HOSPITAL-BASED NEWBORN HEARING SCREENING ACROSS THE GLOBE: A SYSTEMATIC REVIEW & PRELIMINARY REPORT ON GUATEMALA'S PROGRAM

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ABSTRACT

Background: An average of 3 out of every 1000 children are born with a hearing loss. Both hearing screening and intervention services have a significant positive impact on these children. The current study focused on screening services. There were two objectives. 1. Systematic Review: To identify and examine the reported factors and outcomes of hospital-based newborn hearing screening (NHS) programs. Factors and outcomes include screening protocol, type of equipment, coverage rates, referral rates, loss to follow up (LTF) rates and Birth Screening Performance Index (BSPI) 2. Guatemala NHS Program: To examine outcomes from Guatemala's program relative to the systematic review results.

Methods: Eligible studies published in English up to February 2019 were identified through searches of PubMed (NCBI), the Cochrane Library, ASHA Wire and relevant article reference lists. The data were screened using pre-defined inclusion and exclusion criteria to generate a list of eligible articles. Data extracted included age at screening, screening rate, referral rate, LTF rate, and number of newborns identified with permanent congenital hearing loss.

Systematic Review Results: This review identifies factors that affect BSPI, coverage rates, referral rates and loss to follow up rates in hospital-based universal NHS programs. 45 articles were included for data extraction and analysis.

Preliminary Data from Guatemala: Data were collected from January 2018 to March 2019 from four sites across the country. 2,024 newborns in total were screened. The prevalence of permanent congenital hearing loss (PCHL) was 4.9:1000. Of the 10 children with PCHL, one was diagnosed with bilateral microtia and all children received hearing technology and services.

Conclusion: The systematic review identified coverage rates, referral rates, LTF rates and the BSPI in hospital-based universal NHS programs. **S**creening protocol and type of equipment may play a role in referral rates and the BSPI. Multiple factors affect the overall LTF rate including use of an adequate data management system and the level of parental education. The NHS program in Guatemala is feasible, and the preliminary results are in line with other programs across the globe. Measures to expand the program to more geographic locations should continue.

Abbreviations: NHS – newborn hearing screening, JCIH – Joint Committee on Infant Hearing, PCHL – permanent congenital hearing loss, BSPI – Birth Screening Performance Index, HI – High Income, UMI – Upper Middle Income, LMI – Lower Middle Income, IQR – interquartile range, ABR – Auditory Brainstem Response, AABR – Automated Auditory Brainstem Response, OAE - Otoacoustic Emission, LTF – Loss/Lost to Follow-up, RCT – Randomized Control Trial

Keywords: universal newborn hearing screening, screening protocol, JCIH, hearing screening equipment

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1. Introduction

Hearing loss in childhood is a prevalent condition. Over 5% of the population worldwide (466 million people) have disabling hearing loss, and 34 million of these are children (WHO, 2018). Disabling hearing loss for children refers to a hearing loss of greater than 30 dBHL in the better hearing ear. Children identified when they are older than 6 months of age are at a high risk for speech and language delays because they do not have auditory access to the entire speech spectrum. There is also an unequal distribution of hearing loss across the globe [Appendix B]. The greatest prevalence is in areas with less access to audiological services such as the regions of South Asia and Sub-Saharan Africa.

Multiple studies have shown that hearing screening, early identification and early intervention for children with hearing loss has a significant, positive impact on their speech and language development (Moeller, 2000; Yoshinaga-Itano et al., 2017). In the early 2000s, the World Health Organization recommended "a policy for universal neonatal [hearing] screening be adopted in all countries and communities with available rehabilitation services…"(WHO, 2000). Since then, many countries including Guatemala have created or are in the process of implementing their own newborn hearing screening (NHS) programs. Guatemala is a country where 12.3% of the population, about 1,994,000 children, are between 0-5 years of age (United Nations Population Div., 2017). Since there had not been an NHS process for infants in Guatemala, the country's one practicing audiologist to date is collaborating with non-profit organizations to establish a program, starting in three regions of the country. More information on Guatemala's program will be discussed in the second half of this paper.

