Strategies to Scale-Up Global Access and Uptake of Hearing Screening:

A Systematic Review

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I. ABSTRACT

Introduction: Although interventions to address hearing loss exist, access is inequitably distributed across geographic, socioeconomic, and racial axes globally. We sought to determine which scale-up strategies could be useful to bolster the uptake of hearing screening to reduce the global burden of hearing loss. We then provide targeted policy recommendations to aid the implementation of these strategies.

Methods: After evaluating articles from five databases using our inclusion/exclusion criteria, we extracted qualitative and statistical evidence related to the uptake of neonatal, child, and adult hearing screening (NHS, CHS, and AHS), specifically their use, adherence, and satisfaction. Two reviewers independently assessed article quality using the Mixed Methods Appraisal Tool (2018). We then categorized and compared the success of interventions.

Results: Of the 225 articles screened, 29 studies fit our inclusion criteria. Of the 29 articles, 18 describe findings targeting NHS scale-up interventions, five CHS, four AHS, one NHS/CHS, and one CHS/AHS. Interventions assessed were educational (n=3), policy and systemic (n=3), telehealth (n=2), financial and funding (n=2), expanded screening (n=6), and restructured screening programs (n=7). The evidence from these articles suggests that restructure screening programs, the most documented intervention type, could be the most effective in increasing uptake generally and across HIC and UMIC settings, with no null results.

Discussion: We recommend policies and interventions that restructure screening programs or expand their reach as strong options to allocate resources toward in both high- and low-resource settings, relative to existing intervention types previously attempted. More research pertaining to scale-up, especially in lower-income settings, is necessary, however, to make the most appropriate recommendations.

II. INTRODUCTION

According to the 2019 Global Burden of Disease Studies, hearing loss is the third-highest common cause of years lived with disability (YLDs) with an estimated 43.45 million YLDs, spanning about 1.57 billion people (1). If left unchecked, an estimated 25 percent of the global population, or about 2.5 billion people, will experience some degree of hearing loss by 2050. More than half of these cases, however, are considered preventable with improved implementation of hearing healthcare (2).

Although interventions to address hearing loss exist, such as hearing screenings, hearing aids, and cochlear implants, access is inequitably distributed across geographic, socioeconomic, and racial axes, and is especially poor in lower-income settings (3). In fact, of the 500 million people with hearing loss who would benefit from these and other hearing healthcare services, 80 percent have disproportionately poor access due to residence in lower-income countries (4). This disparity has been well-documented across the literature, highlighting the need for specific "scale-up strategies" in hearing healthcare, or interventions and approaches that can increase the capacity to screen and treat patients. Determining what scale-up strategies are effective in bolstering the access and uptake of these interventions could be beneficial in reducing the global burden of hearing loss and preventing further social and economic injury to local and global communities.

At present, articles promoting individual interventions to improve patient use of, adherence to, and/or satisfaction with hearing devices and hearing screenings are available in the literature, with approaches spanning various disciplines, such as health economics, policy, education, and technology. There is, however, an apparent lack of systematic evaluation between these strategies to determine which approaches are not only most effective, but also most cost-

effective, to scale-up hearing healthcare on a global level. With finite resources available for hearing healthcare within and across countries, it is vital to understand which approaches, according to the evidence, may have the highest likelihood of success in improving patient access so that healthcare systems can reap the greatest benefit from their investments. The implementation of a strategy that lacks evidence to support it may fail and possibly waste resources that could have been better allocated otherwise to improve hearing healthcare uptake. Therefore, a comparison of existing strategies is necessary to advise local, national, and global healthcare infrastructure in reaching patients and redressing the burden of hearing loss.

In 2019, *The Lancet* announced a Commission dedicated to investigating the global burden of hearing loss and strategies for reduction. As part of these efforts, the Duke-Margolis Center for Health Policy, in collaboration with the Duke Global Health Institute, has conducted an extensive systematic review to identify key barriers and facilitators to accessing hearing healthcare interventions, including cochlear implants, hearing aids, neonatal hearing screening (NHS), child hearing screening (CHS), and adult hearing screening (AHS) using the Levesque "Access to Care" Framework (5). The results of this systematic review demonstrate how supply-side considerations, like "Approachability", "Acceptability", "Availability", "Affordability and "Appropriateness", and demand-side capabilities, such as "Ability to Perceive", "Ability to Seek", "Ability to Reach", "Ability to Pay", and "Ability to Engage", can impact access to hearing devices and screenings in both high- and low-income countries . The synthesis of this evidence was used to create targeted policy recommendations to redress present obstacles and bolster interventions that improve patients' and providers' interactions with the health system. To complement this work, additional systematic reviews, such as this review, will identify key scale-up strategies to increase patient access and uptake of hearing healthcare interventions,

specifically cochlear implants, hearing aids, and hearing screenings across age demographics, as well as efficacious approaches to hearing healthcare innovation.

Specifically, this systematic review seeks to synthesize evidence on scale-up strategies to increase use, adherence, and satisfaction of hearing screenings across neonatal, pediatric, and adult populations in high-, upper middle-, lower middle-, and low-income countries. A "scale-up strategy" can be defined as an intervention designed to change metrics related to use, adherence, and/or satisfaction upon implementation. This comparison can then be used to create targeted policy recommendations to advise governments, healthcare systems, and providers alike. Our goal is to create a better understanding of which scale-up strategies should be recommended for implementation to increase the uptake of neonatal, child, and adult hearing screenings across the globe. With improved capacity and access for hearing screening, the true burden of hearing loss can be identified, thus demonstrating the need for increased accessibility to treatment and paving the way towards expansion. This review will consider what strategies could effectively expand hearing screening across all age groups and alleviate the overall global burden of hearing loss, rather than target a specific demographic, such as neonates. Therefore, we will unify existing literature to create policy recommendations to help high- and low-income settings determine how to most effectively employ these strategies to improve their capacity for hearing screening.

III. METHODS

This systematic review follows the guidelines of the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) checklist (Appendix I)(6). It is also included under the same PROSPERO protocol (CRD42020214772). The articles for this systematic review were collected from five health databases: *Embase, Global Health & Global Health Archive, Global Index Medicus, PubMed,* and *Web of Science*. Search terms included but were

not limited to “hearing loss”, “deafness”, “barriers”, “facilitators”, “scale up”, “innovation”, “intervention”, and “screening” (Appendix II). For inclusion in this study, an article must possess all four of the following criteria:

1) The article is related to hearing loss.
2) The article describes a scale-up strategy for improving the uptake of NHS, CHS, and/or AHS, as it relates to use, adherence, and/or satisfaction.
3) The article utilizes primary data.
4) The article exists in English, either in its original form or as a translation.

Articles that did not relate to hearing loss, did not describe scale-up strategies for NHS, CHS, and/or AHS, lacked primary data, and/or were unavailable in English were, therefore, excluded from this study. It should be noted that efforts were made to find translated copies of non-English articles, as well as primary manuscripts associated with conference abstracts, but authors were not contacted due to the quantity of articles considered for review.

Each article that two screeners agreed fit the inclusion criteria then underwent title, abstract, and multiple full-text screenings by a primary reviewer using the platform DistillerSR. An article that advanced through these levels of screening was then over-read by a secondary reviewer, working independently, to ensure it met selection criteria and that all extracted material was accurate (Appendix I). Any conflicts were resolved via discussion or the opinion of a tertiary reviewer.

After initial screenings, the 2018 Mixed Methods Appraisal Tool was applied to assess quality, risk of bias, and certainty, as they concern the articles and their results. This process was completed by two investigators, a primary reviewer who evaluated each article and reported the quality, bias, and certainty of the articles and a secondary reviewer who overread the first

reviewer's work (Appendix IV Table 2). Again, any conflicts were resolved via discussion or the opinion of a tertiary reviewer. Each question for an article's designated study design in the questionnaire was considered on a binary system, with "Yes" equating to 1 and "No" equating to 0. "Cannot tell" was also equated to 0, as we lacked certainty in answering and the response made up a negligible two percent of answers. The average score was taken across each article and then for all articles to determine the risk bias and certainty of the evidence included in the systematic review.

During the last full-text screening, data was extracted for inclusion in analysis (Table 1). First, the scale-up strategy and the sample size from the target population were collected. "Scale-up strategy" refers to any intervention designed to cause a change in use, adherence, and/or satisfaction with a hearing healthcare intervention. To be considered "scale-up", studies must demonstrate such a change by reporting the state of the target population both before and after implementation.

Next, outcomes and effects were collected. In this analysis, "outcome" refers to what the authors of each article intend to measure, along with what they hypothesize will happen to this measure when the scale-up strategy is implemented, relative to previous conditions. In most studies of this kind, the authors anticipate some degree of improvement in uptake. Relatedly, "effect" refers to what *actually* occurred due to implementation, as described by the results of the study. This is usually reported as a quantitative measure. As scale-up refers to a change in uptake, this quantitative estimate should measure how uptake has varied from pre-intervention to post-intervention data collection (Table 1).

The articles were then analyzed with Excel and incorporated into a theory of change model to demonstrate the flow of data extraction (Table 1, Figure 1). We reported each article's country of

origin, along with its World Bank classification as a high- (HIC), upper middle- (UMIC), lower middle- (LMIC), or low-income country (LIC) (*World Bank Country and Lending Groups – World Bank Data Help Desk*, n.d.). Additionally, we included each article's hearing screening focus—NHS, CHS, and/or AHS—as well as any additional description of hearing healthcare intervention, such as hearing devices. Next, in addition to reporting the scale-up strategy, the scale-up strategy was inductively grouped with other similar strategies under a name that best described the approaches found in the data—"Funding/Financial", "Policy/Systemic", "Education", "Telehealth", "Expanded Screening", "Restructured Screening", and "Uses Screening". This last category, "Uses Screening", refers to scale-up strategies that improve uptake of hearing healthcare using NHS, CHS, and/or AHS, rather than improving the uptake of screening itself.

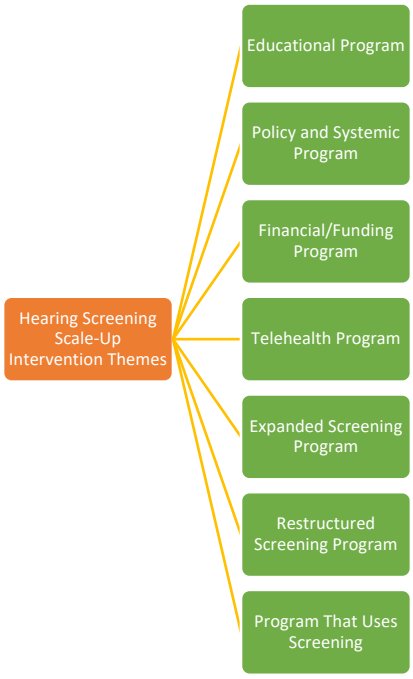
The target population and the sample size within each study are also clearly defined and compared across articles. As the systematic review evaluates scale-up strategies for NHS, CHS, and AHS, data for three distinct populations were collected. "Neonates" or "newborns" refer to patients age zero to 28 days, and, within the articles in this review, may be surveyed by proxy using family members to vouch for the patient. "Children" describe patients age 28 days to 18 years. Similar to neonates, younger members of this population of interest may be surveyed by proxy using family members. Finally, "adults" defines patients age 18 years and older, who may be grouped by veteran status or occupation in the context of studies.

