE	_____	3
	AABR	_____	28
	ABR	_____	2

Effects on Parents

There is substantial literature showing that certain types of newborn screening programs (e.g., screening for cystic fibrosis) can be disruptive and anxiety provoking for parents. Although there is limited evidence on this question for newborn hearing screening programs, the available evidence suggests that most parents are supportive of newborn hearing screening programs. There are feelings of confusion, shock, anger, and concern among a few parents who have a normal-hearing infant who does not pass the initial screen, but these feelings are not nearly so frequent or as strong as they are in other newborn screening programs for conditions such as cystic fibrosis. Whether false positives in newborn hearing screening result in long-term negative effects for parents or for the relationship between parents and the infant has not been adequately investigated.

Difficulties with Follow-up

Many universal newborn hearing screening programs are experiencing difficulty getting all or most of the babies referred from the screening program to complete the diagnostic process and be enrolled in intervention. Attrition rates of as high as 60% between the initial referral and a diagnostic confirmation are not unusual. Such high attrition rates are a problem for UNHS programs regardless of the type of equipment (OAE- or ABR-based) or protocol (one- or two-stage initial screen) used. Even though many more infants with PCHL are being identified as a result of UNHS programs, attrition during the follow-up and diagnostic process is a significant problem.

Limitations of Existing Data

Evidence for the feasibility and practicality of UNHS is quite strong, but evidence concerning secondary effects for parents and family members is only beginning to emerge and is based on small samples or anecdotal reports from operational programs. It is clear that many programs are experiencing difficulties with attrition during the follow-up and diagnostic process, but the factors associated with or contributing to these difficulties are not well documented, nor has there been much work in developing and evaluating solutions.

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Consequences of Neonatal Hearing Loss

There is substantial and convincing evidence that moderate or worse bilateral PCHL has serious negative consequences for children's development, success in school, and later success in life. In recent years, there has been increasing evidence that mild bilateral, unilateral, and fluctuating conductive losses can also have serious negative consequences for children. The preponderance of the evidence indicates that hearing loss of any severity or type has negative consequences for children's development.

Limitations of the Data

The most difficult aspect of estimating the consequences of hearing loss is controlling for all of the confounding variables that may affect the outcomes between groups of hearing-impaired children and groups of normal-hearing children.

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Benefits of Earlier Identification and Intervention

A key question in deciding whether to recommend universal newborn hearing screening is whether children who are identified earlier and given intervention do better than those who are identified later. For obvious reasons, it is not practical to identify hearing-impaired children early and randomly assign them to receive intervention or not to receive intervention. Thus, there are no prospective clinical trials which can be used to address this question. However, there are a number of retrospective studies in which children have been categorized into groups who were identified early or identified later, matched on relevant variables, and assessed on developmental outcomes and success in school-related areas. There are also a number of studies in which the correlation between age of identification and developmental outcomes have been assessed.

Data from these studies consistently show that children who are identified early and given intervention (including amplification, as well as educational intervention) do better than children who are identified later.

Limitations of Available Evidence

Studies which have addressed this question are generally based on small sample sizes in which a group of early identified children are matched with children who are identified later. In such studies, children with more severe hearing losses are likely to be identified early, and late-identified children are more likely to be included if they have more severe language deficits. Most of the studies do not follow the children into the elementary school years. The nature of the early

intervention program provided is often not clearly described, and outcome measures are not as strong as desirable.

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Cost Efficiency of Newborn Hearing Screening

A number of cost-related studies have been done about newborn hearing screening. A few are cost-effectiveness studies in which the costs of two alternative screening protocols are computed and compared to the effects of each protocol. Most are cost estimates in which the costs of a particular protocol are computed without trying to compare that protocol to another. A third type of cost study, cost-benefit analysis, in which the outcomes of a screening program are assessed and assigned a monetary value and compared to the costs of the program, has not been done. Although people frequently ask whether newborn hearing screening is cost beneficial, such studies do not exist.

Although there are several published articles about the costs of newborn hearing screening programs, most of these report results of analyses with outdated technology, are based primarily on hypothetical assumptions about costs and time necessary for various procedures, are incomplete economic analyses, or only consider some of the components or activities associated with early hearing detection and intervention. In spite of these deficiencies, those studies do provide some general information about the costs associated with newborn hearing screening. There are no cost studies which have been done well enough to draw conclusions about the cost effectiveness of different protocols or the cost benefit associated with universal newborn hearing screening.

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Universal Newborn Hearing Screening Issues and Evidence*

by

Karl R. White, Ph.D.

**CDC Workshop on Early Hearing Detection and
Intervention
Atlanta, Georgia**

October 22-23, 1997

***Some of the data included in this presentation is unpublished and should not be cited or used without permission of the authors.**



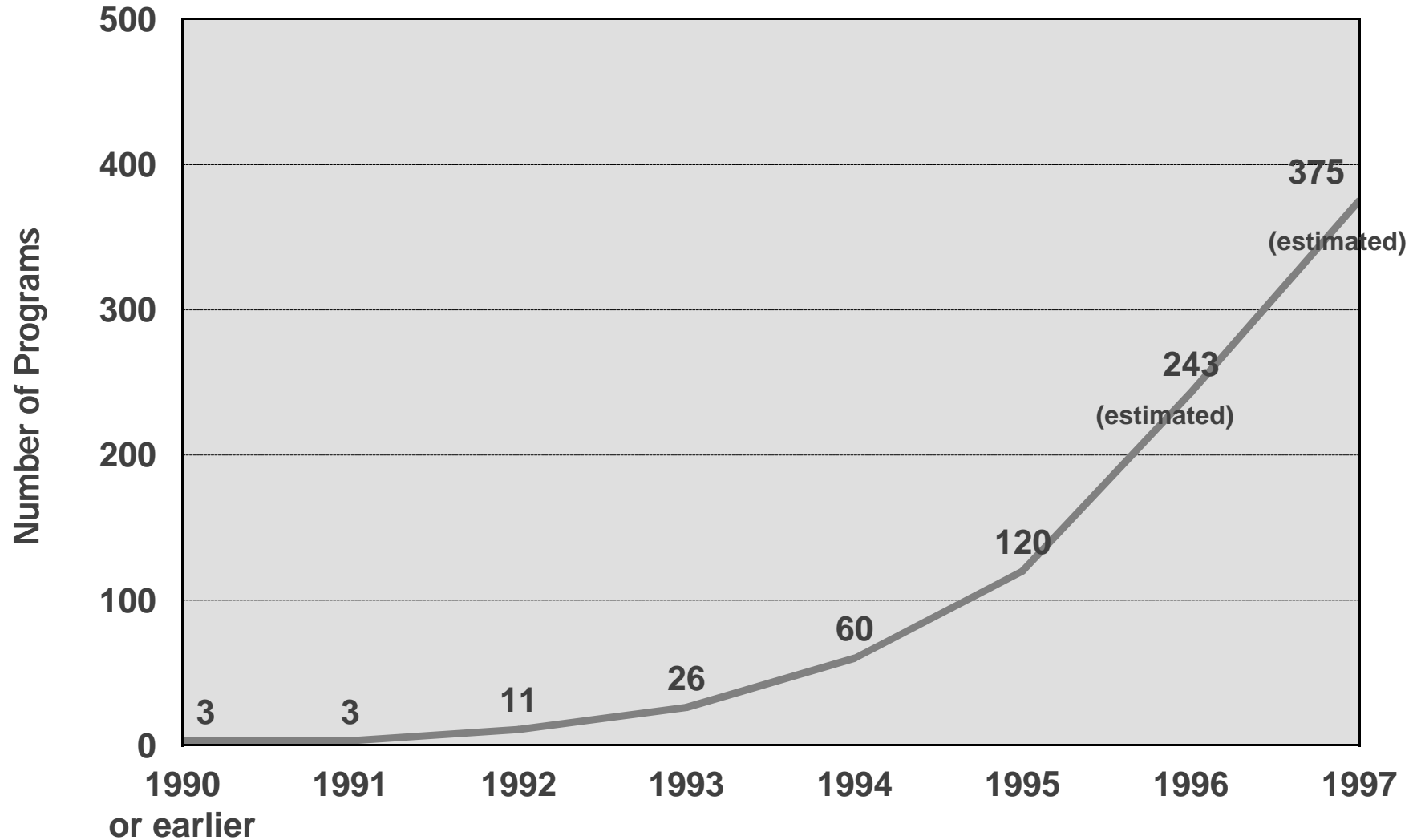


Slide 2



Slide 3

Rate of UNHS Implementation is Accelerating



Updated from: White, K. R., Mauk, G. W., Culpepper, N. B., & Weirather, Y. (1998). Newborn hearing screening in the United States:

Is it becoming the standard of care? In L. Spivak (Ed.), Universal newborn hearing screening (pp. 225-255). Thieme: New York.

Statements Endorsing Early Detection of Hearing Loss

The Department of Education, in collaboration with the Department of Health and Human Services, should issue federal guidelines to assist states in implementing improved [hearing] screening procedures for each live birth.

Commission on Education of the Deaf, 1988

Reduce the average age at which children with significant hearing impairment are identified to no more than 12 months.

Healthy People 2000 Report, 1990

All hearing impaired infants should be identified and treatment initiated by 6 months of age. In order to achieve this ... the consensus panel recommends screening of *all* newborns ... for hearing impairment prior to hospital discharge.

NIH Consensus Statement, 1993

The 1994 [JCIH] Position Statement endorses the goal of universal detection of infants with hearing loss ... [and] recommends the option of evaluating infants before discharge from the newborn nursery.