The purpose of this capstone is to report the preliminary outcomes and impact of the budding program in Guatemala, as well as to identify and examine other hospital-based NHS programs. This includes factors such as the type of equipment used and screening protocol. This also includes outcomes such as screening rates, referral rates, and loss to follow up (LTF) rates.

There are two types of equipment used in NHS programs; otoacoustic emissions (OAEs) and automated auditory brainstem response (AABR). These physiologic measures are appropriate for screening because both are noninvasive and available in automated versions that can easily be utilized by trained hospital staff. Traditional ABR testing is performed by an audiologist, as interpretation of the results is required. Traditional ABR can also be used in a diagnostic capacity to measure a child's hearing levels. Automated ABRs (AABRs) and OAEs have preset pass/refer criteria and provide these results in real-time for screening personnel. Each type of equipment has its strengths and weaknesses because the way the technology operates is different. The pass/refer criteria of the devices also differ because the criteria are set by the manufacturer of the specific equipment.

Though screening protocols vary widely across NHS programs, they can be organized by the type of equipment utilized and the maximum number of screening stages prior to full audiological evaluation. The type of equipment can be broadly classified into four categories:

- AABR-only
- OAEs-only
- Two-tier (OAEs with AABR rescreen)
- Two-technology (AABR and OAEs together at the same screen stage)

Protocols can include one or multiple hearing screening stages. For example, one hospital may use a protocol where all newborns have OAEs for the first stage. If the newborn is referred, or "fails," the screen in one or both ears, they return for a second stage screen using OAEs again. If they are referred for a second time, the third stage screen is completed through AABR. This would be an example of a three-stage two-tier screening protocol. Another hospital may have a two-stage OAEs-only protocol; both the first and the second stage is an OAE screen and there is no third screening stage.

The average protocol includes at least two screening stages in order to limit the number of newborns requiring a full diagnostic evaluation.

High screening rates of 90% or above signify that at least 90% of children born at a site are screened using a physiologic measure before 1 month of age, therefore meeting Joint Committee on Infant Hearing (JCIH) recommendations. A low referral rate refers to minimized false positive screens. This has two significant advantages. Minimizing the false positive rate will help prevent unnecessary burden on parents when their child has normal hearing, and it reduces the number of newborns needlessly

referred for full diagnostic evaluations (Caluraud et al., 2015). Loss to follow-up occurs when infants have not completed the newborn hearing screening process; they have done either one or two screens but have not returned for the next stage screen or a full diagnostic evaluation. A low lost to follow up rate would mean that these newborns are not missed and can therefore receive early intervention if needed.

There are multiple implementation methods that different countries have used to screen the hearing of their newborns. These methods include universal NHS, targeted hearing screening to patients at risk for hearing loss, integrating hearing screening at community-based immunization clinics, and using low-cost noise-maker tools to screen for severe and profound hearing losses only (Krishnan & Donaldson, 2013). Universal NHS occurs when all babies born at a site are screened, regardless of risk for hearing loss or co-morbid conditions. This review will focus solely on universal NHS programs in hospital-based settings; this most closely aligns with pilot programs across the globe and the program in development in Guatemala.

Many factors including limited resources and lack of awareness about hearing loss are potential barriers to creating a successful newborn hearing screening program. Hearing healthcare providers can make decisions prior to the start or expansion of a program that ultimately contribute to outcomes. Malleable factors include the type of equipment used and the screening protocol implemented.