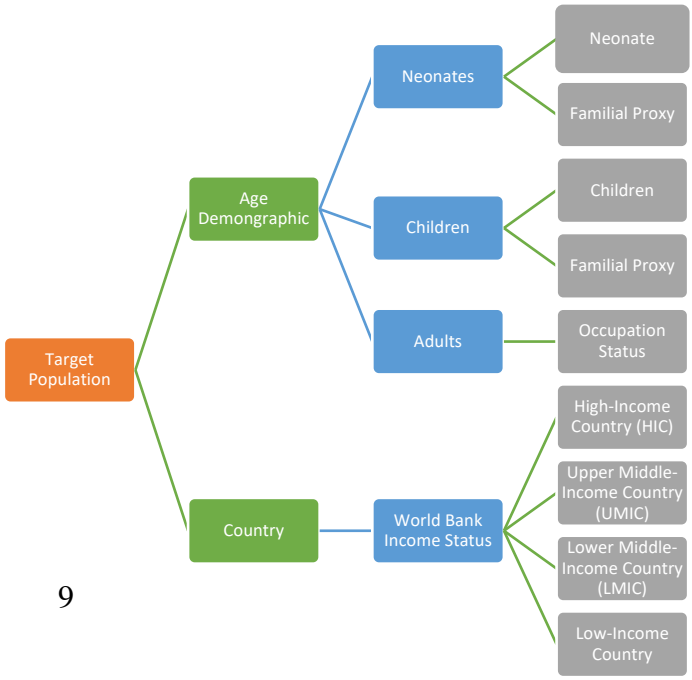
The intended intervention outcome—use, adherence, and/or satisfaction—is also noted as a data item in our collection process. As these terms have been alluded to previously to describe changes in uptake, it is important to clearly define them here. "Use" refers to access and/or consumption of screening services, including measures of coverage, rates of utilization, and

time/age of first access. “Adherence” describes commitment to the screening and diagnostic process and can encompass measures related to referral, follow-up, continuation of screening protocol, and follow-through on recommendations and post-initial screening care. Finally, “satisfaction” refers to fulfillment of needs and expectations for screening services, which is often measured via survey.

Last, data related to the effect/outcome of the scale-up strategy was collected. A successful scale-up strategy, in which uptake increased, was labelled “positive”. An unsuccessful strategy, which decreased uptake, was labelled “negative”. A combination of “positive” and “negative” effects was considered “mixed results”, and no change in uptake was called “null”. Details supporting this categorization were also included as quantitative evidence, including percent changes, p-values, odds ratios, confidence ratios, and other statistical measures. Where applicable, distinctions between sub-groups in the sample were described as well.

The articles evaluated and included were then synthesized to compare NHS, CHS, and AHS scale-up interventions altogether on the bases of type of intervention, outcome, and effect, relative to their country classification. A glossary of the terms above can be found in Appendix III.





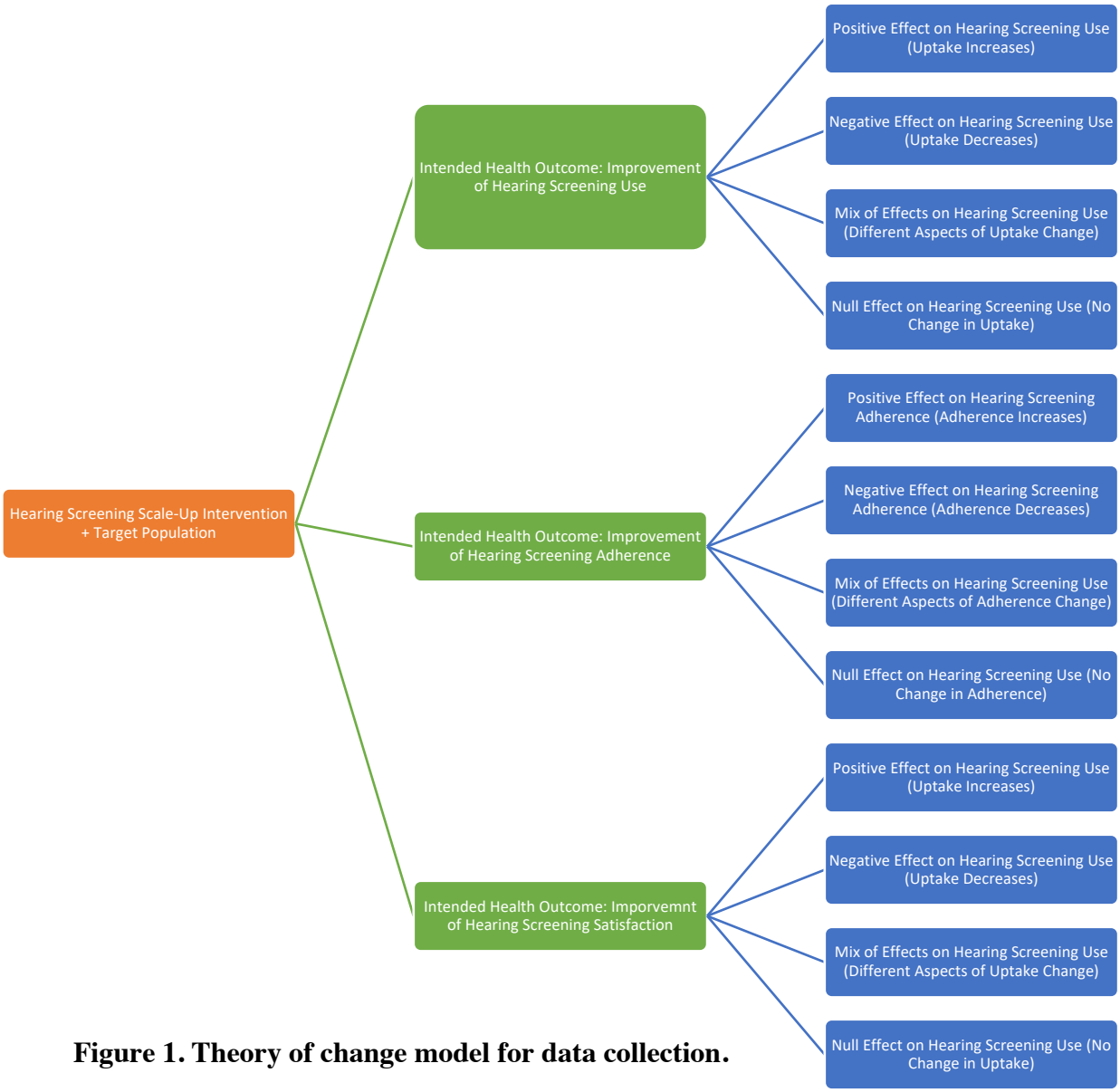


Figure 1. Theory of change model for data collection.

IV. RESULTS

a. OVERALL RESULTS

Of the 225 articles screened, 29 studies fit inclusion criteria and have been analyzed for this systematic review. This pool contains 18 NHS-only articles, five CHS-only articles, four AHS-only articles, one NHS/CHS articles, and one CHS/AHS articles. These studies represent one

LIC, Malawi, one UMIC, Brazil, two LMICs, Vietnam and India, and 15 HICs, Ireland, Israel, France, Italy, the United States, Australia, Canada, New Zealand, Taiwan, Slovakia, South Korea, Denmark, Poland, the Netherlands, and the United Kingdom. Moreover, this collection is comprised of one qualitative study, one quantitative randomized control trial, 16 quantitative non-randomized studies, 10 quantitative descriptive studies, and one mixed methods study (Appendix IV, Table 2).

Across the included articles, 25 had positive effects on scaling-up hearing screening, while only two articles had mixed effects and two articles had null/no effect. There were no articles that had a negative effect on scale-up. These effect categorizations were determined by the quantitative data collected by each article, which demonstrated if the outcome was achieved or not. This included p-values, percent changes, odds ratios, and confidence intervals. Notably, this data showed disparate effects of scale-up in sub-groups in four articles (Table 1).

The 29 articles span seven intervention themes (Table 1). "Education" describes approaches to alter patient knowledge and understanding of screening. "Policy and Systems" refers to interventions that promote screening uptake and accessibility by redressing healthcare or political infrastructure. "Telehealth" uses web- and digital-based programs for patient screening as a way to change capacity and uptake. "Financial/Funding" interventions seek to modify the financial barriers that may prevent patients from seeking care. "Expanded Screening" approaches alter the reach and capacity of existing services, commonly through universalization. Similarly, "Restructured Screening" interventions utilize existing screening programs and revise their infrastructure and protocol to modify capacity and uptake. Finally, "Uses Screening", again, refers to the utilization of screening to alter the capacity and uptake of another hearing healthcare intervention, such as hearing devices (Appendix III; Appendix VI, Table 6).

The distribution of articles and effects across these categories was also evaluated (Table 1). Of the "Education" interventions, which includes health promotion lectures and patient navigation, four articles across four HICs documented effects on use and/or adherence, of which three were positive and one was null. Within "Policy and Systemic" interventions, like legislature and protocol changes, two articles from HICs and one from an LMIC demonstrated positive effects on use and/or adherence. The cases that described "Telehealth" interventions, such as screening applications, showed disparate effects, as one HIC article showed mixed results and one LMIC article showed positive results related to satisfaction. "Financial/Funding" Interventions accounted for two articles, both in HICs and with positive effects on use. Under "Expanded Screening" interventions, 9 articles described universalization or increased geographic reach in different HICs and one LIC, Malawi. Of these, all but two had positive effects on use, adherence, and/or satisfaction. The two non-positive articles were null effect and mixed results, respectively. Finally, seven studies from four HICs and one UMIC documented "Restructured Screening" interventions, such as multistage screening programs, all with positive effects on use and/or adherence. Also, of note, two articles fit the "Uses Screening" category, as one uses NHS as a means to scale-up cochlear implants and the other uses AHS as a means to scale-up hearing aids. Both studies are from HICs, have positive effects, and inherently lead to improved screening uptake as well. All information related to this evaluation, as well as the results of individual studies, can be found in Table 1.

The 2018 Mixed Methods Appraisal Tool (MMAT) was applied to each article in the pool and analyzed quantitatively with binary measures and averages. The average score (out of 1) for the articles was 0.94, indicating that, in general, the articles were of higher quality, with low risk of bias and high certainty of the evidence. The individual articles range from 0.29 (1 articles),

revealing of lower quality with high risk and low certainty, to 1 (n=22), the highest quality with the lowest risk and highest certainty according to the MMAT. Within this range, two articles are 0.71 and four articles are 0.86 (Appendix IV, Table 2).

In addition to analyzing the distribution of articles and interventions across themes, populations, and countries, it is also relevant to stratify the studies across uptake categories—use, adherence, and satisfaction. It should be noted, however, that some articles describe more than one of these categories, prominently use/adherence.