Joint Committee on Infant Hearing, 1994

States with Legislative Mandates

Rhode Island, 1992

Hawaii, 1990

Colorado, 1997

Connecticut, 1997

Mississippi, 1997

States Currently Considering Legislative Mandates

Massachusetts

Minnesota

New York

Oregon

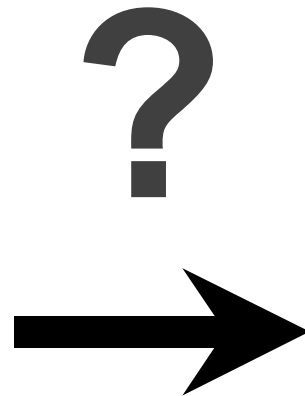
Pennsylvania

Utah

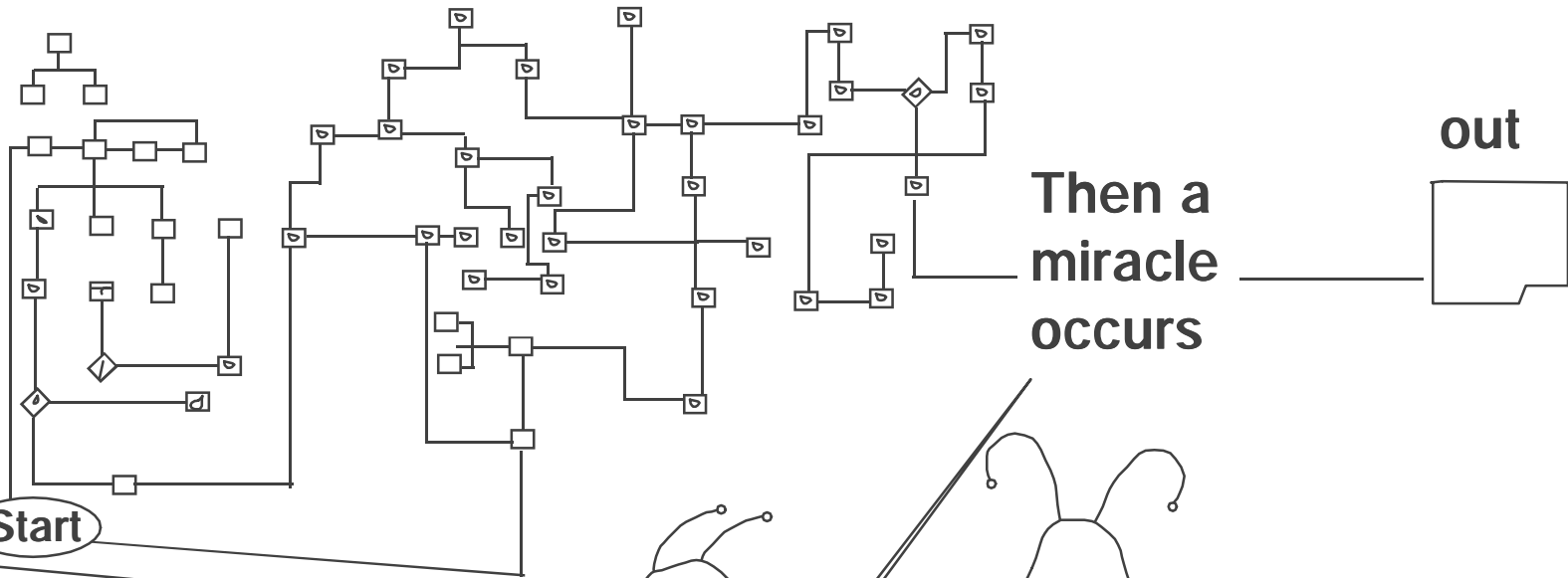
West Virginia

California

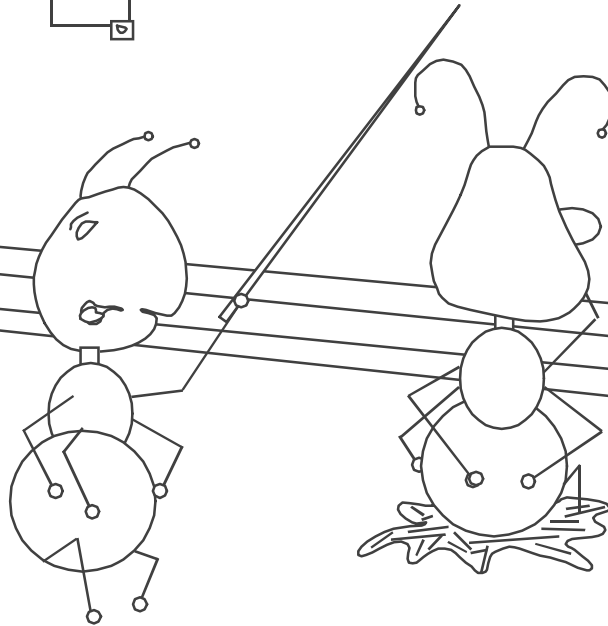
The Solution?



Implementing Universal Newborn Hearing Screening Programs



Good work,
but I think we might
need just a little more
detail right here.



U.S. Preventive Services Task Force (1996)

"While congenital hearing loss is a serious health problem associated with developmental delay in speech and language function, there is little evidence to support the use of routine, universal screening for all neonates."

Because:

- ✓ **a substantial number of infants will be misclassified because the prevalence of hearing impairment is low.**
- ✓ **screening technology is evolving.**
- ✓ **costs and feasibility for universal application are not fully known.**
- ✓ **most importantly, the evidence for efficacy of early intervention is incomplete.**

Purpose of Presentation

- ✓ **Identify key issues related to early hearing detection.**
- ✓ **Present a context and framework to facilitate discussion about those issues.**
- ✓ **Summarize enough of the available evidence to establish a "point of departure" for the group's work.**
- ✓ **Generate discussion about the areas in which important evidence is inadequate.**
- ✓ **Stimulate discussion about how to address gaps in evidence.**

What Are the Issues?

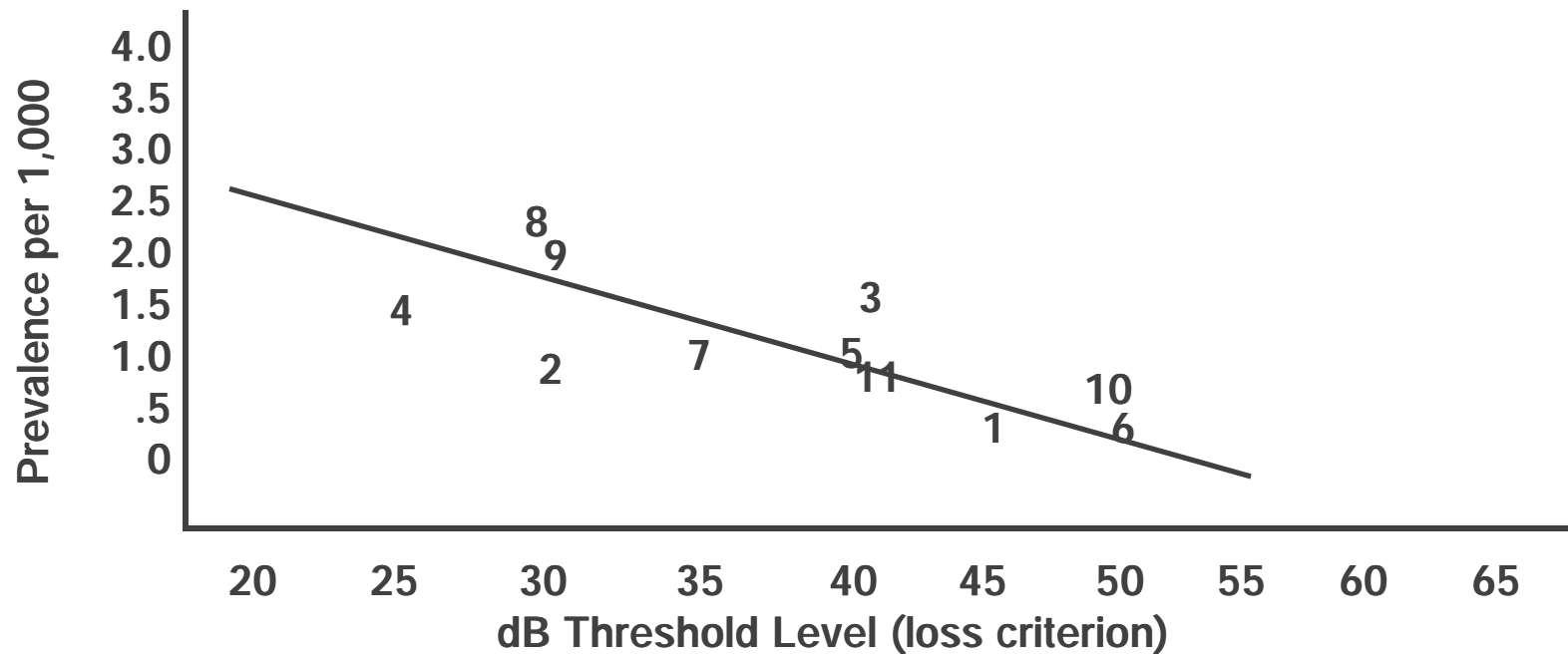
- I. **Prevalence of Congenital Hearing Loss**
- II. **Accuracy of Newborn Hearing Screening Methods**
- III. **Efficiency of Existing Early Detection Programs**
- IV. **Consequences of Neonatal Hearing Loss**
- V. **Benefits Resulting From Earlier Identification and Intervention**
- VI. **Cost Efficiency of Newborn Hearing Screening**

List of Issues modified from 1996 Guide to Clinical Preventive Services, U.S. Preventive Services Task Force

I. Prevalence

- ✓ **Definitional Issues**
- ✓ **Population-based Studies**
- ✓ **Results from Operational Screening Programs**
- ✓ **Late Onset Losses**

Reported Prevalence Rates of Bilateral Permanent Childhood Hearing Loss (PCHL) in Population-based Studies



1. Barr (1980), n = 65,000
2. Downs (1978), n = 10,726
3. Feinmesser et al. (1986), n = 62,000
4. Fitzland (1985), n = 30,890
5. Kankkunen (1982), n = 31,280
6. Martin (1982), n = 4,126,268
7. Parving (1985), n = 82,265
8. Sehlin et al. (1990), n = 63,463
9. Sorri & Rantakallio (1985), n = 11,780
10. Davis & Wood (1992), n = 29,317
11. Fortnum et al. (1996), n = 552,558

Percentage of Sensorineural Hearing Losses Which Are Unilateral

Author (year)	# of Hearing Impaired Children in Sample	% Unilateral
Kinney (1953)	1307	48%
Brookhauser, Worthington & Kelly (1991)	1829	37%
Watkin, Baldwin, & Laoide (1990)	171	35%

Rate Per 1000 of Permanent Childhood Hearing Loss in UNHS Programs

Site	Sample Size	Prevalence Per 1000	% of Refers with Diagnosis
Rhode Island (8/90 - 12/91)	3,300	3.98	73%
Rhode Island (3/93 - 6/94)	16,395	1.71	42%
Colorado (1/92 - 12/96)	41,976	2.56	48%
New York (1/96 - 12/96)	27,938	1.65	67%
Utah (7/93 - 12/94)	4,012	2.99	67%
Hawaii (1/96 - 12/96)	9,605	4.15	98%
New Jersey (1/93 - 12/95)	15,749	3.30	?

Acquired Permanent Childhood Hearing Losses

- ✓ Traditionally assumed to be as prevalent as congenital PCHL.
- ✓ In states where UNHS programs have operated for 5+ years and hundreds of newborns with PCHL have been identified (RI, CO, UT, HI), few, if any, acquired hearing losses are being identified.
- ✓ Although research is lacking, an emerging hypothesis is that many, if not most, children previously considered to have acquired PCHL were really mild congenital losses which were progressive.

II. Accuracy of Screening Tests

- ✓ Auditory Brainstem Response (ABR)
- ✓ High-Risk Indicators
- ✓ Behavioral Evaluation at 7-9 Months
- ✓ Otoacoustic Emissions (OAE)
- ✓ Automated ABR

Sensitivity of Various UNHS Techniques

- ♦ **Although various rates of sensitivity are reported, there are no studies of UNHS with sufficient sample sizes to definitively establish sensitivity for any of the techniques.**
- ♦ **Weakness with existing studies of "sensitivity"**
 - ✓ **Small sample sizes.**
 - ✓ **One screening technique compared to another screening technique (e.g., OAE vs. ABR).**
 - ✓ **All screening passes are not followed.**
 - ✓ **Samples include only high-risk babies.**

Accuracy of ABR for Newborn Hearing Screening

Hyde, Riko, and Malizia (1990)

- ♦ 713 at-risk infants screened with ABR prior to hospital discharge.
- ♦ Children evaluated by "blind" examiners at mean of 3.9 years of age (range 3-8years).
- ♦ Results based on 1367 ears with reliable ABR and pure tone thresholds.

		Hearing Status	
		Impaired	Normal
ABR Screen (40 dBHL)	Refer	44	57
	Pass	1	1265

Sensitivity = 98%
Specificity = 96%

		Hearing Status	
		Impaired	Normal
ABR Screen (30 dBHL)	Refer	45	125
	Pass	0	1197

Sensitivity = 100%
Specificity = 91%

Hyde, M.L., Riko, K., & Malizia, K. (1990). Audiometric accuracy of the click ABR in infants at risk for hearing loss. *J Am Acad Audiol*, 1, 59-66.

Accuracy of ABR for UNHS

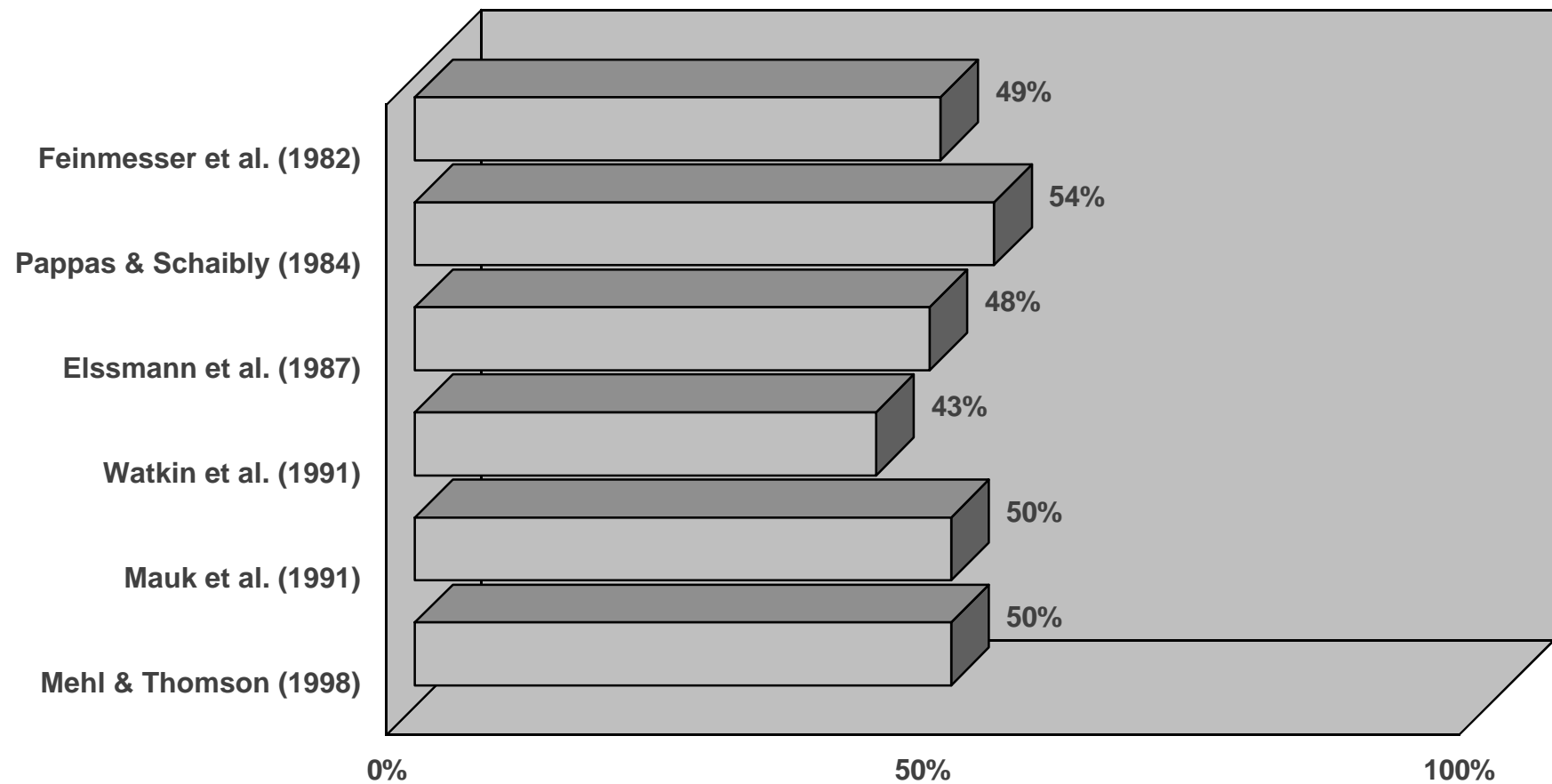
Saint Barnabus Medical Center, NJ

- ♦ 15,749 infants born from 1/1/93 to 12/31/95 screened with Nicolet Compass ABR system without sedation.
- ♦ Normal care nursery babies screened at 35 dB HL; NICU and High Risk screened at 40 dB HL and 70 dB HL.
- ♦ Screening done by audiologists, usually within 24 hours of birth.
- ♦ Babies with a High Risk Indicator who passed initial screen were re-evaluated at 6 months.

<u>Births</u>	<u># and % Screened</u>	<u># and % Referred</u>	<u>PCHL # and Prevalence</u>
16,229	15,749 (97%)	485 (3.1%)	52 3.3/1000

Barsky-Firkser, L., & Sun, S. (1997). Universal newborn hearing screenings: A three year experience. *Pediatrics*, *99*(6), 1-5.

What Percentage of Hearing Impaired Children were High Risk as Infants?



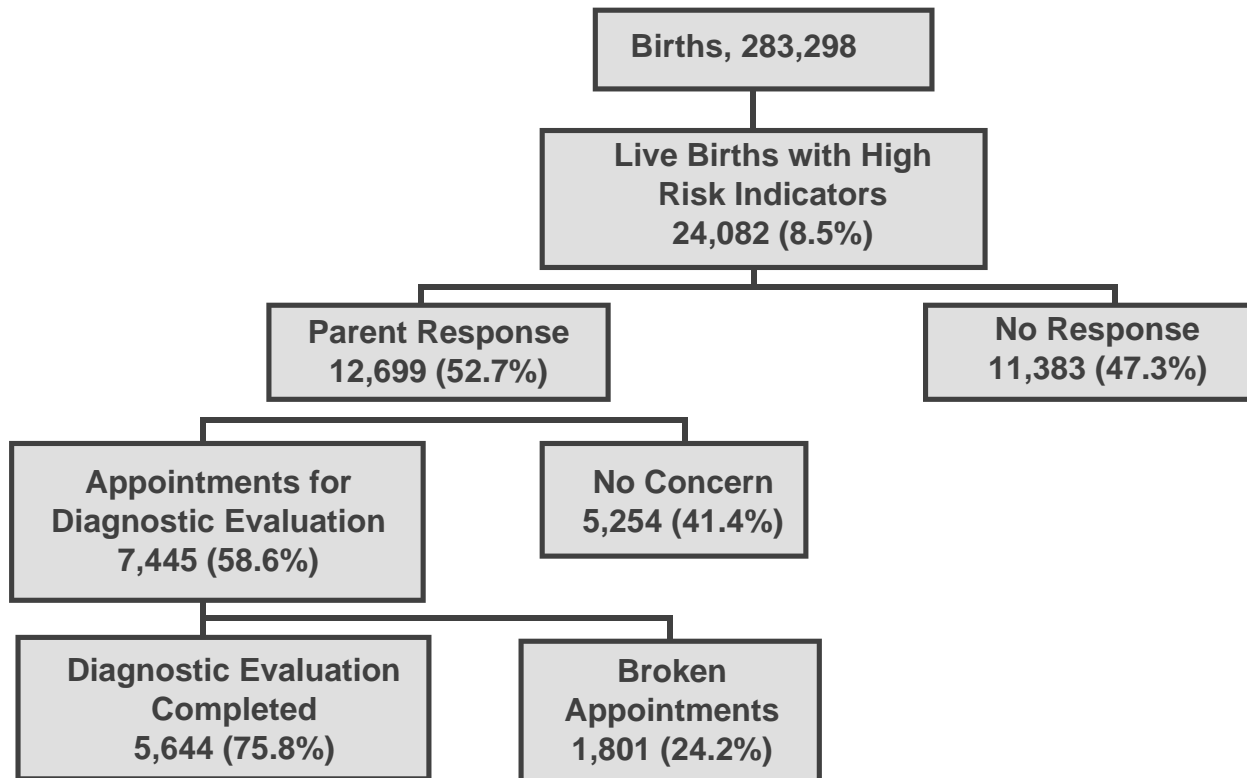
Accuracy of High Risk Based UNHS Programs

Mahoney and Eichwald (1987)

- ✓ Program operational from 1978-1995.
- ✓ JCIH indicators incorporated into legally required birth certificate.
- ✓ Computerized mailing and follow-up, and free diagnostic assessments at regional offices and/or mobile van.
- ✓ Program now discontinued because:
 - ◆ parents only made appointments for about 1/2 the children who had a risk indicator.
 - ◆ only about 1/2 of the children with an appointment showed up.
 - ◆ of difficulty obtaining accurate information from hospitals for some risk indicators.

Mahoney, T.M., & Eichwald, J.G. (1987). The ups and "downs" of high-risk hearing screening: The Utah statewide program. Seminars in Hearing, 8(2), 155-163.

Results of Birth Certificate Based High Risk Registry to Identify Hearing Loss in Utah (1978-1984)



Summary: 23.4% of live births with high-risk indicators completed a diagnostic evaluation; .36 SNHL per 1000 identified.

Mahoney, T.M., & Eichwald, J.G. (1987). The ups and "downs" of high-risk hearing screening: The Utah statewide program. *Seminars in Hearing*, 8(2), 155-163.

Accuracy of Home-Based Behavioral Screening

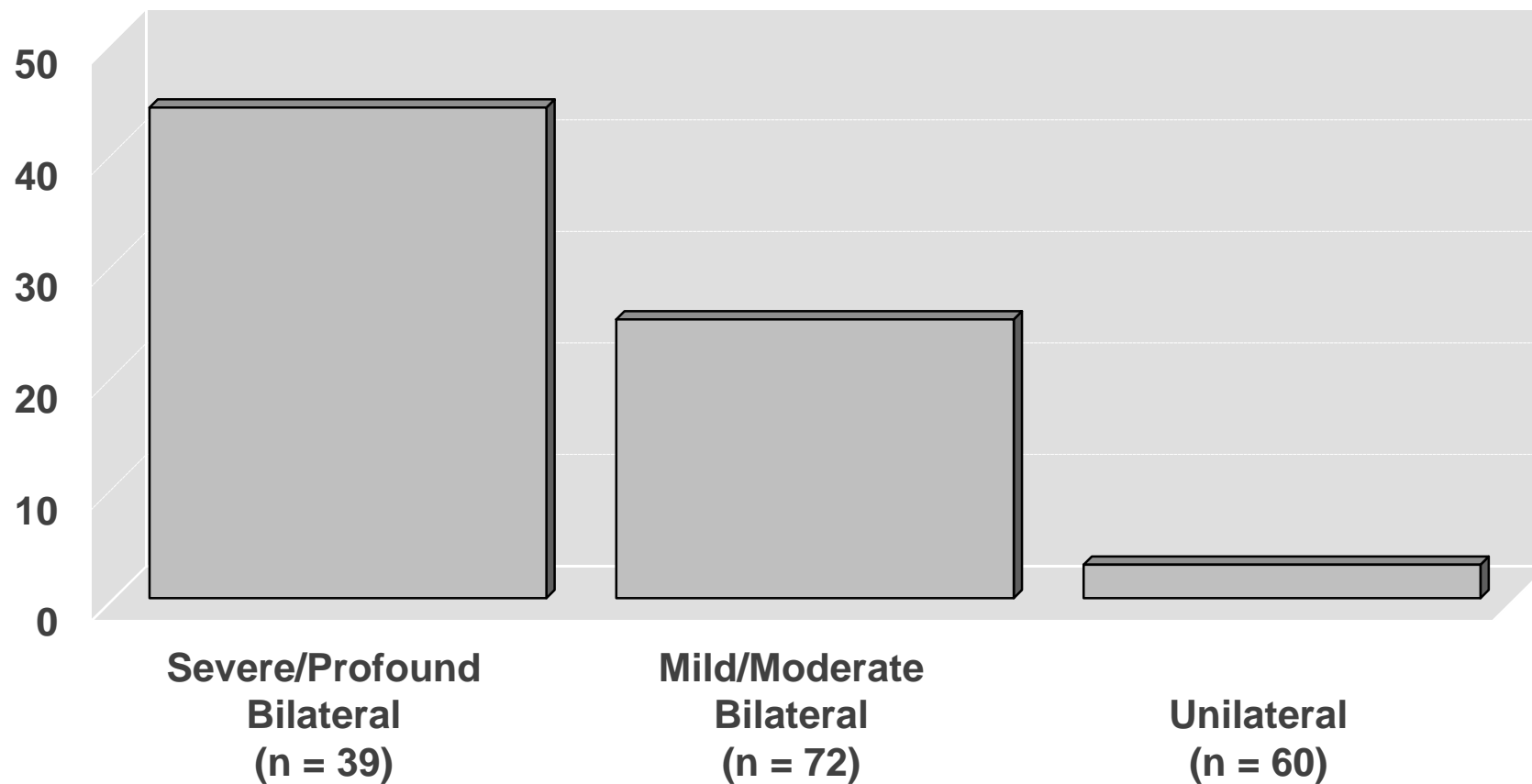
Watkin, Baldwin and Laoide, 1990*

- ♦ Retrospective analysis of 171 hearing impaired children to determine how they were identified.
- ♦ Hearing loss first noticed by:

	Home visitor or School Screening	Parent	Other than Parent (e.g., teacher, doctor, etc.)
Severe/profound Bilateral (n = 39)	18 (46%)	10 (26%)	11 (28%)
Mild/Moderate Bilateral (n = 72)	51 (71%)	14 (19%)	7 (10%)
Unilateral	34 (57%)	18 (30%)	8 (13%)

*Parental suspicion and identification of hearing impairment. *Archives of Disease in Childhood*, 65, 846-850.

Percentage of Hearing Impaired Children in Watkin, et al. (1990) Identified by Home Screening at 7-9 Months of Age



Accuracy of OAE-Based Newborn Hearing Screening

Plinkert et al. (1990)

- Sample:** 95 ears of high-risk infants
- Comparison:** TEOAE vs. ABR (≥ 30 dB) @ mean age = 9 weeks)
- Results:** TEOAE compared to ABR: sensitivity = 90%;
specificity = 91%

Plinkert, P.K., Sesterhenn, G., Aroid, R., & Zenner, H.P. (1990). Evaluation of otoacoustic emissions in high-risk infants by using an easy and rapid objective auditory screening method. European Archives of Otorhinolaryngology, 247, 356-360.