Table 1. Synthesis of Themes and Outcomes

	Effects			
Type of Scale Up Intervention	**Outcome: Use**	**Outcome: Adherence**	**Outcome: Satisfaction**	**Effect Type**
Education	Education interventions have been demonstrated to increase hearing screening utilization. For example, an Israeli educational intervention improved use from 19 percent to 49 percent over two years in a healthcare setting (7). Similarly, an Australian educational program improved hearing screening uptake in farmers by 26 percent (8).	Continuation of hearing healthcare after initial screening improved in response to educational intervention implementation in both LIC and HIC settings. In Malawi, follow-through with referrals improved from 3 percent to 53 percent while an analogous positive effect was documented in the Netherlands of 11 percent improvement (9,10). A study in the US also recorded a similar phenomenon with only 7.4 percent of patients nonadherent to follow-up instead of 38.2 percent initial nonadherence. Additionally, a decrease in time to follow-up was reported, from 105.9 days to 67.9 days (11).		Positive
Policy and Systems	Interventions involving policy and health systems improved both screening coverage and age of access. In Italy, coverage increased from 29.2 percent to 78.3 percent over nine years across, with analogous increases observed across individual regions surveyed (12). In the US, implementation led to a decrease in the average age of screening from 7.9 weeks to 5.1 weeks (13). A similar phenomenon was observed in Vietnam, with a decrease to less than 24 months on average (14).	Policy and health systems interventions had a notable improvement on adherence to screening and hearing healthcare protocols in both HICs and LMICs. In the US, loss to follow-decreased from 18 percent to 7 percent, while, in Vietnam, the number of children who progressed to receiving treatment increased from 36.4 percent to 61.2 percent (13,14). Moreover, in Vietnam, fewer families attempted to treat hearing loss independent of the health system, decreasing from 63.6 percent to 38.8 percent (14).		Positive
Financial/Funding	Interventions that remove financial barriers for hearing screening improve screening coverage. Such a positive effect was observed in Taiwan, with an increase from 87.34 percent to 99.64 percent, and in South Korea, with an increase from 50 percent to 70 percent (15,16).	Financial interventions improved follow-up after initial screening, with a Taiwanese study showing an increase from 40.74 percent to 100.00 percent after implementation (15).		Positive
Telehealth		Telehealth interventions were effective in improving continuation of treatment, specifically in an LMIC setting. In an Indian setting, the adaptation of ABR as an application improved follow-up by 11 percent, relative to in-person screening (10).	Telehealth interventions had no impact in HICs. In fact, in a United States study, patients had no preference between Telehealth and in-person screenings. (17).	Mixed
Expanded Screening	Although expanded screening interventions are well-documented, they have variable impact on uptake of hearing screening. Screening coverage increased in Italy from 34.4 percent to 93.8 percent over six years, in New Zealand from 86 percent to 91 percent over four years, in Slovakia from 42 percent to 95 percent in two years, and in Denmark from 88.6 percent to 94.8 percent in seven years (18–21). Similarly, the number of patients receiving screenings in a study from the United States study quadrupled during a four-year period (22). In Italy, the mean ages of screening and diagnosis also improved over two years from an average of 32 months to less than a year of age (21). On the other hand, a study from the Netherlands reported no change in hearing screening coverage when UNHS services were paired with home metabolic screening (23). Moreover, in Poland, the increase in the number of patients screened was not correlated in a change of health behavior but, rather, reflective of population growth (24).	The impact of expanded screening interventions was also variable relative to the continuation of care. In Denmark, follow-through increased from 89.5 percent to 99 percent over eight years. Additionally, the time to diagnosis post-initial screening decreased from 3.6 months to 0.7 months over seven years (20). Conversely, in the Netherlands, complete participation in screening protocols in the home improved by more than 10 percent compared to in the clinic but this margin was not much greater nor significant when paired with home metabolic screening (23).	Expanded screening interventions had positive impacts on patient satisfaction. For example, 97.5 percent of hearing specialists, 82.5 percent of pediatricians, and 89.5 percent of mothers preferred UNHS over a targeted screening program in the US (25).	Mixed

Restructured Screening	Restructured screening interventions had various positive impacts on the uptake of hearing screening. These programs were observed to improve screening coverage in Ireland from 0 percent to 98 percent and in Italy from 89.8 percent to 92 percent in six years (26,27). Similarly, a study from the US improved the screening coverage in the first month of life from 24 percent to 61 percent and in the third month of life from 82 percent to 96 percent over a two-year period (28). Restructured screening interventions also reduced age of screening from 63.6 days to 35.8 days in the US over two years and from 48.66 days to 24.53 days in Brazil over eight years (28,29). Finally, in France, increased use of a restructured program, from 14.9 percent to 75.8 percent, reduced the need for referral from 2.9 percent to 0.6 percent (30).	Interventions that restructured existing programs improved measures of adherence in both HICs and UMICs. Studies from the US saw increases in follow-up from 36 percent to 91 percent and from 61 percent to 75 percent, respectively (31,32). Similarly, a Brazilian study reported an improvement in adherence by families from 32 percent to 85 percent over nine years (29).		Positive
Uses Screening	Interventions that use screening as a means to improve uptake of additional hearing healthcare. In Australia, utilizing screening early in life decreased the age of hearing aid fitting and cochlear implant survey in Australia (33). In Canada, earlier use of screening correlated with earlier diagnosis and use of hearing aids (34). Finally, in the US, hearing aid use was up to 7.4 percent in patients who were screened in the last year compared to those who had not, at 3.3 percent (35).	The employment of screening was also shown to successfully promote the adherence to other hearing healthcare interventions. For example, in Canada, use of hearing screening was correlated to longer routine hearing aid wear, compared to those who did not use hearing screening (34).		Positive

b. USE

As previously mentioned, "use" describes access to and/or uptake of screening services. This could be described in the literature with outcomes such as utilization of screening, screening coverage, and time or age of first use. Of the articles reviewed, 23 articles pertaining to use fit the inclusion criteria. Of these, there are 15 NHS articles, two CHS, four AHS, and one CHS/AHS. This subsect of the literature encompasses one LMIC, Vietnam, one UMIC, Brazil, and 13 HICs, Ireland, Israel, France, Italy, the United States, Australia, Canada, New Zealand, Taiwan, Slovakia, South Korea, Denmark, Poland, and the Netherlands. Only four articles of the 23 had non-positive effects—two with mixed results and two with null effects. All intervention categories except Telehealth were considered and attempted as a means to improve use (Table 3).

The category pertaining to "Education" contains two interventions, one of which has positive results. A series of health-promotion lectures for AHS for older adults in Israel improved patient screening rates from 19 percent to 49 percent in two years. On the other hand, the other study describes an unsuccessful enhanced hearing health education program for older

AHS patients in the Netherlands. More specifically, this intervention failed to improve attendance to a general practitioner, indicative of a lack of change in screening coverage (7, 36).

There are also three articles related to "Policy and Health Systems" interventions, each with a positive effect. The first study, from Italy, describes legislation to prescribe and promote UNHS across the state, which yielded a steady increase in screening coverage over a nine-year period from 29.3 percent to 78.3 percent. In the second study, policy and benchmark changes for NHS at the hospital-level in the United States demonstrated a notable decrease in age of initial screening from nearly eight weeks to five weeks of age. Finally, a Vietnamese study explored the impact of health-system level changes to early hearing detection interventions (EHDI), specifically for CHS, and found that this also decreased age of initial screening to less than 24 months of age (12,14,37).

Only two articles describe "Financial/Funding" interventions, both with effective scale-up. One article describes a government-funded UNHS program in Taiwan from that increased screening coverage from 87.34 percent to 99.64 percent. The other study implemented a government-subsidized financial aid program for low-income UNHS patients in South Korea to also improve screening coverage from 50 to 70 percent (15,16).

The category "Expanded Screening" is the most common strategy to improve use. Of the eight articles, three explore the impact of making NHS programs universal or nationwide in Slovakia, Denmark, Italy, respectively. These interventions demonstrated significant increases in NHS coverage across each state, and, in Italy, the average age of identification decreased from 32 months to less than a year of age. A similar intervention for universal CHS in preschools was implemented in New Zealand, with an analogous increase in screening coverage from 86 percent to 91 percent as a result. In this study, however, there was lower screening coverage for Māori

and Pacific indigenous people, as well as for children from socioeconomically disadvantaged backgrounds, from large households, in rented homes, with younger mothers, with worse health conditions, and with greater geographic mobility, suggesting that universalized preschool screenings may an effective, but inequitable, intervention. Additionally, in Australia, a program to increase the reach of CHS and AHS services showed a two-fold increase in screening coverage in rural areas and underserved populations (18–22).

The strategy of "Expanded Screening", however, lacks consistently positive effects. In fact, one study demonstrated no effect on its outcome and two interventions have mixed effects. Unlike the previously described articles in this category, a study from Poland demonstrated that improvements in nationwide NHS programming uptake were caused by proportional increases in birth rate, rather than scale-up, meaning there was no net effect by the intervention. Additionally, in the Netherlands, UNHS administered in the home, alongside home metabolic screening programs, demonstrated no change in actual hearing screening coverage but improved participation in the larger metabolic screening program. A third study in Australia, which focused on expanding AHS to the farming community, showed an increase in screening service utilization, but by only one community, suggesting some degree of uncertainty in the actual effect of the strategy for unknown reasons (8,23,24).

Furthermore, there are five articles "Restructured Screening" interventions that describe attempts to improve use, all of which have positive effects. Of these articles, two describe revamping existing NHS programs to include a two-stage protocol in Ireland and Brazil, respectively. In Ireland, coverage was measured to be 98 percent after an intervention introduced hearing screening to the study population. In Brazil, the mean age of newborns at screening was halved from 48.66 days to 24.53 days of age over eight years. In France, deferring preliminary

screening before the age of one month was also effective in improving screening coverage fivefold from 14.9 percent screening coverage to 75.8 percent. An Italian intervention that incorporates a well-infant nursery hearing screening program for newborns also demonstrated a modest but statistically significant improvement in screening coverage from 89.8 percent to 92 percent. Finally, a study from the United States, which targeted outpatient prescreening through the WIC program, led to a decrease in the mean age of hearing loss diagnosis for infants from 63.6 days of age to 34.8 days (26–30).

It is also pertinent to discuss any intervention that "Uses Screening" as means to improve the uptake of other forms of hearing healthcare. In the literature, there are three articles that specifically accomplish this relative to "Use". All three studies use screening, either NHS or AHS geared toward veterans, to improve hearing aid use and fitting in Australia, Canada, and the United States, respectively, with positive effects accomplished by each intervention. The Australian study also demonstrates promising results with improving utilization of cochlear implants with NHS as well (33–35).

c. ADHERENCE

As defined in the Methods section, "adherence" refers to commitment to the screening and diagnostic process. This includes interventions that promote referral, follow-up, continuation of screening protocols, follow-through on recommendations, post-initial screening care and more. In this systematic review, 12 articles were included that described attempts to change adherence. Of these 12, there are seven NHS articles, four CHS, and one NHS/CHS. The 12 articles, nine of which detail interventions in HICs, including the United States, Canada, Denmark, the Netherlands, the United Kingdom, and Taiwan, along with one LIC, Malawi, one

UMIC, Brazil, and two LMICs, India and Vietnam, describing adherence as a measure of uptake were overwhelmingly positive, with only one article having mixed results (Table 4).

Only two of these articles describe adherence with respect to "Education" interventions. One study describes a patient-navigation intervention for NHS and CHS in the Appalachian region of the United States, which lowered patient nonadherence from 38.2 percent to 7.4 percent. Additionally, the follow-up period was decreased upon implementation of patient navigation from 105.9 days after birth to 67.9 days. The other article describes a multi-component educational model for CHS in Malawi, the only LIC represented in this review. This intervention improved patient screening follow-up from 3 percent to 53 percent. Interestingly, despite differences in location and associated income, both interventions demonstrated positive results in increased continuation of care (9,11).

The intervention category "Policy and Health Systems" also contains two adherence articles. The first study describes policy and benchmark changes at the hospital level for NHS in the United States that improved loss to follow-up from 18 percent to 7 percent. The second article details health system-level changes to EHDI for CHS in Vietnam, an LMIC, yielding an increase in patient progression to treatment from 36.4 percent to 61.2 percent. This coincides with a decrease in the number of patients who are nonadherent and seek informal remedies, from 63.6 percent to 38.8 percent. Loss to follow-up decreased and progression to hearing loss treatment increased in both studies (13,14).