Kennedy et al. (1991)



- Sample:** 370 infants (223 NICU, 61 normal nursery with risk factors, and 86 normal Inursery with no risk factors)
- Comparison:** TEOAE, ABR (≥ 35 dB), and Automated ABR (≥ 35 dB) all at 1 month vs. behaviorally confirmed hearing loss, mean age = 8 months
- Results:** TEOAE identified same 3 infants with sensorineural hearing loss as ABR and automated ABR

Kennedy, C.R., Kimm, L., Dees, D.C., Evans, P.I.P., Hunter, M., Lenton, S., & Thornton, R.D. (1991). Otoacoustic emissions and auditory brainstem responses in the newborn. Archives of Disease in Childhood, 66, 1124-1129.

Rhode Island Hearing Assessment Project (RIHAP)

- ♦ 1850 infants (normal and special care) screened prior to hospital discharge with TEOAE and ABR
- ♦ Referrals for either TEOAE or ABR were rescreened at 3-6 weeks and referred for diagnosis as necessary

Fail TEOAE	■	■	■	■	■	■	■	■	■	■	■	n = 11
Fail ABR	■	■	■	■	■	■	■	■	■	□	■	n = 10
NICU	□	□	□	■	■	■	□	■	■	■	■	n = 7
High-Risk	□	□	■	■	□	■	□	■	■	■	■	n = 7

 Failed test, Present in NICU, Risk Factor Present
 Passed Test, Not in NICU, Risk Factors Absent

White, K.R., & Behrens, T.R. (Editors) (1993). The Rhode island Hearing Assessment Project: Implications for universal newborn hearing screening. Seminars in Hearing, 14(1).

Accuracy of TEOAE 2-Stage Screen*

Sensorineural Loss

		Hearing Status	
		Impaired	Normal
RIHAP Screen	Refer	11	79
	Pass	0	1643

"Sensitivity" = 100%

"Specificity" = 95%

***Note:** Analysis is based on heads. Infants initially screened but lost to follow-up or rescreen because of parent refusal, lost contact, or repeated broken appointments (> 3) are not included.

White, K.R., Vohr, B.R., Maxon, A.B., Behrens, T.R., McPherson, M.G., & Mauk, G.W. (1994). Screening all newborns for hearing loss using transient evoked otoacoustic emissions. International Journal of Pediatric Otorhinolaryngology, 29, 203-217.

Accuracy of Automated ABR

Hall, Kileny, Ruth, & Kripal
(1987) (336 ears)

		Conventional ABR	
		Refer	Pass
ALGO I	Refer	18	11
	Pass	0	307

Sensitivity = 100%
Specificity = 97%

Jacobson, Jacobson, & Spahr
(1990) (447 ears)

		Conventional ABR	
		Refer	Pass
ALGO I	Refer	33	17
	Pass	0	397

Sensitivity = 100%
Specificity = 96%

Accuracy of Automated ABR (continued)

Von Wedel, Schauseil-Zipf and Doring (1988) (100 ears)

		Conventional ABR	
		Refer	Pass
ALGO I	Refer	8	4
	Pass	2	86

Sensitivity = 80%
Specificity = 96%

Hermann et al. (1995)
(304 ears)

		Conventional ABR	
		Refer	Pass
ALGO I	Refer	42	6
	Pass	0	256

Sensitivity = 98%
Specificity = 100%

Accuracy of Automated ABR Summary of 4 Studies (1187 ears)

		Conventional ABR	
		Refer	Pass
ALGO I	Refer	101	38
	Pass	2	1046

Sensitivity = 96%

Specificity = 98%

Herrmann, B.S., Thornton, A.R., & Joseph, J.M. (1995). Automated infant hearing screening using the ABR: Development and validation. American Journal of Audiology, 4(2), 6-14.

NIH Study: Identification of Neonatal Hearing Impairment Multi-Center Study Based at University of Washington

- ✓ **Null Hypothesis: ABR, TEOAE, and DPOAE are equally effective for newborn hearing screening.**
- ✓ **7178 infants (4510 NICU and 2668 normal nursery) screened prior to discharge with ABR, TEOAE, and DPOAE in random order.**
- ✓ **Screening results will be compared with ear specific VRA at 8-12 months.***
- ✓ **Other issues investigated:**
 - ✓ **Influence of co-existing medical factors on characteristics of OAE and ABR.**
 - ✓ **Optimum stimulus and recording parameters for OAE.**
 - ✓ **Time and cost-efficiency of ABR and OAE.**
 - ✓ **Influence of external and middle ear status, test environment, and tester characteristics.**

Data collection completed October, 1997; data expected to be reported April 1998.

III. Efficiency of Existing UNHS Programs

- ✓ **Coverage and Referral Rates**
- ✓ **Effects of UNHS on Age of Identification**
- ✓ **Effects on Parents**
- ✓ **Follow-up**

Births Per Year, Percent of Babies Screened, and Reported Referral Rates of Universal Newborn Hearing Screening Programs

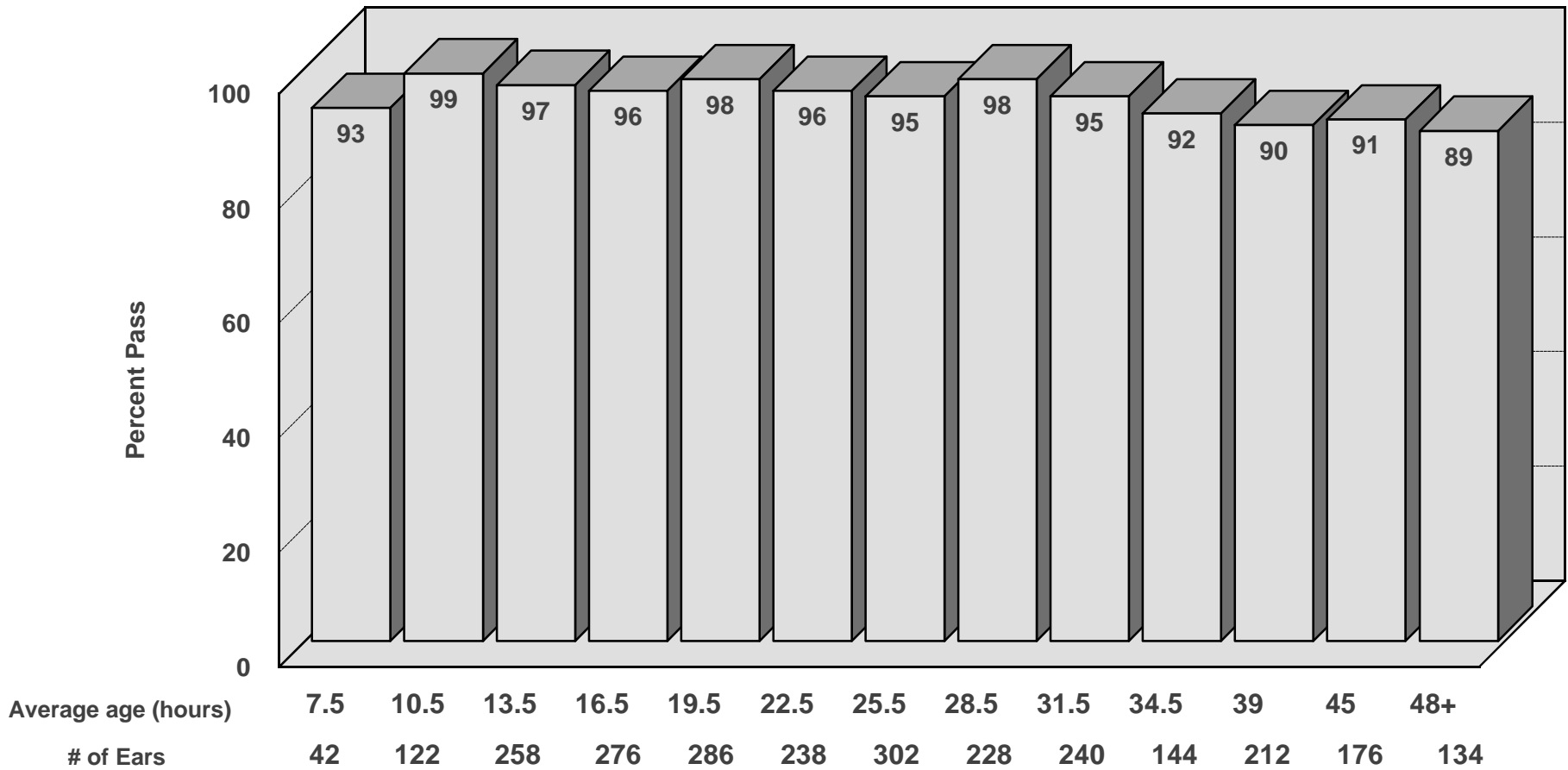
	# of Hospitals	Average Births Per year	Percent Babies Screened Before Discharge	Reported Pass Rate at Discharge
OAE-Based Programs ^a	64	2140	94.9	91.6%
ABR-Based Programs ^b	56	1348	96.2	96.0%
All Programs	120	1767	95.5	93.7%

^a55 of 64 OAE-based programs were TEOAE, 9 were DPOAE

^b54 of 56 ABR-based programs were automated ABR

White, K.R., Mauk, G.W., Culpepper, N. B., & Weirather, Y. (1998). Newborn hearing screening in the United States: Is it becoming the standard of care? In L. Spivak (Ed.), Universal newborn hearing screening (pp. 225-255. Thieme: New York.

TEOAE Pass Rate for Inpatient Newborn Hearing Screen for Infants at Different Ages



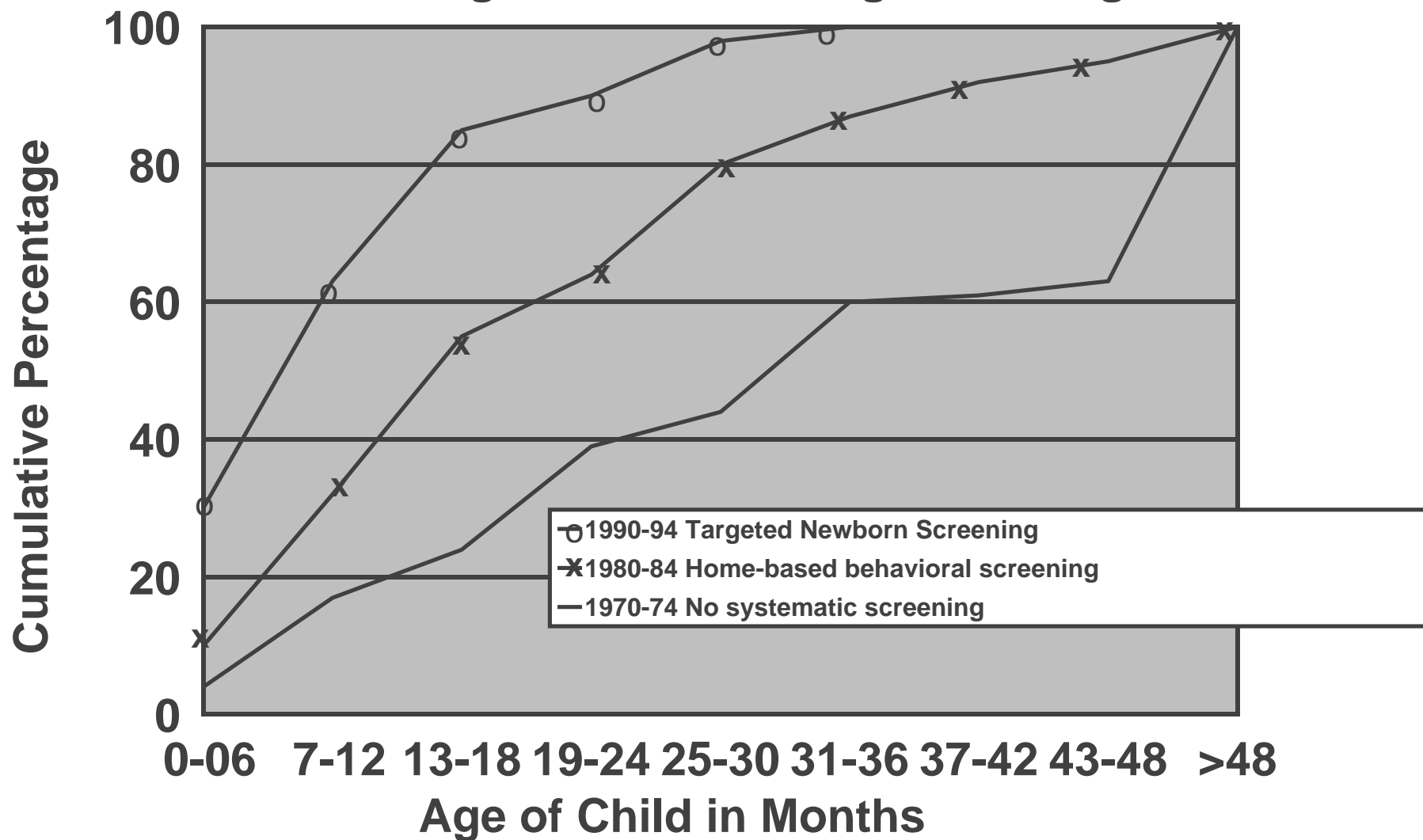
Maxon, A.B., White, K.R., Culpepper, B., & Vohr, B.R. (1997). Maintaining acceptably low referral rates in TEOAE-based newborn hearing screening programs. *Journal of Communication Disorders*, 30(6), 457-475.