One article uniquely explores a "Financial and Funding" intervention to improve adherence. In Taiwan, a government-funded program for UNHS, which specifically covers the cost of automatic auditory brainstem response screening, positively impacted adherence and uptake of hearing screening. The article described a significant decrease in the need for referral

compared to patients screened without this financial coverage from 2.82 percent to 0.95 percent. This may mean more individuals were able to adhere to screening protocol regardless of hearing status or perceived need. Additionally, this intervention yielded improvements in follow-up, increasing form 40.74 percent to 100.00 percent (15).

Only one article explicates the effects of a "Telehealth" intervention on adherence. An Indian study utilized telehealth diagnostic auditory brainstem response (ABR) follow-up for CHS. This intervention had a significant improvement in follow-up of 11 percent, relative to conventional in-person methods, due to the accessibility of ABR through a digital platform. It is also worth noting that this study was set in villages in South India, indicative of success in an especially low-resource settings (10).

Two articles describe changes to adherence due to "Expanded Screening" interventions. The first study focuses on nationwide NHS programming in Denmark, which decreased the time infants progressed through hearing screening protocols to receiving a diagnosis from 3.5 months to 0.7 months. The second study evaluates home screening for UNHS using home intake and home metabolic screening programs in the Netherlands. The authors of this study report mixed results for this intervention due to differences in participation across the treatment groups participating in the study. More specifically, home intake had stronger adherence than well-baby clinics, at 86.9 percent compared to 75.2 percent, but weaker compliance than home metabolic screening at 88.9 percent (20,23).

The most articles fall within the category of "Restructured Screening", with three articles from the United States and one from Brazil. The first American study describes using a two-tiered program for CHS, specifically in preschools. This had a positive effect, significantly improving follow-up from 36 percent of patients to 91 percent of patients after implementation

relative to the control. The second article from the United States pertains to targeted outpatient rescreening for NHS among patients enrolled in the WIC program. This was also successful, as the loss to follow-up rate was reduced to 9.1 percent for the WIC group, compared to 28.7 percent for the non-WIC group. The third American article explores the addition of accommodations for patients and their families to existing UNHS programs, which had increased the return rate to 61 percent after discharge and boosted follow-up to 75 percent. Finally, in Brazil, a study describing a two-stage protocol for outpatient NHS displayed success, with adherence more than doubling from 32 percent to 85 percent in less than a decade (28,29,31,32).

It is also worth noting that one study describes the use of NHS to identify and early fit hearing aids in Canadian neonates. This intervention, which falls under "Uses Screening", has a positive effect in scaling-up hearing healthcare, leading to earlier access to hearing aid treatment at an average age of 6.3 months, relative to other infants with hearing loss at an average age if 34.5 months (34).

d. SATISFACTION

As previously described, "satisfaction" describes the fulfillment of needs and expectation for screening services, often evaluated via survey. In this systematic review, there are only two articles that describe interventions to improve patient satisfaction as a means to improve uptake, one NHS article and one AHS article. Both are from the United States but describe satisfaction with disparate effects across different demographic groups (Appendix V, Table 5).

The NHS article described an "Expanded Screening" intervention, specifically comparing a universal NHS (UNHS) program to a more targeted one. The researchers administered a questionnaire to 95 post-partum mothers randomly selected from an obstetric ward, as well as 80 hearing healthcare providers, to gauge their satisfaction with UNHS relative to targeted

programs. The survey demonstrated that most mothers (89.5 percent) and providers (97.5 percent of hearing specialists, 82.5 percent of physicians) favored UNHS over targeted screening, with notable variation between mothers and providers and between kinds of providers (25).

Conversely, the AHS article focused on a "Telehealth" approach, in which audiometric screenings were performed in a clinical setting using tablet-based applications. In this study, 107 adult patients that had been referred for audiometric testing were surveyed to determine satisfaction with the tablet-based screening, relative to traditional screening methods. A survey administered to the patients after the intervention demonstrated that he application had no effect on patient satisfaction and uptake, as patients showed no preference for the telehealth intervention relative to conventional audiometry (17).

V. DISCUSSION

a. ANALYSIS

We synthesized existing evidence on scale-up strategies for NHS, CHS, and AHS to then make policy recommendations and appropriately allocate scarce resources.

Education interventions were only partially successful, have limited literature to support their implementation, and have not been documented outside of HICs. Not only do education interventions lack evidence related to impact on patient satisfaction, but it also had mixed results relative to use, with data showing improved utilization by only some patients. Education interventions, however, were entirely positive in improving adherence to screening, especially as it pertains to uptake of referral and follow-up. Despite the high quality of the articles available, the inconsistent support in the literature of only 5 articles, primarily from HICs rather than equally dispersed across income levels, education interventions may not be a suitable entry point for policy recommendation and resource allocation. It should be noted, however, that the only

LIC intervention in this review pertained to hearing screening education in Malawi, with positive results, so this approach may be a promising option for lower-resource settings and may warrant more research in LICs.

Both policy and funding interventions were successful in the instances documented in the literature, but these approaches have similar limitations to education strategies. Policy and health system interventions encompass only two HICs and one LMIC in the three articles that explore its outcome, but all three articles have positive effects for use and/or adherence. This category also fails to explore the impact on patient satisfaction, but notable improvements were seen in age of screening, coverage, and loss to follow-up. Financial and funding interventions only accounted for two articles in this review, both in HICs and positive effects on use and adherence, especially as they pertain to improving coverage and need for referral. They also lack description of impact on satisfaction for low-income settings. While the strictly positive results make these two scale-up strategies promising options, the lack of evidence does not make these approaches strong contenders at the present time.

Telehealth, also under-researched with only two articles described in this review, showed success in an LMIC setting, but not an HIC setting, suggesting it may be an inequitable intervention to implement. While this category did explore the impact on patient satisfaction, patients in an HIC showed no improved affinity to the technology. It was, however, associated with improved follow-up in an LMIC. With more research, telehealth may prove to be a useful tool for LMICs to improve uptake of hearing screening, but presently much uncertainty still surrounds the effectiveness of this strategy, including questions of equitable access to the necessary technology and internet.

Expanding screening reach was the second-most attempted and successful intervention type, but was also only described in HICs, one of which experienced no impact. Of the nine articles studied, all but two had a positive effect on use, satisfaction, and/or adherence. Of the non-positive articles, one intervention had no effect and one had mixed results. Additionally, it is also important to note that most of these articles specifically pertain to implementing UNHS programs. While there is the most evidence for this strategy, especially across all three categories of uptake, the lack of documentation in LMICs is a cause for concern about the adaptability of this approach on a global scale.

Finally, the evidence from these articles suggests that restructured screening programs, the second-most documented intervention type, could be effective in increasing uptake generally and across HIC and UMIC settings, with few adverse results. Simply put, restructured screening programs have a large research base and no reported negative results. Similar to expanded screening, restructured programs have no documentation LMIC, which could be a problem for widespread implementation.

Despite the difference in aim, it is interesting to also consider the approaches that used hearing screening as a means to scale-up other forms of hearing healthcare. While only three articles fall under this category, all three yielded positive effects in improving use and adherence for hearing devices in HICs, inherently increasing the use and adherence of screening when employed as a prerequisite for treatment. This indirect approach has limited support, as this category does not contain interventions in lower-income settings or studies that measure changes in satisfaction. Nevertheless, it could be an effective way to improve hearing screening uptake while also treating patients with hearing loss, but this is outside the scope of this review.

b. POLICY IMPLICATIONS

While each intervention type brings unique strengths, specific evidence-based strategies should be considered the first line of prevention in addressing the global burden of hearing loss, reducing hearing healthcare disparities, and improving access to global hearing screening. Furthermore, with limited and disparate resource availability, it is necessary to create recommendations and prioritizations for regional, national, and international health systems to adopt. Thus, from this analysis, we recommend policies and interventions that restructure screening programs or expand their reach as an effective strategy for resource allocation in both high- and low-resource settings, relative to existing intervention types previously attempted. Although such interventions have not been documented in lower-income countries, they would target existing infrastructure to emphasize physical accessibility and reachability. As expanded and restructured screening articles make up approximately half of this review, there is significant evidence to suggest a higher likelihood of successful implementation to improve use and adherence especially. Policies that dictate, enforce, and support mandatory hearing screenings, as well as incentivize multi-tier protocols, would improve availability of screening while also appropriately using resources as needed.

Because expanded and restructured screening interventions, despite being strong candidates for scale-up strategies relative to the other approaches, lack evidentiary support for success in UMICs and LMICs, additional approaches should be recommended either in addition to expanded and restructured screening, or as a second line of defense. Specifically, the literature, albeit in a limited capacity, demonstrates that education and telehealth can be successful for improving adherence in lower-income settings. With assistance from the global community or improved access to resources, this should also be considered for implementation, thus necessitating policy regarding telemedicine, privacy, and internet access.

The above recommendations do not dismiss any one strategy as being "ineffective" or "unworthy" but, rather, attempts to denote which may have the most success in the face of constraint. Of course, educating patients about hearing loss and screening or providing funding to improve patients' ability to pay or redressing the health infrastructures that house this programming would be beneficial. Health literacy, affordability, and freedom from system-level barriers are vital for healthcare accessibility. When there are limited resources to alleviate a global issue, however, we must recommend what is the most cost-effective and most likely to be efficacious.

a. LIMITATIONS

Various limitations in data collection and data availability significantly inhibit our ability to accurately report and draw inferences regarding existing success in global hearing screening scale-up and related policy recommendations to improve access and uptake. Nevertheless, the synthesis of existing data can be beneficial to our comprehension and in developing a path forward.

First, as the article collection phase for this project was completed by July 2021, the scope of scale-up strategies analyzed is temporally limited, mostly to the conditions of a "pre-pandemic" world. In light of COVID-19 pandemic, the strategies reported and inferences made may not be the most accurate portrayal of the present needs, challenges, and access related to hearing screening in a "post-pandemic" world. COVID-19 has exacerbated inequity in healthcare infrastructure across the world, but it is unknown to what extent. Thus, this systematic review and its recommendations do not reflect the effects of the pandemic as on hearing healthcare use, adherence, and satisfaction, creating potential for erroneous comprehension and intervention.

Another noteworthy limitation is based on generalizability regarding implementation of the policy recommendations. In the proposed policies, there is an implicit belief that a given country or region has the necessary physical and human capital to implement change, when this is simply not the case. Not all technological interventions can be used successfully in all settings, whether it is because of climate, access to electricity, ability to maintain machinery, or other considerations. Similarly, this scarcity applies to human knowledge and personnel, with limited individuals to run tests and then understand and report accurate results. While community health workers could be an asset in this situation, issues related to compensation, training, and supervision then arise. These shortages in resources are often more severe with decreasing income in a country or region, meaning that LMICs are disproportionately affected by these challenges while also bearing a disproportionate share of the need. In order for these policy recommendations to be implemented properly in the regions that need this change the most, steps must be taken to equip health systems with the resources necessary to actually and successfully scale-up hearing screening and subsequent care. Until then, these findings may not be generalized to all places.

More limiting to this review than the data that *could* be collected was the data that *could not* be collected. Stark gaps exist in the literature that prevent a complete understanding of global hearing loss and, therefore, the presentation of the most pertinent policy recommendations. More specifically, the literature on scale-up strategies is skewed toward HICs, as only one LIC, two LMIC, and one UMIC were represented in this review, compared to 15 HICs. This could be due to HICs' greater availability of funds, technology, personnel, and knowledge for hearing healthcare and hearing loss research, as well as an increased capacity to allocate these resources to hearing loss relative to other morbidities when compared to UMICs, LMICs, and LICs. Thus,

recommendations made here, which are based on the existing literature, may not be completely appropriate or feasible in all regions, or countries. This could likely be extrapolated to comparison between urban and rural settings within countries, as similar inequities exist along axes of income on a more microcosmic scale.