Targeted Neonatal Screening vs. Universal Home-Based at 7-9 Months

- ✓ Five-year cohorts of all births in Copenhagen county beginning in 1970, 1980, and 1990 were analyzed for age of hearing loss identification (bilateral, ≥ 25 dB HL)
- ✓ Prior to 1975, no systematic procedures were used for early identification of hearing loss.
- ✓ Beginning in 1975, a home-based behavioral hearing screening procedure (BOEL) was done for 95%+ of all children by trained health visitors.
- ✓ From 1990-1994, an OAE newborn hearing screening was done for about 20% of births.
- ✓ About 37,000 births in each 5-year cohort.

Parving, A., & Salomon, G. (1996). The effect of neonatal universal hearing screening in a health surveillance perspective - a controlled study of two health authority districts. *Audiology*, 35, 158-168.

Cumulative Percentage of the Age at Which Children are Identified Using Different Hearing Screening Protocols



Possible Adverse Effects for Parents of Various Hearing Screening Results

- ♦ **False-Positive**
 - ✓ Adversely affect parent-child bonding (e.g., rejection or over-protection).
 - ✓ Anger, resentment, or confusion when child is confirmed normal.
 - ✓ Lingering concerns about whether child's hearing is normal.

- ♦ **False Negative**
 - ✓ Inappropriate confidence that child hears normally, thus delaying identification.

- ♦ **True Positive**
 - ✓ Emotional stress during time of emerging parent-child relationship.
 - ✓ Incomplete or inaccurate information may be used to make future reproductive decisions.

Adapted from Clayton, E.W. (1992). Screening and treatment of newborns. Houston Law Review, 29(1), 85-148.

Do False-Positives in Newborn Hearing Screening Create Parental Anxiety?

Example #1: Screening for Cystic Fibrosis (Tluczek et al., 1992)

- ✓ 104 parents of children who failed IRT.
- ✓ 54-item questionnaire (17 items about parental anxiety, 5 point Likert Scale).
- ✓ Data collected by phone or personal interview after the initial test.

Example #2: Screening for Congenital Hearing Loss (Uzcategui, 1997)

- ✓ 171 parents of children who failed initial newborn hearing screen.
- ✓ 46-item question, 4 point Likert Scale.
- ✓ Data collected by mailed questionnaire (40% response rate, n = 69) after child failed initial screen.

Tluczek, A., Mischler, E. H., Farrell, P. M., Fost, N., Peterson, N. M., Carey, P., Bruns, W. T., & McCarthy, C. (1992). Parents' knowledge of neonatal screening and response to false-positive cystic fibrosis testing. *J Dev Behav Pediatr*, *13*(3), 181-6.

Uzcategui, C. A. D. (1977). Refer rate and parental anxiety in newborn hearing screening in the state of Colorado. Unpublished doctoral dissertation, University of Colorado at Boulder, Colorado.

Parents strongly agreed or agreed that the following terms described their reaction to initial screening test results:

	Tluczek (1992)	Uzcategui (1997)
Concern/Fear	98%	45%
Shock	76%	25%
Anger	48%	14%
Confusion	61%	35%

Other Findings from Tulczek (1992) Cystic Fibrosis Study

- ✓ **Less ambiguity about what test results mean if results are explained in person.**
- ✓ **6% of parents (all of whom were notified by phone) had "lingering anxiety" a year later about whether their child had cystic fibrosis.**
- ✓ **75% did not know their child was being screened for cystic fibrosis.**

Parents' Perceptions of Screening

Questionnaires administered by nurses to 169 babies born between 6/1/94 and 7/15/94.

Question	% Answering Yes
If you were in a hospital where you had to give your permission to have your baby's hearing screened, would you give it?	98%
If this screening were conducted for a fee of approximately \$30, would you be willing to pay it?	71%
Do you believe that any anxiety caused by your baby not passing the hearing screening would be outweighed by the benefits of early detection if a hearing loss was found to be present?	88%

Barringer, D.G., & Mauk, G.W. (1997). Survey of parents' perceptions regarding hospital-based newborn hearing screening. *Audiology Today*, 9(1), 18-19.

Parents' Views About Newborn Hearing Screening

Watkin, Beckman, and Baldwin, 1995

- ✓ 208 parents of children with sensorineural hearing loss (average age of child = 12.3 years) answered written questionnaires.
- ✓ None of the children participated in a newborn hearing screening program.
- ✓ 58.5% response rate.
 - ◆ 58% wished their child had been identified earlier.
 - ◆ Only those whose children's impairments were mild or who were confirmed in the first 18 months of life were satisfied with the age of confirmation.
 - ◆ 89% preferred having a newborn hearing screening program instead of what they had.

Watkin, P.M., Beckman, A., & Baldwin, M. (1995). The views of parents of hearing impaired children on the need for neonatal hearing screening. British Journal of Audiology, 29, 259-262.

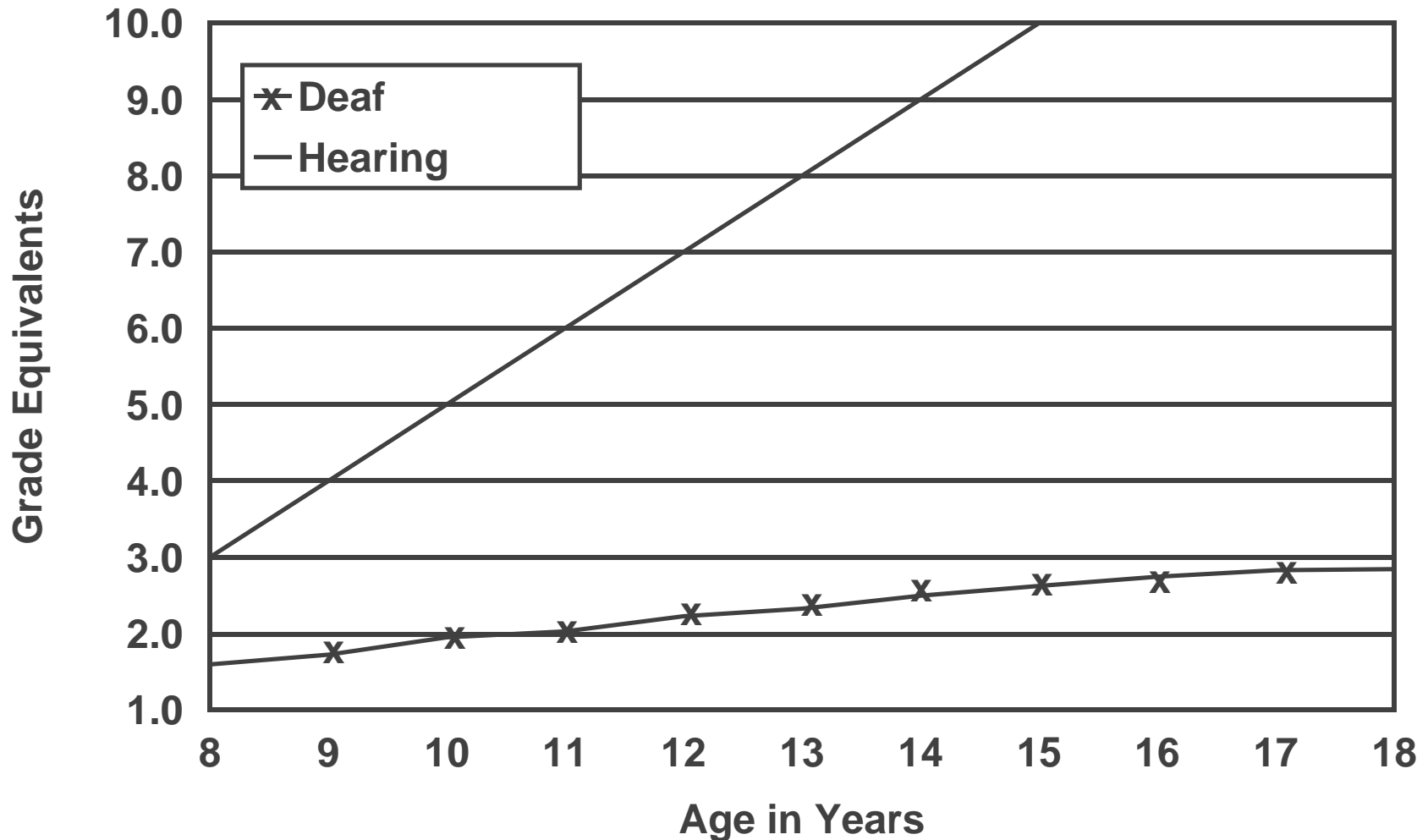
Tracking "Refers" is a Major Challenge

	Births	Screened	Initial Refer	Rescreen	Rescreen Refer	Conclusive Diagnosis	PCHL # (Prevalence)
Colorado (1/92 - 12/96)	??	41,796	2,709 (6%)	NA	NA (6%)	1,296 (48%)	107 (2.56/1000)
Rhode Island (1/93 - 12/96)	53,121	52,659 (99%)	5,397 (10%)	4,575 (85%)	677 (1.3%)	??	111 (2.12/1000)
Hawaii (1/96 - 12/96)	10,584	9,605 (91%)	1,204 (12%)	991 (82%)	121 (1.3%)	119 (98%)	40 (4.15/1000)
Utah (9/96-10/97)	4,572	4,012 (91%)	561 (14%)	330 (59%)	36 (0.8%)	24 (67%)	12 (2.99/1000)
New York (1/96-12/96)	28,951	27,938 (96.5%)	1,953 (7%)	1,040 (53%)	245 (0.8%)	165 (67%)	46 (1.65/1000)

IV. Consequences of Neonatal Hearing Loss

- ✓ **Severe/Profound PCHL Losses**
- ✓ **Mild Bilateral and Unilateral PCHL Losses**
- ✓ **Fluctuating Conductive Loss**

Reading Comprehension Scores of Hearing and Deaf Students



Schildroth, A.N., & Karchmer, M.A. (1986). Deaf children in America. San Diego: College Hill Press.

Studies of Unilateral Hearing Loss

	Sample Size		Average Age
	USNHL	Matched Control	
Keller & Bundy (1980)	13	13	12.0 yrs
Peterson (1981)	24	24	7.5 yrs
Bess & Thorpe (1984)	25	25	10.0 yrs
Blair, Peterson, & Viehweg (1985)	8	8	7.5 yrs
Culbertson & Gilbert (1986)	25	25	10.0 yrs

Effects of Unilateral Hearing Loss

	Standardized Mean Difference Effect Size for		
	Math	Language	Social
Keller & Bundy (1980)	-.26	-.44	
Peterson (1981)	-.53	-.50	
Bess & Thorpe (1984)			-.47
Blair, Peterson, & Viehweg (1985)	-1.28	-1.17	
Culbertson & Gilbert (1986)	-.19	-.63	-.47

Effects of Mild Fluctuating Conductive Hearing Loss

Teele, et al., 1990

- ♦ 194 children followed prospectively from 0-7 years.
- ♦ Days child had otitis media between 0-3 years assessed during normal visits to physician.
- ♦ Data on intellectual ability, school achievement, and language competency individually measured at 7 years by "blind" diagnosticians.
- ♦ Results for children with less than 30 days OME were compared to children with more than 130 days adjusted for confounding variables.