It is also worth noting that there is a large skew in the literature towards NHS, likely due to a combination of resource availability, ease of diagnosis, and social misconceptions about the demographics affected by hearing loss. More specifically, in most sociocultural settings, assumptions are made about commonality of neonatal, child, and adult hearing loss, with impairment in the elderly being an inevitability and not "necessitating" screening and birth defects demanding greater emphasis on EHDI. Thus, this imbalance in the literature indicates that these policy recommendations may not be as feasible or effective in screening children or adults and scaling up CHS and AHS, respectively, as how programs need to target families with infants may be different from how they need to target families with older children or adults in any phase of life.

Moreover, there is a strong bias toward "Use" and "Adherence' articles relative to studies that describe "Satisfaction". This could be due to more subjective measurements that can be employed to evaluate use and adherence or possibly reflective of the need for interventions that increase access over the desire for interventions from which patients can choose based on personal preference. While these are true, studies that analyze patient satisfaction should not be overlooked. In fact, we should strive toward a future in which patients are able to choose and pursue the method of access that is most appropriate for their circumstances. Thus, further research is needed in evaluating intervention success.

Few articles in the literature also explore screening outside a healthcare setting. For example, there seems to be a gap in the literature regarding NHS for newborns not delivered in a hospital or clinic, which is particularly common in lower-income settings. This further detracts from a true understanding of global conditions relative to access and uptake, as it is infeasible in some settings for health centers to be conduits of hearing screening immediately after birth.

Finally, this analysis was completed on the global- and country-levels, meaning we looked only at the country reported, rather than the specific regions of interest. This means lower-income settings within an HIC were not distinguished. In future reviews, it could be beneficial to evaluate scale-up interventions specifically in low resource settings in HICs to consider the influence of this specific phenomenon of disparity on uptake, adherence, and satisfaction.

To best redress the burden of hearing loss and improve access and uptake to global hearing screening, we must, therefore, call for and improve support for increased research efforts related to hearing loss and hearing screening, especially in lower income settings and for non-newborn populations. Without these valuable additions to our knowledge, we fail to have complete insight into the burden of disease and have a greater likelihood of misallocating resources due to assumptions made from limited literature.

VI. CONCLUSION

This systematic review sought to create the most accurate depiction of hearing screening scale-up strategies in the present global health sphere. With this information, policy recommendations could be created in an attempt to alleviate the global burden of hearing loss with improved screening uptake. After evaluating NHS, CHS, and AHS articles from across high- and low-income settings and grouping them into categories based on approach, it is clear that expanded and restructured screening programs are not only the best documented strategy,

but also the most successful approach in the literature. These two strategies, however, fail to reflect changes in uptake in lower-resource settings, but this is the case across all axes of analysis in this review. Across categories, interventions, and the literature itself, there is a stark gap in knowledge related to hearing screening scale-up in lower-income regions for both children and adults. While this review creates a potential entry point for scale-up intervention on a global scale, it is apparent that additional research must be pursued to enable identification of the most appropriate strategies and policies for HICs, UMICs, LMICs, and LICs alike and to allocate resources with greater certainty.

VII. REFERENCES

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VIII. APPENDIX I – PRISMA CHECKLIST

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	

Section and Topic	Item #	Checklist item	Location where item is reported
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	
Study characteristics	17	Cite each included study and present its characteristics.	
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	
DISCUSSION			

Section and Topic	Item #	Checklist item	Location where item is reported
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	
	23b	Discuss any limitations of the evidence included in the review.	
	23c	Discuss any limitations of the review processes used.	
	23d	Discuss implications of the results for practice, policy, and future research.	
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	
Competing interests	26	Declare any competing interests of review authors.	
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

For more information, visit: http://www.prisma-statement.org/

IX. APPENDIX II – SEARCH TERMS

Topic: **What are the barriers to, and the innovating strategies of scaling-up hearing healthcare interventions?**
Searcher: SJK
Date: 7.6.2020
Database (including vendor/platform): PubMed (NLM)

Set		Results
1	"Hearing Loss"[Mesh] OR “hearing”[tiab] OR deaf[tiab] OR deafness[tiab]	135361
2	"Hearing Tests"[Mesh] OR "Hearing aids"[Mesh] OR "Neonatal Screening"[Mesh] OR "Prostheses and Implants"[Mesh] OR "Self-Help Devices"[Mesh] OR "assistive technology"[tw] OR "assistive technologies"[tw] OR "assistive device"[tw] OR "assistive devices"[tw] OR Intervention[tiab] OR interventions[tiab] OR screening[tiab] OR screenings[tiab]	1980566
3	"Health services accessibility"[Mesh] OR "Health policy"[Mesh] OR "health planning"[Mesh] OR healthcare[tiab] OR "health care"[tiab]	933494

4	Access[tiab] OR accessibility[tiab] OR uptake[tiab] OR scale[tiab] OR "scale up"[tiab] OR "scaling up"[tiab] OR "scale-up"[tiab] OR adopt[tiab] OR adopted[tiab] OR adopts[tiab] OR barrier[tiab] OR barriers[tiab] OR strategy[tiab] OR strategies[tiab] OR strategic[tiab] OR innovative[tiab] OR innovation[tiab] OR implement[tiab] OR implementing[tiab] OR implemented[tiab] OR implements[tiab] OR adoption[tiab] OR improvement[tiab] OR improve[tiab] OR improved[tiab] OR improves[tiab] OR improving[tiab] OR increase[tiab] OR increasing[tiab] OR increases[tiab] OR increased[tiab] OR reduce[tiab] OR reducing[tiab] OR reduces[tiab] OR reduced[tiab] OR decrease[tiab] OR decreased[tiab] OR decreasing[tiab] OR decreases[tiab] OR stigma[tiab] OR stigmas[tiab] OR cost[tiab] OR costs[tiab] OR burden[tiab] OR training[tiab] OR development[tiab] OR develop[tiab] OR developing[tiab] OR developments[tiab] OR developed[tiab] OR develops[tiab]	12883956
5	1 AND 2 AND 3 AND 4	1211
6	NOT (animals[mh] NOT humans[mh])	1211
7	NOT (Editorial[ptyp] OR Letter[ptyp] OR Comment[ptyp])	1199
8	+ grey lit, not RCTs, guidance/white papers	

Database (including vendor/platform): Embase

Set		Results
1	'hearing impairment'/exp OR hearing:ti,ab OR deaf:ti,ab OR deafness:ti,ab	193409

2	'hearing test'/exp OR 'hearing aid'/exp OR 'newborn screening'/exp OR 'prostheses and orthoses'/exp OR 'self help device'/exp OR "assistive technology":ti,ab,de,tn OR "assistive technologies":ti,ab,de,tn OR "assistive device":ti,ab,de,tn OR "assistive devices":ti,ab,de,tn OR Intervention:ti,ab OR interventions:ti,ab OR screening:ti,ab OR screenings:ti,ab	2423418
3	'health care access'/exp OR 'health care policy'/exp OR 'health planning'/exp OR healthcare:ti,ab OR "health care":ti,ab	993088
4	Access:ti,ab OR accessibility:ti,ab OR uptake:ti,ab OR scale:ti,ab OR "scale up":ti,ab OR "scaling up":ti,ab OR scale-up:ti,ab OR adopt:ti,ab OR adopted:ti,ab OR adopts:ti,ab OR barrier:ti,ab OR barriers:ti,ab OR strategy:ti,ab OR strategies:ti,ab OR strategic:ti,ab OR innovative:ti,ab OR innovation:ti,ab OR implement:ti,ab OR implementing:ti,ab OR implemented:ti,ab OR implements:ti,ab OR adoption:ti,ab OR improvement:ti,ab OR improve:ti,ab OR improved:ti,ab OR improves:ti,ab OR improving:ti,ab OR increase:ti,ab OR increasing:ti,ab OR increases:ti,ab OR increased:ti,ab OR reduce:ti,ab OR reducing:ti,ab OR reduces:ti,ab OR reduced:ti,ab OR decrease:ti,ab OR decreased:ti,ab OR decreasing:ti,ab OR decreases:ti,ab OR stigma:ti,ab OR stigmas:ti,ab OR cost:ti,ab OR costs:ti,ab OR burden:ti,ab OR training:ti,ab OR development:ti,ab OR develop:ti,ab OR developing:ti,ab OR developments:ti,ab OR developed:ti,ab OR develops:ti,ab	17015556
5	1 AND 2 AND 3 AND 4	1561
6	AND [humans]/lim	1488

Database (including vendor/platform): Web of Science

Set		Results
1	TS=(hearing OR deaf OR deafness)	167092

2	TS=(Tests OR " hearing aids" OR "hearing aid" OR implant OR implants OR "assistive technology" OR "assistive technologies" OR "assistive device" OR "assistive devices" OR Intervention OR interventions OR screening OR screenings)	7149639
3	TS=("health care" OR "healthcare" OR "health policy" OR "health services")	617332
4	TS=(Access OR accessibility OR uptake OR scale OR "scale up" OR "scaling up" OR "scale-up" OR adopt OR adopted OR adopts OR barrier OR barriers OR strategy OR strategies OR strategic OR innovative OR innovation OR implement OR implementing OR implemented OR implements OR adoption OR improvement OR improve OR improved OR improves OR improving OR increase OR increasing OR increases OR increased OR reduce OR reducing OR reduces OR reduced OR decrease OR decreased OR decreasing OR decreases OR stigma OR stigmas OR cost OR costs OR burden OR training OR development OR develop OR developing OR developments OR developed OR develops)	24609137
5	1 AND 2 AND 3 AND 4	1577

Database (including vendor/platform): Global Health & Global Health Archive

Set		Results
1	DE "hearing impairment" OR DE "deafness" OR TI (hearing OR deaf OR deafness) OR AB (hearing OR deafness OR deaf)	8231

2	DE "screening" OR DE "mandatory screening" OR DE "prenatal screening" OR DE "anonymous testing" OR TI(Tests OR " hearing aids" OR "hearing aid" OR implant OR implants OR "assistive technology" OR "assistive technologies" OR "assistive device" OR "assistive devices" OR Intervention OR interventions OR screening OR screenings) OR AB(Tests OR " hearing aids" OR "hearing aid" OR implant OR implants OR "assistive technology" OR "assistive technologies" OR "assistive device" OR "assistive devices" OR Intervention OR interventions OR screening OR screenings)	496760
3	DE "health services" OR DE "community health services" OR DE "maternity services" OR DE "Medicaid" OR DE "medical services" OR DE "public health services" OR DE "school health services" OR DE "traditional health services" OR DE "health care" OR DE "primary health care" OR TI(health OR "healthcare") OR AB(health OR "healthcare")	677091