<u>Outcome Measure</u>	<u>Effect Size for Less vs. More OME</u>
WISC-R Full Scale	.62
Metropolitan Achievement Test	
Math	.48
Reading	.37
Goldman Fristoe Articulation	.43

Teele, D.W., Klein, J.O., Chase, C., Menyuk, P., Rosner, B.A., and the Greater Boston Otitis Media Study Group (1990). Otitis media in infancy and intellectual ability, school achievement, speech, and language at age 7 years. The Journal of Infectious Diseases, 162, 685-694.

V. Benefits Resulting From Earlier Identification and Intervention

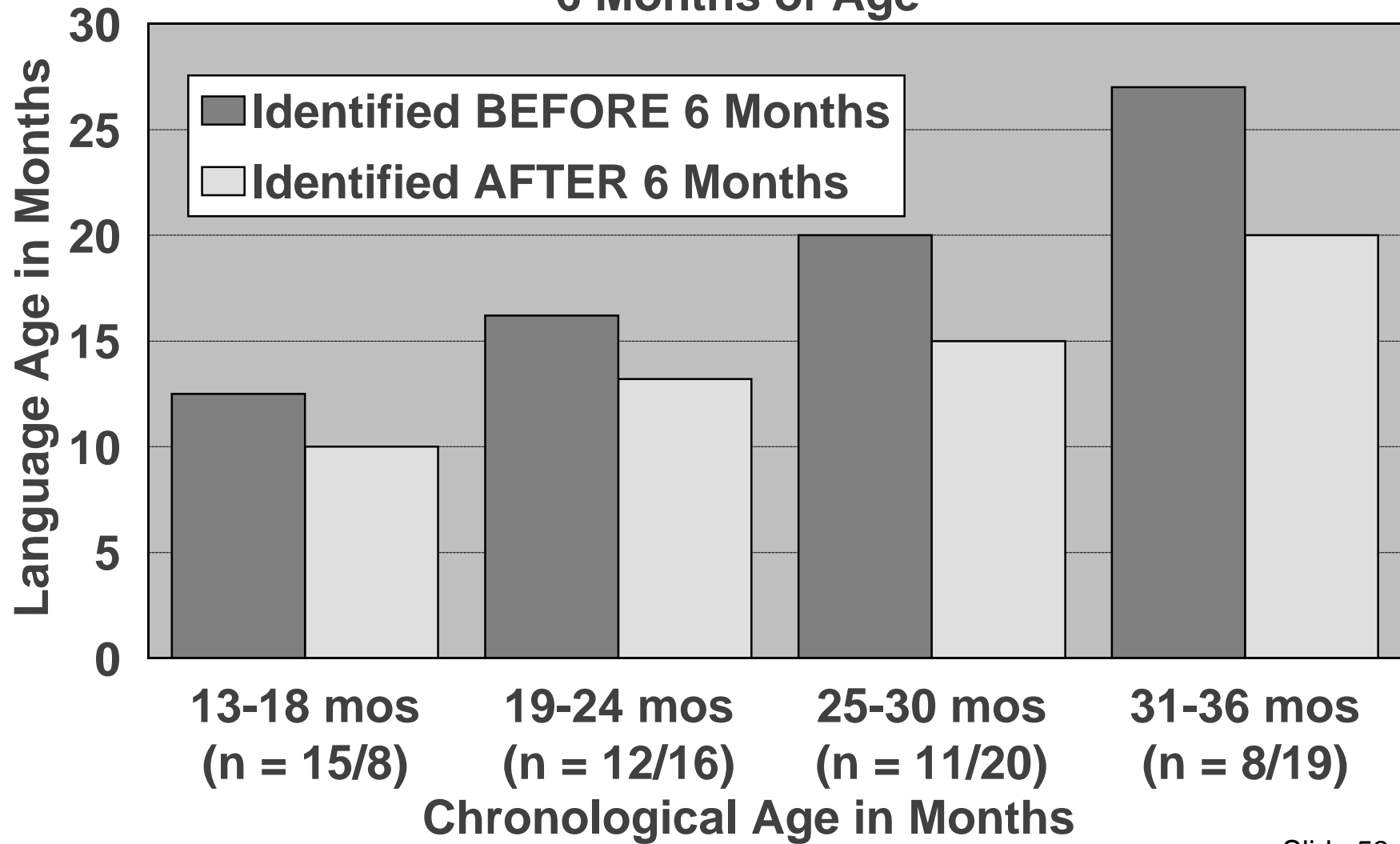
- ✓ **Prospective randomized trials have not been done.**
- ✓ **Most existing evidence is weakened by:**
 - ◆ **potential for selection bias.**
 - ◆ **lack of long-term follow-up to assess "wash-out" effect.**
 - ◆ **small sample sizes.**
 - ◆ **subjective assessments of outcomes.**

Yoshinaga-Itano, et al., 1996

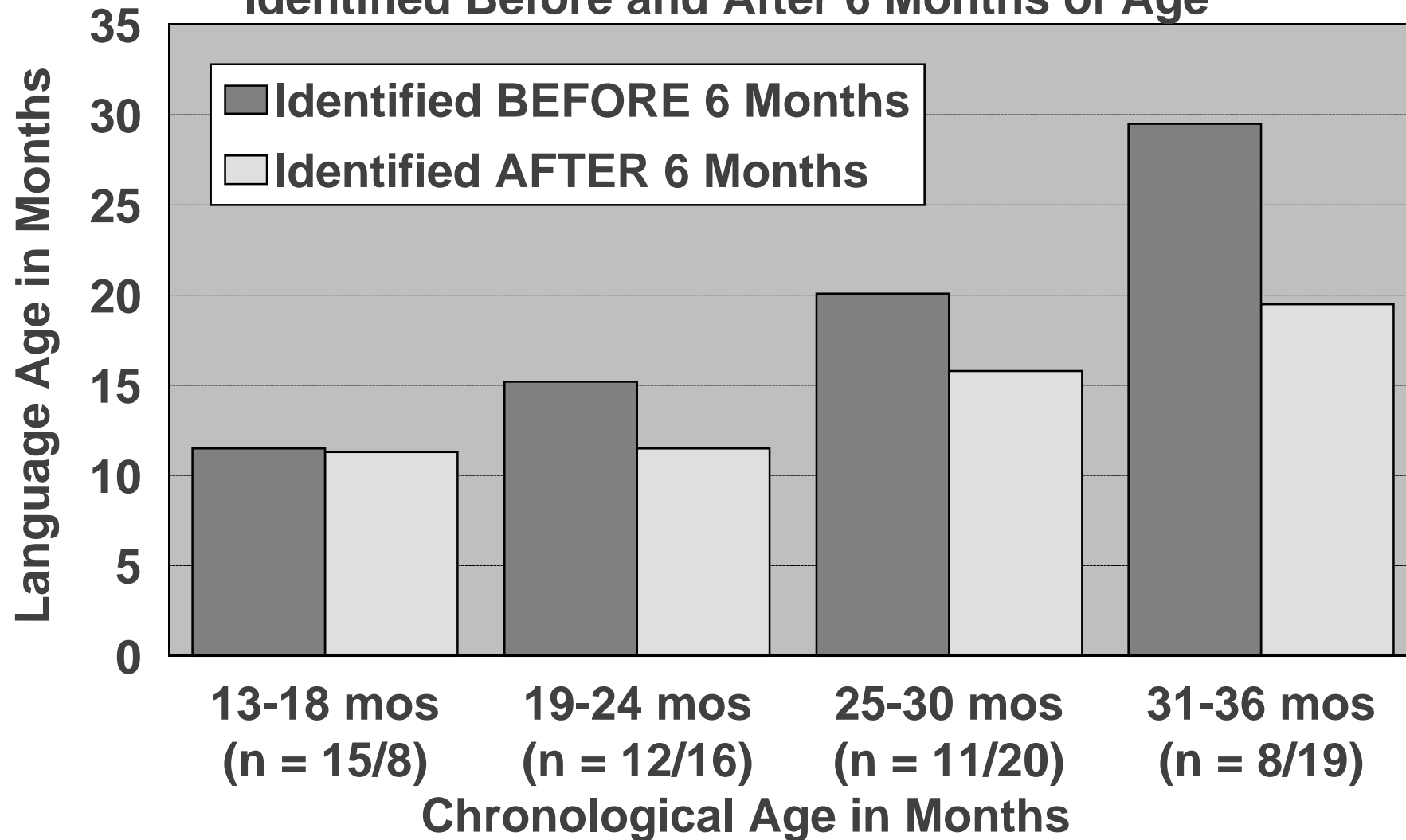
- ♦ Compared language abilities of hearing-impaired children identified before 6 months of age (n = 46) with similar children identified after 6 months of age (n = 63).
- ♦ All children had bilateral hearing loss ranging from mild to profound, and normally-hearing parents.
- ♦ Language abilities measured by parent report using the Minnesota Child Development Inventory (expressive and comprehension scales) and the MacArthur Communicative Developmental Inventories (vocabulary).
- ♦ Cross-sectional assessment with children categorized in 4 different age groups.

Yoshinaga-Itano, C., Sedey, A., Apuzzo, M., Carey, A., Day, D., & Coulter, D. (July 1996). The effect of early identification on the development of deaf and hard-of-hearing infants and toddlers. Paper presented at the Joint Committee on Infant Hearing Meeting, Austin, TX.

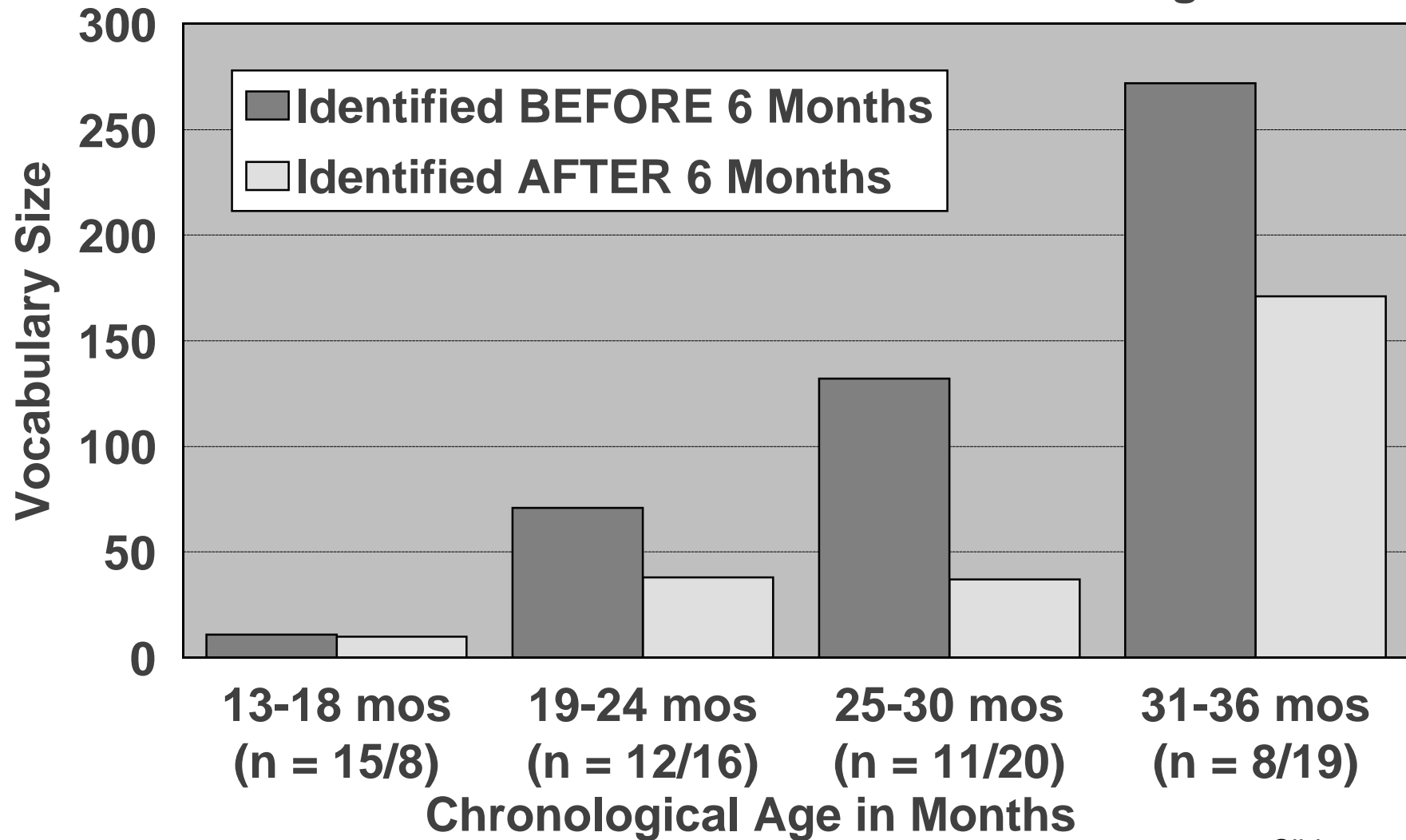
Language Comprehensive Scores for Hearing Impaired Children Identified Before and After 6 Months of Age



Expressive Language Scores for Hearing Impaired Children Identified Before and After 6 Months of Age



Vocabulary Size for Hearing Impaired Children Identified Before and After 6 Months of Age



Apuzzo and Yoshinaga-Itano, 1995

- ♦ 69 hearing impaired children categorized in 4 groups according to age of identification.