4	TI(Access OR accessibility OR uptake OR scale OR "scale up" OR "scaling up" OR "scale-up" OR adopt OR adopted OR adopts OR barrier OR barriers OR strategy OR strategies OR strategic OR innovative OR innovation OR implement OR implementing OR implemented OR implements OR adoption OR improvement OR improve OR improved OR improves OR improving OR increase OR increasing OR increases OR increased OR reduce OR reducing OR reduces OR reduced OR decrease OR decreased OR decreasing OR decreases OR stigma OR stigmas OR cost OR costs OR burden OR training OR development OR develop OR developing OR developments OR developed OR develops) OR AB(Access OR accessibility OR uptake OR scale OR "scale up" OR "scaling up" OR "scale-up" OR adopt OR adopted OR adopts OR barrier OR barriers OR strategy OR strategies OR strategic OR innovative OR innovation OR implement OR implementing OR implemented OR implements OR adoption OR improvement OR improve OR improved OR improves OR improving OR increase OR increasing OR increases OR increased OR reduce OR reducing OR reduces OR reduced OR decrease OR decreased OR decreasing OR decreases OR stigma OR stigmas OR cost OR costs OR burden OR training OR development OR develop OR developing OR developments OR developed OR develops)	2202891
5	1 AND 2 AND 3 AND 4	738

Database (including vendor/platform): Global Index Medicus

Set		Results
1	tw:(hearing OR deaf OR deafness)	13877
2	tw:(test OR tests OR aid OR aids OR "assistive technology" OR "assistive technologies" OR "assistive device" OR "assistive devices" OR Intervention OR interventions OR screening OR screenings)	505570

3	Tw:(Health OR healthcare)	347353
4	TW:(Access OR accessibility OR uptake OR scale OR "scale up" OR "scaling up" OR "scale-up" OR adopt OR adopted OR adopts OR barrier OR barriers OR strategy OR strategies OR strategic OR innovative OR innovation OR implement OR implementing OR implemented OR implements OR adoption OR improvement OR improve OR improved OR improves OR improving OR increase OR increasing OR increases OR increased OR reduce OR reducing OR reduces OR reduced OR decrease OR decreased OR decreasing OR decreases OR stigma OR stigmas OR cost OR costs OR burden OR training OR development OR develop OR developing OR developments OR developed OR develops)	887563
5	1 AND 2 AND 3 AND 4	797

X. APPENDIX III – GLOSSARY OF TERMS

1. **NHS** – neonatal hearing screening
2. **CHS** – child hearing screening
3. **AHS** – adult hearing screening
4. **HIC** – high-income country, as defined by the World Bank
5. **UMIC** – upper middle-income country, as defined by the World Bank
6. **LMIC** – lower middle-income country, as defined by the World Bank
7. **LIC** – low-income contrry, as defined by the World Bank
8. **YLD** – years lived with disability
9. **PRISMA** – Preferred Reporting Items for Systematic Reviews and Meta-Analyses
10. **MMAT** – Mixed Methods Appraisal Tool (2018 edition)
11. **Scale-up strategy** – an intervention that demonstrated a change in uptake by reporting the state of the target population both before and after implementation; an intervention designed to change metrics related to use, adherence, and/or satisfaction upon implementation
    a. **Successful** – uptake increased; "positive" effect observed
    b. **Unsuccessful –** uptake decreased; "negative" effect observed
    c. **Mixed Results** – a combination of "positive" and "negative" effects observed
    d. **Null** – no change in uptake
12. **Outcome** – what the authors of each article seek to measure, along with what they hypothesize will happen to this measure upon scale-up strategy implementation, relative to previous conditions

13. **Effect** – how the measure actually changed due to the implementation of the scale-up strategy, often described quantitatively in the results of the study
14. **Neonates/Newborns** – patients age 0-28 days who may be surveyed by proxy using family members to vouch for the patient
15. **Children** – patients age 28 days to 18 years and may include young members who are surveyed by proxy using family members
16. **Adults** – patients age 18 years and older, who may be groups by veteran status or occupation
17. **Use** – access and/or consumption of screening services, including measures of coverage, rates of utilization, and time/age of first access
18. **Adherence** – commitment to the screening and diagnostic process and can encompass measures related to referral, follow-up, continuation of screening protocol, and follow-through on recommendations and post-initial screening care
19. **Satisfaction** – fulfillment of needs and expectations for screening services, which is often measured via survey
20. **Education** – theme that describes approaches to alter patient knowledge and understanding of hearing screening
21. **Policy and Systems** – theme that describes interventions that promote screening uptake and accessibility by redressing healthcare and/or political infrastructure
22. **Telehealth** – theme that involves the use of web- and digital-based programs for patient screening as a means to change capacity and uptake
23. **Financial/Funding** – theme that describes interventions which seek to alter the financial barriers that may prevent patients from seeking care

24. **Expanded Screening** – theme that refers to approaches that modify the reach and capacity of existing services, commonly through universalization
25. **Restructured Screening** – theme that describes interventions which utilize existing screening programs and revise their infrastructure and protocol to alter capacity and uptake
26. **Uses Screening** – theme that refers to the utilization of screening to change the capacity and uptake of another hearing healthcare intervention, such as hearing devices

XI. APPENDIX IV – MMAT (2018) RESULTS (TABLE 2)

REFERENCE	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5	TOTAL (S1+S2+1/2/3/4/5)	AVG (SUM/7)
(07)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	0	1	1	0	0	0	0	0	0	0	0	0	0	6	0.85714286
(08)	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0	0	0	2	0.28571429
(09)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	7	1
(10)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(11)	1	1	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	7	1
(12)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(13)	1	1	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	7	1
(14)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	0	1	0	0	0	0	0	0	0	0	0	0	6	0.85714286
(15)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	0	1	0	0	0	0	0	0	0	0	0	0	6	0.85714286
(16)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(17)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(18)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(19)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(20)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(21)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(22)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	0	0	1	0	0	0	0	0	0	0	0	0	0	5	0.71428571
(23)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(24)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(25)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	0	1	0	0	0	0	0	6	0.85714286
(26)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(27)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(28)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(29)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(30)	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	7	1
(31)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(32)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(33)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(34)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1
(35)	1	1	0	0	0	0	0	0	0	0	0	0	1	1	1	1	1	0	0	0	0	0	0	0	0	0	0	7	1

Table 2 : Mixed Methods Appraisal Tool (MMAT) (2018) Results for Bias and Certainty

KEY:

Screening Question 1 (S1): Are there clear research questions?
Screening Question 2 (S2): Do the collected data allow to address the research question?
1: QUALITATIVE
1.1: Is the qualitative approach appropriate to answer the research question?
1.2: Are the qualitative data collection methods adequate to address the research question?
1.3: Are the findings adequately derived from the data?
1.4: Is the interpretation of results sufficiently substantiated by data?
1.5: Is there coherence between qualitative data sources, collection, analysis, and interpretation
2: QUANTITATIVE RANDOMIZED CONTROLLED TRIALS
2.1: Is randomization appropriately performed?
2.2: Are there groups comparable at baseline?
2.3: Are there complete outcome data?
2.4: Are outcome assessors blinded to the intervention provided?
2.5: Did the participants adhere to the assigned intervention?
3: QUANTITATIVE NON-RANDOMIZED
3.1: Are the participants representative of the target population?
3.2: Are measurements appropriate regarding both the outcome and intervention (or exposure)?
3.3: Are there complete outcome data?
3.4: Are the confounders accounted for in the design and analysis?
3.5: During the study period, is the intervention administered (or exposure occurred) as intended?
4: QUANTITATIVE DESCRIPTIVE
4.1: Is the sampling strategy relevant to address the research question?
4.2: Is the sample representative of the target population?
4.3: Are the measurements appropriate?
4.4: Is the risk of non-response bias low?
4,5: Is the statistical analysis appropriate to answer the research question?
5: MIXED METHODS
5.1: Is there an adequate rationale for using a mixed method design to answer the research question?
5.2: Are there different components of the study effectively integrated to answer the research question?
5.3: Are the outputs of the integration of qualitative and quantitative components adequately interpreted?
5.4: Are divergence and inconsistencies between quantitative and qualitative results adequately addressed?
5.5: Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?

XII. APPENDIX V – USE, ADHERENCE, SATISFACTION, AND COMPOSITE DATA TABLES (TABLES 3-6)

a. USE

Reference	Country	World Bank Classification	Scale-Up Strategy: 1) Intervention 2) Target Population	Intervention Type	Does the Intervention Outcome Target "Use"?	Other Intended Outcomes	Overall Effect of the Scale-Up Intervention on Outcome	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 1)	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 2)	Effect of the Scale-Up Intervention on Population Sub-Groups (if applicable)
(7)	Israel	HIC	1) Health promotion lectures for patients and providers 2) 206 adult patients ages 65 and older and two control groups (the first being 101 people registered with the same general practitioner, and the second 87 people registered with another general practitioner).	Education	Use		Positive only (scale up achieved)	Amongst patients, there was a significant increase in numbers undergoing a hearing test, from 19 per cent before the intervention to 49 per cent two years later, while in the two control groups there was little change		
(8)	Australia	HIC	1) community-based demonstration project to address hearing health screening conducted in the Australian state of New South Wales 2) farmers from the Far West, North Coast and New England regions of the state of New South Wales (NSW)	Education	Use		Mixed results	In comparing data from 2007 with that of 2008 when the project operated, providers in New England reported an increase of 26% in service utilization by farmers, although the sample was small (29–39 farmers). Providers from the North Coast reported only one instance where a client had indicated that the local promotion had been central to attending for a hearing assessment		
(12)	Italy	HIC	1) ad hoc UNHS legislation (Health Regional Laws) prescribing or encouraging UNHS 2) The number of Hospitals involved in the four reporting years was 618 in 2003, 607 in 2006, 711 in 2008 and 505 in 2011. They accounted for 532,221 births in 2003 (corresponding to 97.8% of total live births), 541,970 births in 2006 (corresponding to 96.7% of total live births), 535,577 births in 2008 (corresponding to 92.8% of total live births), and 527,308 births in 2011 (corresponding to 96.7% of total live births). (2,137,076 neonates between 2003-2011)	Policy and Health Systems	Use		Positive only (scale up achieved)	As shown in Fig. 2, screening coverage increased progressively from 29.3% (156,048 out of 532,221 births) in 2003 to 48.4% (262,103 out of 541,970 births) in 2006, to 60.6% (324,537 out of 535,577 births) in 2008 and to 78.3% (413,212 out of 527,308 births) in 2011. Similarly, screening coverage increased during the 2003-2011 reporting period also in all the five geographical areas: from 62.2% (85,291 out of 137,175 births) to 97.1% (137,013 out of 141,067 births) in the North West; from 36.6% (33,315 out of 90,957 births) to 95.9% (93,433 out of 97,427 births) in the North East; from 17.3% (16,927 out of 97,797 births) to 71.5% (73,940 out of 103,395 births) in the Center; from 12.0% (16,795 out of 140,327 births) to 74.4% (96,260 out of 129,366 births) in the South; and from 5.6% (3,720 out of 65,965 births) to 27.4% (12,566 out of 56,053 births) in the Islands. The larger increase was observed in the Southern Regions, where the coverage in 2011 was 62.4 percentage points greater than in 2003, followed by the North-East (59.3 percentage points of increase), the Regions of the Center (54.2 percentage points of increase), the North-West (34.9 percentage points of increase), and the Islands (21.8 percentage points of increase).		