0 - 2 months (n = 14)

13 - 24 months (n = 30)

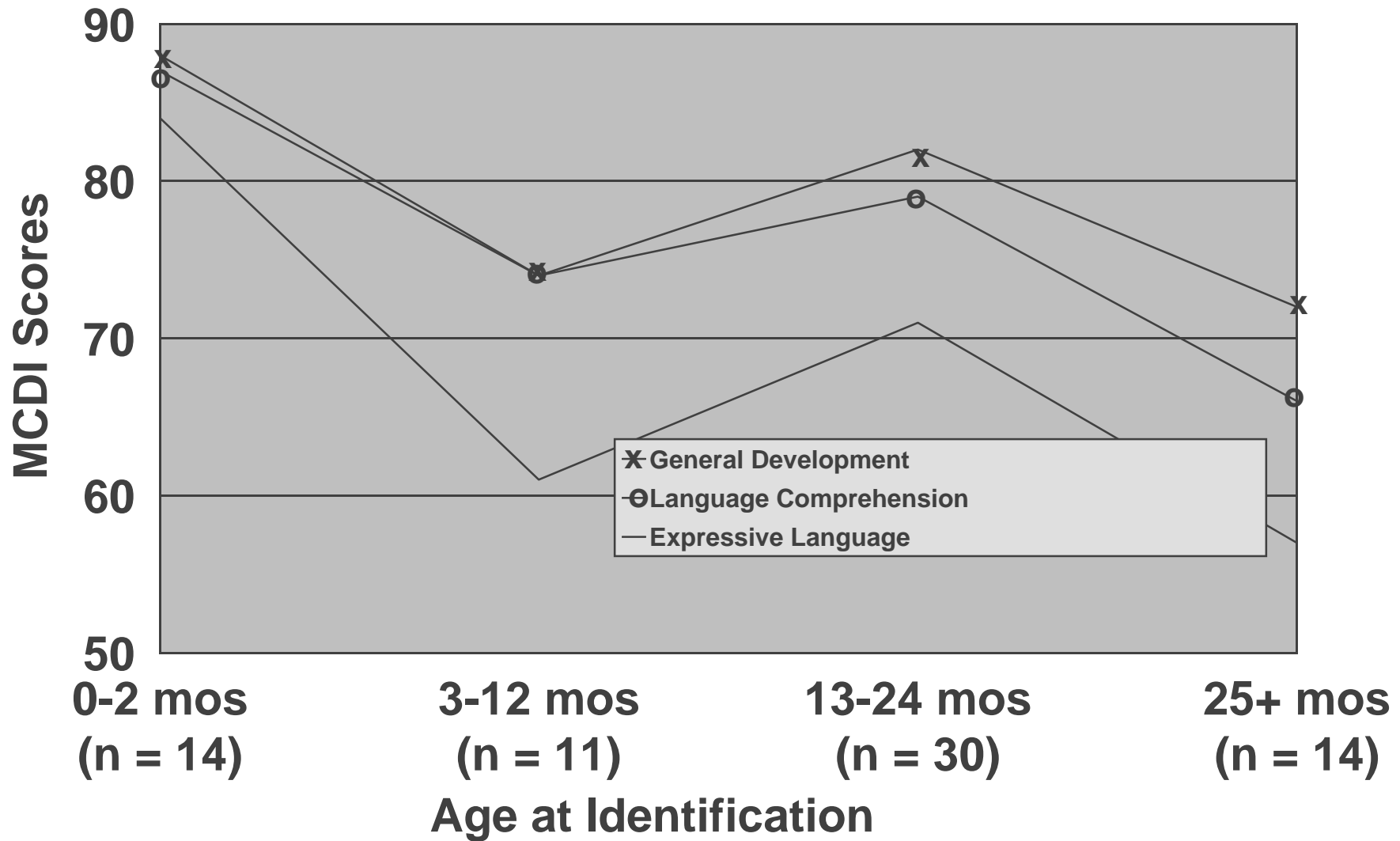
3 - 12 months (n = 11)

25+ months (n = 14)

- ♦ Groups reasonably similar with respect to age at time of testing, degree of hearing loss, and DQ. Children with substantial cognitive delays were eliminated.
- ♦ All subjects participating in similar early intervention programs, but began at different ages.
- ♦ Outcome data based on parent report using Minnesota Child Development Inventory.
- ♦ Results based on covariance adjustments for degree of hearing loss and cognitive ability.

Apuzzo, M. L., & Yoshinaga-Itano, C. (1995). Early identification of Infants with Significant Hearing Loss and the Minnesota Child Development Inventory. Seminars in Hearing, 16(2), 124-139.

MCDI Scores for Deaf and Hard of Hearing Children Identified at Different Ages



Watkins, 1987

- ♦ Comparisons made among 3 groups of bilaterally hearing-impaired children (n = 23 in each group)
 - Group #1: Received average of 9 months home intervention before 30 months age, followed by preschool intervention.
 - Group #2: Attended preschool beginning at an average of 36 months.
 - Group #3: Received no home intervention and no preschool intervention.
- ♦ Children matched on hearing severity (PTA ~ 85 dBHL), presence of other handicaps, and analysis of covariance used to adjust for age at post test, age of mother, SES, and number of childhood middle ear infections.
- ♦ Data collected by uninformed, trained examiners when children were 10 years old.

Watkins, S. (1987). Long term effects of home intervention with hearing-impaired children. American Annals of the Deaf, 132, 267-271.

Watkins, S. (1983). Final Report: 1982-83 work scope of the Early Intervention Research Institute, Logan, Utah: Utah State University.

Standardized Mean Difference Effect Sizes for Group Comparisons from Watkins, 1987

	EI & Preschool vs. Preschool	EI & Preschool vs. Nothing
Woodcock-Johnson Reading (letter/word identification & word attach)	0.64	0.99
Woodcock-Johnson Calculation	0.83	1.09
Peabody Picture Vocabulary Test	0.70	0.76
Arizona Articulation	0.26	0.78
% of Child's Communication Understood by Non-Family	0.92	1.23
% of Non-Family Communication Understood by Child	0.71	1.12
Meadow-kendall Social Adjustment	0.74	0.94
Child Behavior	0.61	0.96

Appropriate Intervention is Substantially Different For Different Groups of PCHL Children

- ✓ **Children with Recurrent Conductive Losses**
- ✓ **Children with Unilateral Losses**
- ✓ **Children with Mild/Moderate Bilateral Losses**
- ✓ **Children with Severe/Profound Bilateral Losses**

VI. Cost Efficiency of Newborn Hearing Screening

- ✓ What does early detection and intervention cost?
- ✓ Is protocol A more cost-effective than protocol B?
- ✓ Is early hearing detection and intervention cost-beneficial?

Most Existing Studies of Newborn Hearing Screening Costs Have Conceptual or Methodological Weaknesses

- ✓ **Too much of the cost estimate is based on hypothetical assumptions or unverified self reports.**
- ✓ **Cost analyses are incomplete (e.g., fringe benefits, overhead, costs to parents, equipment, etc., are not included).**
- ✓ **Only part of the early detection and intervention process is included in cost figures.**
- ✓ **Standard economic analysis concepts and procedures are ignored or misused.**

Actual Costs of Operating a Universal Newborn Hearing Screening Program

	Cost
Personnel	\$ 60,654
Screening Technicians (avg. 103 hrs./week)	
Clerical (avg. 60 hrs./week)	
Audiologist (avg. 18 hrs./week)	
Coordinator (avg. 20 hrs./week)	
Fringe Benefits (28% of Salaries)	16,983
Supplies, Telephone, Postage	12,006
Equipment	5,575
Hospital Overhead (24% of Salaries)	<u>14,557</u>
TOTAL COSTS	\$110,775

Cost Per Infant Screened = \$110,775 ÷ 4,253 = \$26.05

Maxon, A. B., White, K. R., Behrens, T. R., & Vohr, B. R. (1995) Referral rates and cost efficiency in a universal newborn hearing screening program using transient evoked otoacoustic emissions (TEOAE). Journal of the American Academy of Audiology, 6, 271-277.

Cost of a TEOAE-Based Universal Newborn Hearing Screening Program at Logan Regional Hospital (Utah)

February and March 1996

	Hours Worked	Average Rate/Hour	Cost
Personnel			
Screening	65.40	\$ 9.45	\$ 617.84
Rescreening	9.48	10.72	101.65
Screening Management	15.32	8.94	136.95
Program Management	5.23	10.15	53.12
Patient Management	12.90	11.05	142.54
Scoring	9.73	11.97	116.54
Total Personnel	118.07	\$9.90	\$1,168.63
Fringe Benefits (30% of salaries)	\$350.59		
Supplies	416.97		
Equipment	446.00		
Overhead (20% of costs)	476.44		
Total Costs	\$2,858.62		

Cost Per Baby = \$2,859 / 385 babies = \$7.42

Weirather, Y., Korth, N., White, K. R., Downs, D., Woods-Kershner, N. (1997). Cost analysis of TEOAE-based universal newborn hearing screening. Journal of Communication Disorders, 30(6), 457-475.

CDC Cost Study (1997)

- ✓ **Multi-center pilot UNHS cost study using 6 hospitals (one each in CO, GA, LA, TN, UT, and VA).**
- ✓ **Cost estimates based on self-report questionnaires with site visits to 4 of 6 sites.**
- ✓ **Standardized estimates used for equipment and overhead costs.**

Grosse, S. (September, 1997). The costs and benefits of universal newborn hearing screening. Paper presented to the Joint Committee on Infant Hearing, Alexandria, VA.

Results of CDC Cost Study

Cost category	3 Hospitals using TEOAE	3 Hospitals using AABR
Staff time	\$13.04	\$10.73
Equipment	0.91	2.63
Supplies	0.51	9.33
Overhead	3.49	3.34
Total Cost (Range)	\$17.96 (\$15-\$22)	\$26.03 (\$22-\$30)
Initial refer rate	8%	2%
Screening minutes per child	31.4	42.9
Audiologist minutes per child	17.0	5.4

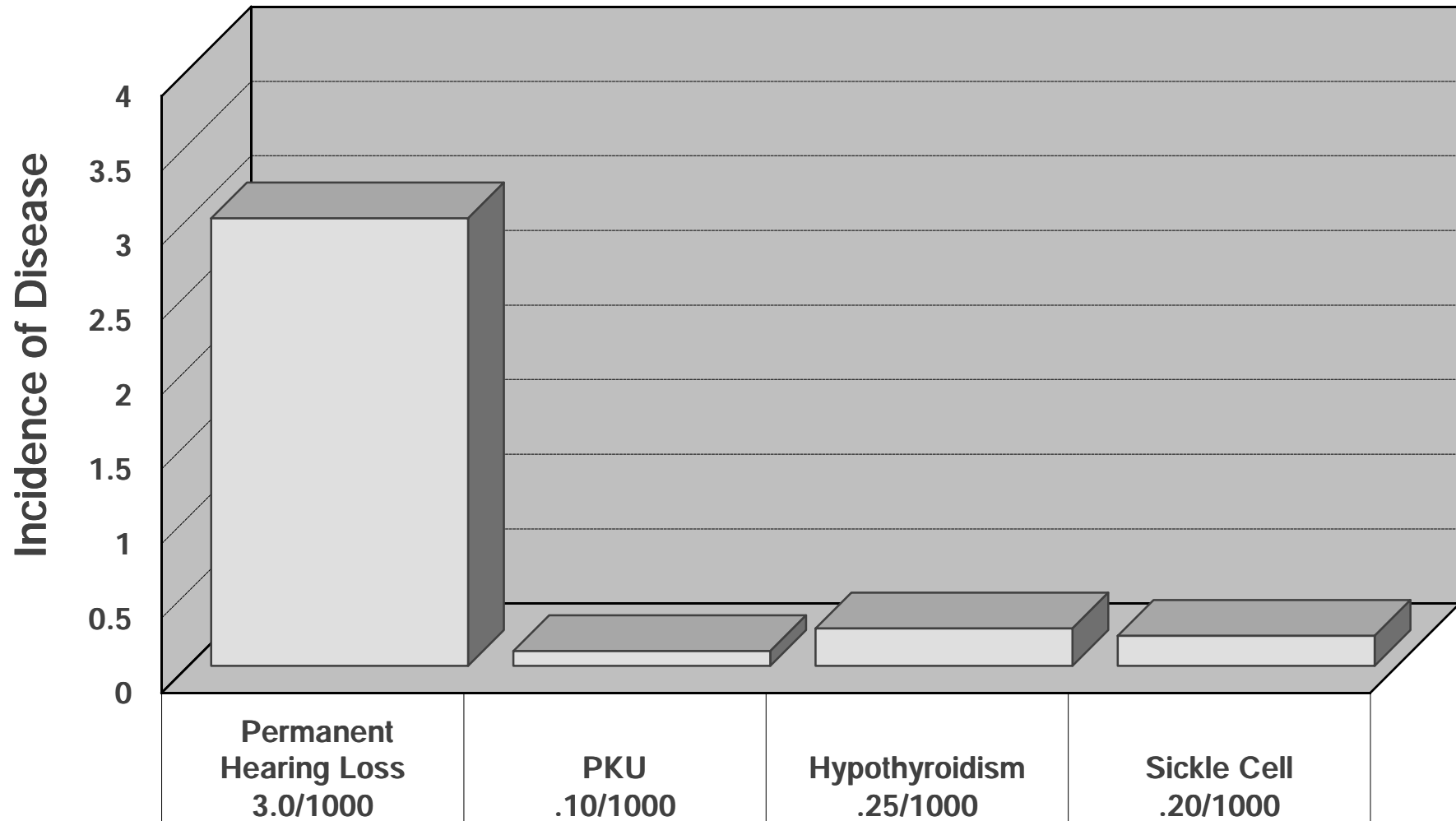
Stevens et al. (1997) Hearing Screening Cost Analysis

- ◆ Staff at 10 screening programs completed questionnaires regarding costs.
- ◆ Fringe benefits and overhead were added using standard multipliers.
- ◆ Included were 5 high-risk screening programs
3 universal screening programs
2 home visitor or surveillance programs

	Results Per 1000 Births	
<u>Type of Program</u>	<u>Mean Cost</u>	<u>Range</u>
High Risk	\$ 8,184	(\$6,894 to \$9,645)
Universal	\$22,480	(\$21,250 to \$23,940)
Home Visitor	\$32,120	(\$18,590 to \$36,190)

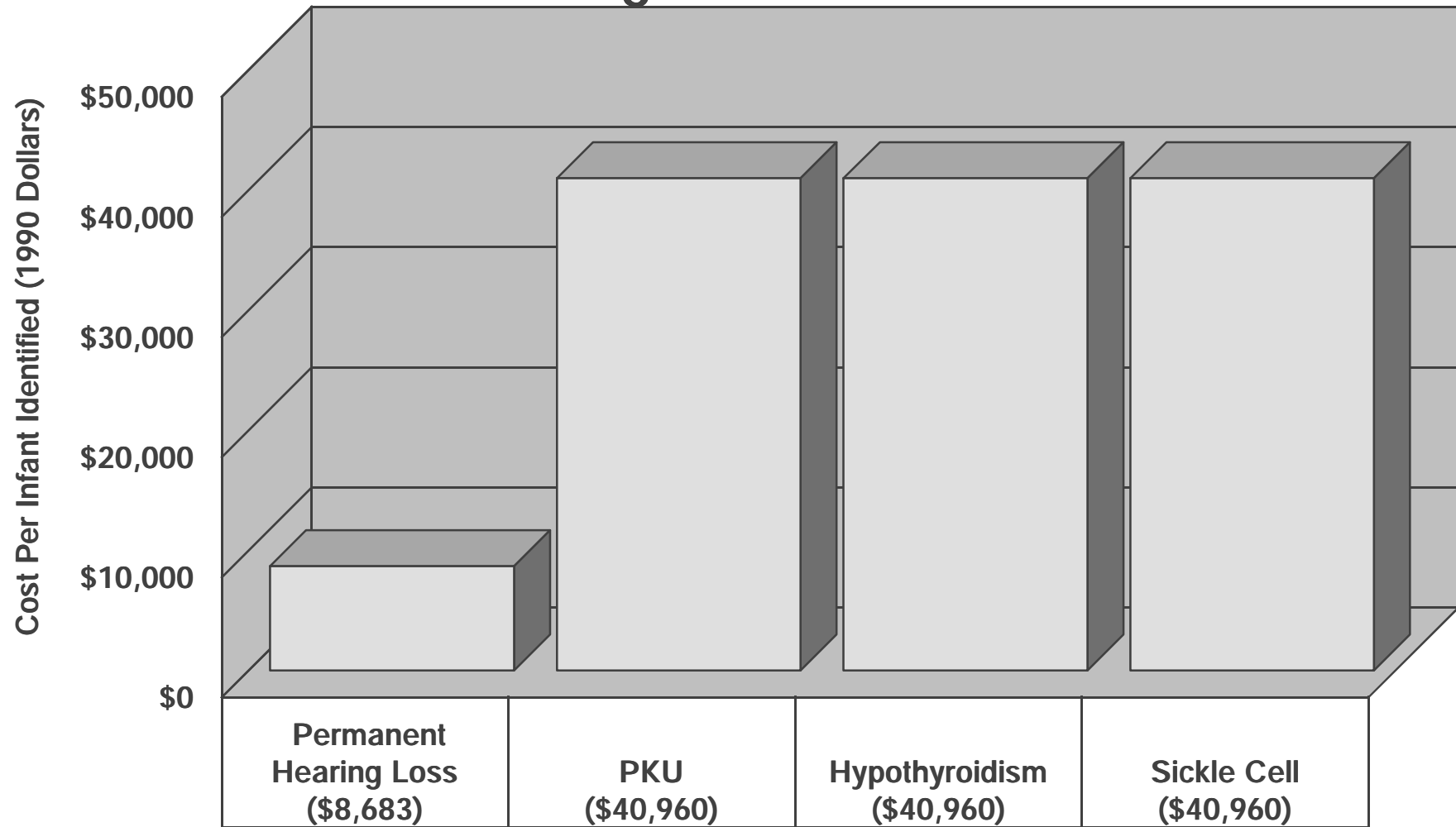
Stevens, J.C., Hall, D.M.B., Davis, A., Davies, C.M., & Dixon, S. (1997). The costs of early hearing screening in England and Wales. Unpublished manuscript, Royal Hallamshire Hospital, Sheffield, England.

Prevalence of Various "Screenable" Diseases Among Newborns



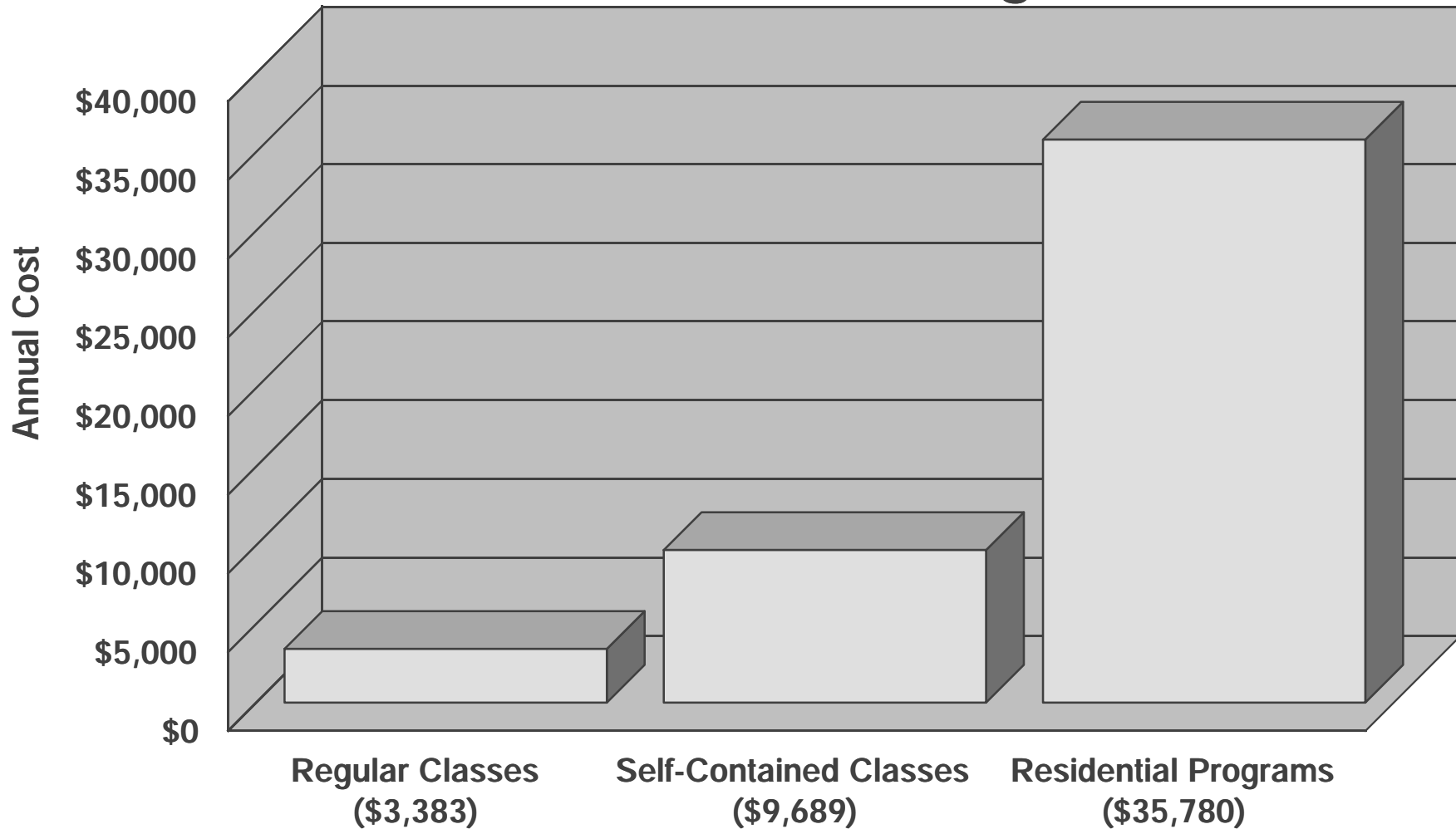
Johnson, J.L., Mauk, G.W., Takekawa, K.M., Simon, P.R., Sia, C.C.J., & Blackwell, P.M. (1993). Implementing a statewide system of services for infants and toddlers with hearing disabilities. *Seminars in Hearing*, 14(1), 105-119.

Cost of Identifying Infants with Various Diseases Using Current Screening Protocols in Rhode Island



Johnson, J.L., Mauk, G.W., Takekawa, K.M., Simon, P.R., Sia, C.C.J., & Blackwell, P.M. (1993). Implementing a statewide system of services for infants and toddlers with hearing disabilities. *Seminars in Hearing*, 14(1), 105-119.

Cost of Educating Children with Hearing Loss in Various Settings



Johnson, J.L., Mauk, G.W., Takekawa, K.M., Simon, P.R., Sia, C.C.J., & Blackwell, P.M. (1993). Implementing a statewide system of services for infants and toddlers with hearing disabilities. *Seminars in Hearing*, 14(1), 105-119.