(13)	US	HIC	1) policy and benchmark changes to neonatal hearing screening diagnostic criteria at hospital level 2) 111 infants in retrospective study	Policy and Health Systems	Use	Adherence	Positive only (scale up achieved)	Loss to follow up decreased from 18% (25 of 139 infants) to 7% (8 of 111 infants)	age of screening decreases from an average of 7.9 weeks to average of 5.1 weeks (4.78 if eliminating NICU outliers)	
(14)	Vietnam	LMIC	1)health-system level changes to EDHI program in Hai Phong, Vietnam 2) A total of 190 children (<15 years old) with hearing impairments and whose records were maintained by the participating service providers for 2 years (2013–2014) were interviewed. Children >15 years old who are afflicted with hereditary hearing impairment and whose families declined participation in the research were excluded.	Policy and Health Systems	Use	Adherence	Positive only (scale up achieved)	The ages at which interventions were initiated significantly decreased compared with the ages of implementation before the proposed solutions were applied ($p<0.05$), that is, children with hearing loss were provided treatments, except for language intervention, at 26.5 months. At <24 months, the disease was discovered and initially diagnosed, a definitive diagnosis was made, and hearing aids were fitted. Note, however, that the ages at which these events take place continue to fluctuate substantially from 1 to 49 months (Fig. 2).	The number of children with hearing loss who receive treatment increased (61.2% vs. 36.4%), and the number of children whose families are compelled to apply self-intervention decreased (38.8% vs. 63.6%; $p<0.001$).	
(15)	Taiwan	HIC	1) Government-funded programming for UNHS--government covered cost of automatic auditory brainstem response screening 2) 3,361 neonates from Taipei hospital between August 2009 to July 2010	Financial/Funding	Use	Adherence	Positive only (scale up achieved)	The coverage rate of the study was significantly higher than that of the control group (99.64% vs. 87.34%, P<.001).	A significant decrease of the referral rate was achieved in the study group when compared with the control group (0.95% vs. 2.82%, P < .001). The follow-up rate of the study group was significantly higher than that of the control group (100.00% vs. 40.74%, P < .001).	
(16)	South Korea	HIC	1) hospital-based NHS program in South Korea, where screening costs for low-income families are paid by the National Health Authority. 2)NHS process for 13805 newborns delivered in a tertiary referral center of South Korea from 2005 through 2014 was reviewed.	Financial/Funding	Use		Positive only (scale up achieved)	In this hospital-based NHS program, the screening rate plateaued at ~50% when the National Health Authority was not involved but increased to ~70% when the cost for low-income families was covered by the government.		
(18)	New Zealand	HIC	1) Universal preschool health screening in NZ 2) 252,273 preschool aged children who qualified if (1) they were ever resident in New Zealand (NZ), (2) lived in NZ for at least 6 months during the reference year, (3) were alive at the end of the reference year, (4) either appeared in any hospital (including emergency) admissions, community pharmaceutical dispensing or general practitioner enrolment datasets during the reference year or (5) had a registered birth in NZ	Expanded Screening	Use		Positive only (scale up achieved)	Screening coverage increased from 86% to 91% from 2011 to 2015		Māori and Pacific children were less likely to complete the checks than non-Māori and non-Pacific children (for VHTs: Māori: OR=0.60[95% CI 0.61 to 0.58], Pacific: OR=0.58[95% CI 0.60 to 0.56], for nurse checks: Māori: OR=0.63[95% CI 0.64 to 0.61], Pacific: OR=0.67[95% CI 0.69 to0.65] and for SDQ-T Māori: OR=0.76[95% CI 0.78 to 0.75], Pacific: OR=0.37[95% CI 0.38

										to 0.36]). Children from socioeconomically deprived areas, with younger mothers, from rented homes, residing in larger households, with worse health status and with higher rates of residential mobility were less likely to participate in the B4 School Check than other children.
(19)	Slovakia	HIC	1)mandatory UNHS for early detection of hearing loss 2) 162,758 Slovakian newborns	Expanded Screening	Use		Positive only (scale up achieved)	In year 2005 - 42% of newborns in Slovakia were screened, in 2006 - 66% newborns and in 2007 - 94, 99% (three small newborn departments do not yet have equipment for OAE screening). Comparing the number of identified cases with bilateral severe permanent HL or deafness before and after UNHS, 22.8% more cases of PHL were identified in the first year of UNHS.		
(20)	Denmark	HIC	1) nationwide NHS programming in Denmark following Danish Health and Medicines Authority 2) neonates born in Denmark between 2005-2014	Expanded Screening	Use	Adherence	Positive only (scale up achieved)	We recorded an annual increase in average screening density; from 88.6% in 2006 to 94.8% in 2013.	Furthermore, in 2006, 89.5% had completed the hearing screening programme within 30 days and in 2014 this figure had increased to 99%. The average time to diagnosis decreased from 3.5 months in 2006 to 0.7 months in 2013.	
(21)	Italy	HIC	1) universal NHS programming throughout Italy 2)20841 newborns in 11 Italian hospitals (20,051 were well born babies (WB), while 790 babies (3.8%) presented identified audiological risk factors (BRF).)	Expanded Screening	Use		Positive only (scale up achieved)	The coverage rate of newborn screening was only 34.4% in 2006. The overall coverage rate in the study period was 93.8% (2010-2012).	The average age of identification of congenital hearing loss was around 32 months of age in 2006. The mean age of identification in 2010-2012 was 5.31 +/- 3.95 months for well born newborns and 11.28+/- 7.73 months in at-risk group	
(23)	Netherlands	HIC	1) Home screening for UNHS via home intake and home metabolic screening programs 2)3336 newborns from Denmark	Expanded Screening	Use	Adherence	Mixed results	In setting B, where UNHS was performed during the same home visit as the screening for metabolic diseases on the fourth to seventh day after birth, the proportion referred was lowest, 1.4%. In setting A where neonatal hearing screening was performed at the WBC the proportion of referred children was 2.3%. In setting C where the screening was performed during the first home visit by a nurse from the WBC the proportion was 2.7%. These differences were not significant (Pearson's chi square test 3.32, df2, p.19). Refer rate per screening stage was lowest for setting B at each of the screening stages.	Total participation across three stages differs between arms. Well-baby clinics (control)=75.2%, home metabolic screening= 88.9%, home intake = 86.9%	

(24)	Poland	HIC	1) Polish Universal Newborn Hearing Screening Program supported by the Great Orchestra for Christmas Charity (WOŚP) foundation 2)4,672,704 newborns in Poland from 2003-2015	Expanded Screening	Use		Null / no effect	The proportion of newborns screened in Poland increased after 2002 due to the support afforded by WOŚP to the Universal Neonatal Hearing Screening Program. In the first year of the Program's operation, the number of newborns entered into the Program's database and screened for hearing impairment was 78,748. In the subsequent year, neonates born throughout Poland were included in the Program, with 336,460 newborns (i.e., 97% of all neonates born in Poland) entered into the Program's database. From then onward, 359,439 newborns on average were included in the Program each year, yielding a total of 4,672,704 newborns screened during the evaluated 12-year period		
(25)	Netherlands	HIC	1)enhanced hearing health education 2)687 older adults living at home	Education	Use		Positive only (scale up achieved)	Hearing aid use 1 year after screening was 6.3%, 4.1%, and 7.4% in the same arms, compared with 3.3% in the control arm (P<.01).		follow-up).
(26)	Ireland	HIC	1) 2-stage UNHS screening program in West of Ireland; 2) 26,281 neonates studied over the eight-year period.	Restructured Screening	Use		Positive only (scale up achieved)	No NHS done before (typical hearing screening occurred previously only for children at a minimum of 7-9 months of age); after the pilot UNHS at the two hospitals in which the program was implemented, a 98% coverage rate was achieved (effect = no NHS to high coverage of NHS)		
(27)	Italy	HIC	1) well-infant nursery hearing screening program for EDHI with transient evoked otoacoustic emission 2) 3379 newborns from 2003-2008	Restructured Screening	Use		Positive only (scale up achieved)	The coverage rate increased progressively from 89.8% to 92% from 2003 to 2009. The referral rate was 1.51% after second stage with a specificity value of 98.78%. The four-stage screening performed for G3 reduced the numbers of global audiology assessment to 0.91% with a final global specificity of 99.4 0.4%		only one community showed increased use of services (New England region)
(29)	Brazil	UMIC	1) two-stage protocol for outpatient NHS 2) 14205 infants from case files obtained from 2004 to 2013	Restructured Screening	Use	Adherence	Positive only (scale up achieved)	The adherence of the families was 32% in 2004 and increased to 85% in 2013	the mean age of the screened newborns was 48.66 days in 2005 and 24.53 days in 2013.	
(30)	France	HIC	1) Deferred preliminary screening (T3) before the age of 1 month 2) 115,435 neonates; All the facilities implemented the UNHS protocol, with 47 out of 51 using the recommended techniques. 99.7% of the 115,435 newborns were screened	Restructured Screening	Use		Positive only (scale up achieved)	In the perinatal network making extensive use of T3 (75.8% versus 14.9% elsewhere), 0.6% of the infants were referred to a diagnostic center, versus 2.9% in the rest of the region (2016, $p < 0.001$).		Twenty-two per cent of patients and 19 per cent of the first control group reported that their physicians suggested undergoing a hearing test; the second control group subjects (whose general practitioners received no specific educational intervention) showed no change.

(33)	Australia	HIC	1)NHS before both HA fitting and CI surgery 2)417 pediatric patients (221 male, 196 female) treated at age younger than 3	Uses Screening	Use		Positive only (scale up achieved)	ANOVA with Tukey pairwise comparison indicated significant differences for age at HA fitting (p50.001) and age at CI surgery (p50.001) across the NHS groups. Mean age at HA fitting was significantly older for Group 1 compared to Groups 2, 4, and 5. Groups 2 and 3 were not significantly different to each other, but Groups 1 and 3 were significantly different to Groups 4 and 5 (Figure 5), where the mean age at HA fitting and CI surgery for the five time periods; Group 1: Pre-NHS, Group 2: VIHSP, Group 3: VIHSP plus 4 NICUs, Group 4: Transition in implementation, and Group 5: Post-NHS	ANOVA with Tukey pairwise comparison indicated significant differences for age at HA fitting (p50.001) and age at CI surgery (p50.001) across the NHS groups. Mean age at HA fitting was significantly older for Group 1 compared to Groups 2, 4, and 5. Groups 2 and 3 were not significantly different to each other, but Groups 1 and 3 were significantly different to Groups 4 and 5 (Figure 5), where the mean age at HA fitting and CI surgery for the five time periods; Group 1: Pre-NHS, Group 2: VIHSP, Group 3: VIHSP plus 4 NICUs, Group 4: Transition in implementation, and Group 5: Post-NHS	
(34)	Canada	HIC	1) NHS to identify and early fit HAs 2) 709 children born between 1980 and 2003. who were diagnosed with hearing loss at the Audiology Clinic of the Children's Hospital of Eastern Ontario (CHEO)?	Uses Screening	Use	Adherence	Positive only (scale up achieved)	Children who had been screened in infancy were diagnosed significantly earlier (mean6.3 months; 95% CI, 5.6 to 7.0) than referred children with risk factors (mean34.5 months; 95% CI, 29.6 to 39.4) who in turn were diagnosed significantly earlier than referred children without risk factors (mean51.8 months, 95% CI, 48.51 to 55.14) (F (2,706)110.4, pB.001). It should be noted that of the 26 children identified through the IHP, the 21 NICU babies were diagnosed significantly later (Mann-Whitney U 5.50, PB.05) (mean7.0 months, 95% CI, 5.3 to 8.6) than the babies from the WBN (mean2.3 months, 95% CI, 0.8 to 3.7).		
(35)	US	HIC	1) AHS to improve HA use 2) 2305 older veterans	Uses Screening	AHS use improves HA use		Positive only (scale up achieved)	Hearing aid use 1 year after screening was 6.3%, 4.1%, and 7.4% in the same arms, compared with 3.3% in the control arm (P<.01).		1) AHS to improve HA use 2) 2305 older veterans

Table 3: Extraction data on scale-up pertaining to "Use"

b. ADHERENCE

Reference	Country	World Bank Classification	Scale-Up Strategy: 1) Intervention 2) Target Population	Intervention Type	Intended Intervention Outcome Targets Adherence?	Other Intended Intervention Outcome?	Overall Effect of the Scale-Up Intervention on Outcome	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 1)	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 2)	Effect of the Scale-Up Intervention on Population Sub-Groups (if applicable)
(9)	Malawi	LIC	1) multi-component educational intervention including (i) an information booklet; (ii) personalized counselling by a community health worker and an expert mother; (iii) a text message reminder to address poor uptake of hearing healthcare 2) 30 eligible families of children (<18 years) with hearing loss	Education	increased uptake of hearing healthcare referrals (adherence)		Positive only (scale up achieved)	Increased uptake of hearing healthcare referrals in hearing camps: in contrast to the earlier formative research, where uptake of referral at outreach camps was only 3%, uptake following the intervention was 53%)		
(10)	India	LMIC	1)telemedicine diagnostic ABR follow up for CHS conducted by VHWs 2) VHWs conducted DPOAE screening in 91 villages and hamlets in two administrative units (blocks) of a district in South India. VHWs who fulfilled the post training evaluation criteria were recruited for the screening program. VHWs screened 1335 children in Group A (in person follow up) and 1480 children in Group B (telemedicine follow-up).	Education	Adherence		Positive only (scale up achieved)	integration of tele-ABR resulted in 11% improvement in follow-up compared to in-person ABR at a tertiary care hospital.		1)telemedicine diagnostic ABR follow up for CHS conducted by VHWs 2) VHWs conducted DPOAE screening in 91 villages and hamlets in two administrative units (blocks) of a district in South India. VHWs who fulfilled the post training evaluation criteria were recruited for the screening program. VHWs screened 1335 children in Group A (in person follow up) and 1480 children in Group B (telemedicine follow-up).
(11)	US	HIC	1)Patient navigator intervention which focuses on elimination of breakdowns in communication, parent decisional support, and coordination of care through the complex EHDI system; Navigators identified specific barriers to care and then assisted participants by taking actions tailored to the specific needs of the individual 2) 61 dyads were enrolled in the study. Dyads=guardian-infant pairs, in which infants had abnormal NHS and recruited within first week after birth	Education	Adherence		Positive only (scale up achieved)	The percentage of participants nonadherent to diagnostic follow-up during the first 6 months after birth was significantly lower in the patient navigator arm compared with the standard of care arm (7.4% vs. 38.2%) (P = .005).	The timing of initial follow-up was significantly lower in the navigator arm compared with the standard of care arm (67.9 days after birth vs. 105.9 days, P = .010).	
(13)	US	HIC	1) policy and benchmark changes to neonatal hearing screening diagnostic criteria at hospital level 2) 111 infants in retrospective study	Policy and Health Systems	Adherence	Use	Positive only (scale up achieved)	Loss to follow up decreased from 18% (25 of 139 infants) to 7% (8 of 111 infants)	age of screening decreases from an average of 7.9 weeks to average of 5.1 weeks (4.78 if eliminating NICU outliers)	

(14)	Vietnam	LMIC	1)health-system level changes to EDHI program in Hai Phong, Vietnam 2) A total of 190 children (<15 years old) with hearing impairments and whose records were maintained by the participating service providers for 2 years (2013–2014) were interviewed. Children >15 years old who are afflicted with hereditary hearing impairment and whose families declined participation in the research were excluded.	Policy and Health Systems	Adherence	Use	Positive only (scale up achieved)	The ages at which interventions were initiated significantly decreased compared with the ages of implementation before the proposed solutions were applied ($p<0.05$), that is, children with hearing loss were provided treatments, except for language intervention, at 26.5 months. At <24 months, the disease was discovered and initially diagnosed, a definitive diagnosis was made, and hearing aids were fitted. Note, however, that the ages at which these events take place continue to fluctuate substantially from 1 to 49 months (Fig. 2).	The number of children with hearing loss who receive treatment increased (61.2% vs. 36.4%), and the number of children whose families are compelled to apply self-intervention decreased (38.8% vs. 63.6%; $p<0.001$).	
(15)	Taiwan	HIC	1) Government-funded programming for UNHS--government covered cost of automatic auditory brainstem response screening 2) 3,361 neonates from Taipei hospital between August 2009 to July 2010	Financial/Funding	Use	Adherence	Positive only (scale up achieved)	The coverage rate of the study was significantly higher than that of the control group (99.64% vs. 87.34%, P<.001).	A significant decrease of the referral rate was achieved in the study group when compared with the control group (0.95% vs. 2.82%, P < .001). The follow-up rate of the study group was significantly higher than that of the control group (100.00% vs. 40.74%, P < .001).	
(20)	Denmark	HIC	1) nationwide NHS programming in Denmark following Danish Health and Medicines Authority 2) neonates born in Denmark between 2005-2014	Expanded Screening	Adherence	Use	Positive only (scale up achieved)	We recorded an annual increase in average screening density; from 88.6% in 2006 to 94.8% in 2013.	Furthermore, in 2006, 89.5% had completed the hearing screening programme within 30 days and in 2014 this figure had increased to 99%. The average time to diagnosis decreased from 3.5 months in 2006 to 0.7 months in 2013.	
(23)	Netherlands	HIC	1) Home screening for UNHS via home intake and home metabolic screening programs 2)3336 newborns from Denmark	Expanded Screening	Adherence	Use	Mixed results	In setting B, where UNHS was performed during the same home visit as the screening for metabolic diseases on the fourth to seventh day after birth, the proportion referred was lowest, 1.4%. In setting A where neonatal hearing screening was performed at the WBC the proportion of referred children was 2.3%. In setting C where the screening was performed during the first home visit by a nurse from the WBC the proportion was 2.7%. These differences were not significant (Pearson's chi square test 3.32, df2, p.19). Refer rate per screening stage was lowest for setting B at each of the screening stages.	Total participation across three stages differs between arms. Well-baby clinics (control)=75.2%, home metabolic screening= 88.9%, home intake = 86.9%	1) Home screening for UNHS via home intake and home metabolic screening programs 2)3336 newborns from Denmark
(28)	US	HIC	1) targeted outpatient prescreening through WIC program 2) 260 infants associated with WIC program	Restructured Screening	Adherence	Use	Positive only (scale up achieved)	Among WIC-eligible intervention infants, the lost to follow-up rate over 2 years was 9.6%, compared with 28.7% for nonintervention infants in the same hospitals and 18.1% for nonintervention hospitals. The average age of hearing confirmation for the WIC intervention group was 34.8 days, compared with 63.6 days in non-WIC infants.	Average age at hearing confirmation across the 2 years combined for the WIC rescreening group was 34.8 days (median, 27.5 days; range, 2–283 days), which was significantly lower than nonWIC infants in the same hospitals (average, 63.6 days; median, 45 days;	

									range, 1–411 days; P < .0001) and was also significantly lower than in control hospitals (average, 49.4 days; median, 29 days, range, 1–495 days; P = .0007). Timely follow-up (rescreening by 1 month and diagnosis by 3 months of age) was examined by comparing the proportion of infants who had hearing confirmation by these target ages. By 1 month of age, 61% of study infants were confirmed by screening or diagnostic tests compared with only 24% of nonstudy infants in the same hospitals (P < .0001). By 3 months of age, 96% of study infants had received hearing confirmation, compared with 82% of nonstudy infants in the same hospitals (P < .0001)	
(29)	Brazil	UMIC	1) two-stage protocol for outpatient NHS 2) 14205 infants from case files obtained from 2004 to 2013	Restructured Screening	Adherence	Use	Positive only (scale up achieved)	The adherence of the families was 32% in 2004 and increased to 85% in 2013	the mean age of the screened newborns was 48.66 days in 2005 and 24.53 days in 2013.	
(31)	US	HIC	1) Using otoacoustic emission (OAE) testing in a single-visit two-tiered program for preschool hearing screenings 2)3257 low-income preschool-aged children	Restructured Screening	Adherence		Positive only (scale up achieved)	Referral rate decreased from 8% to 5% (P = 0.0014), and follow-up improved from 36% to 91% (P<0.0001).		
(32)	US	HIC	1)the addition of accommodations, like dedicated project secretaries, free daycare for siblings, cab vouchers for transport, and elimination of requirement for health maintenance organization referrals, to UNHS programming 2)2031 newborns. Seventy percent of the obstetrical population is indigent. The racial mix is 70% black, 20% Hispanic, and 10% white and other. The mothers' mean age at delivery is 25 years. Fourteen percent of infants are born before 37 weeks of gestation or are of low birth weight (	Restructured Screening	Adherence		Positive only (scale up achieved)	Collecting a complete database profile for each newborn, establishing rapport with the family, and offering immediate follow-up appointments yielded a 61% return rate after discharge. The addition of a dedicated project secretary, free day-care for siblings, and cab vouchers for transportation and the elimination of a requirement for health maintenance organization referrals increased follow-up yield to 75%.		

(34)	Canada	HIC	1) NHS to identify and early fit HAs 2) 709 children born between 1980 and 2003. who were diagnosed with hearing loss at the Audiology Clinic of the Children's Hospital of Eastern Ontario (CHEO)?	Uses Screening	Adherence	Use	Positive only (scale up achieved)	Children who had been screened in infancy were diagnosed significantly earlier (mean6.3 months; 95% CI, 5.6 to 7.0) than referred children with risk factors (mean34.5 months; 95% CI, 29.6 to 39.4) who in turn were diagnosed significantly earlier than referred children without risk factors (mean51.8 months, 95% CI, 48.51 to 55.14) (F (2,706)110.4, pB.001). It should be noted that of the 26 children identified through the IHP, the 21 NICU babies were diagnosed significantly later (Mann-Whitney U 5.50, PB.05) (mean7.0 months, 95% CI, 5.3 to 8.6) than the babies from the WBN (mean2.3 months, 95% CI, 0.8 to 3.7).		

Table 4: Extraction data on scale-up pertaining to "Adherence"

c. SATISFACTION

Reference	Country	World Bank Classification	Scale-Up Strategy: 1) Intervention 2) Target Population	Intervention Type	Intended Intervention Outcome Targets Satisfaction?	Other Targeted Intended Intervention Outcome	Overall Effect of the Scale-Up Intervention on Outcome	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 1)	Actual Effect of the Scale-Up Intervention on the Outcome (Effect 2)	Effect of the Scale-Up Intervention on Population Sub-Groups (if applicable)
(17)	US	HIC	1) audiometric screening with tablet-based applications in typical clinic locations. 2) Participants included 107 adult patients referred for audiometric testing to assess hearing loss.	Telehealth	Satisfaction		Null / no effect	Patients showed no preference for either conventional audiometry or the tablet-based device.		
(26)	US	HIC	1) hospital based UNHS program (instead of targeted program) 2) 95 post-partum women selected randomly from an obstetric ward at a university hospital and for 40 primary care pediatricians and 40 hearing specialists (audiologists and otolaryngologists) randomly selected from a local community.	Expanded Screening	Expanded Screening		Positive only (scale up achieved)	The majority of respondents in all three groups (97.5% hearing specialists, 82.5% pediatricians, and 89.5% mothers) favored universal over targeted screening. Differences in mean preference scores were statistically significant between hearing specialists and pediatricians ($p < 0.005$) and between hearing specialists and mothers ($p < 0.005$).		

Table 5: Extracted data on scale-up pertaining to "Satisfaction"