This paper will attempt to assess whether the screening protocol and type of equipment results in different coverage, referral and LTF rates for babies born in hospitals. The paper also examines whether Guatemala's program is consistent with peer reviewed programs and has the potential to be a feasible and efficacious NHS program. Other systematic reviews have assessed newborn hearing screening in the following ways:

- Retrospectively studied children with hearing loss to assess who had undergone NHS (Nelson, Bougatsos, Nygren, & 2001 US Preventive Services Task Force, 2008)
- Compared screening versus no screening in cohort studies (Wolff et al., 2010)
- Assessed early versus later treatment/intervention after screening (Wolff et al., 2010)
- Assessed diagnostic accuracy of NHS (Wolff et al., 2010)

One systematic review explored LTF rates in universal NHS programs, reasons for LTF and strategies to reduce LTF (Ravi et al., 2016). This paper expands on Ravi and colleague's systematic review because it is a novel attempt at exploring two additional outcomes, coverage and referral rates. This review also checks for significant differences in these outcomes in relation to the screening protocol and equipment type.

2. Methods

A systematic review was conducted on studies of universal NHS programs and their structures. The purpose is to compare and analyze the implementation of such programs from around the world. A systematic search of evidence from the literature was performed per the Cochrane Collaboration guidelines and the PRISMA statement (Higgins, Sutton, & Cochrane Collaboration, 2008; Moher et al., 2009) [Appendix F]. Methods of data extraction, analysis, and inclusion and exclusion criteria were pre-specified and documented within the systematic review protocol as described below.

2.1. Inclusion Criteria

Types of studies: Research articles written or translated in English in peer-reviewed journals were included. Studies were included when the full text was available. Only studies reporting primary data were included. Studies that provided an overview or a review of more than one program were excluded along with commentary or opinion pieces.

Participants: Studies were included if the participants were human newborns between the ages of 0 and 23 months of age at the time of the first screen.

Interventions: Studies were included if they used a universal NHS approach where all newborns, regardless of risk for hearing loss, were screened. Screenings also must be conducted in a hospital or medical facility. The location was key to best control for the environment of the screening, as many studies do not document the specific ambient noise level of the screening environment. The hospital specification was also used to better standardize and compare the methodology across studies.

Outcomes: Studies were included if the outcomes contained coverage rates, referral rates and loss to follow-up rates. Each study must also document the prevalence of permanent congenital hearing loss (PCHL) and address the process after a positive diagnosis of hearing loss.

2.2. Study Identification

The literature search was conducted on three publication databases: PubMed, The Cochrane Library, and ASHA Wire. These databases were searched during the period of February 2018 to February 2019. Additional articles were obtained from reference lists of relevant studies. Searches included the following keywords:

- "assessment of newborn hearing screening program"
- "neonatal hearing screening pilot program AND outcomes"
- "newborn hearing screening program implementation"
- "neonatal hearing screening AND screening protocol"
- "hospital-based newborn hearing screening"

2.3. Screening and Eligibility

The search resulted in 733 articles over the three databases and 23 articles from relevant reference lists. 621 articles remained after duplicates were removed. A total of 388 articles from the initial search remained after the following criteria were applied; the full text was available, the articles were written or translated into English, and the study population consisted of human children aged birth to 23 months.

The remaining 388 titles generated by the search were scrutinized. Relevant abstracts were retrieved and assessed for suitability based on the inclusion and exclusion criteria. Eighty-two studies met the inclusion criteria and progressed to the next stage of screening that involved obtaining and reviewing the full-text of the articles.

The full-text review of the 82 articles found 37 articles failed to meet the inclusion criteria. The data from the remaining 45 articles were extracted and synthesized. The flow chart as per PRISMA requirements can be found on Appendix F.

2.4. Data Extraction and Synthesis

All eligible articles were read, and a table of evidence was created to assess key components of each study [Appendix A]. The table included details of research design, study location characteristics (WHO Region, World Bank Classification), participant count, and outcome measures (coverage rate, referral rate, loss to follow up and prevalence of PCHL). A Birth Screening Performance Index (BSPI) was calculated from the data as well (Finitzo et al., 1998). The index is the percent of newborns screened multiplied by the percent who pass the screening. This single number allows multiple studies and research sites with different screening protocols to be compared.

2.5. Individual Quality Assessment

A methodological quality appraisal tool [Table 1] was developed based on the National Institutes of Health Quality Assessment Tool (NIH, 2014), as used in another NHS systematic review (Ravi et al., 2016). Each study was rated for the questions on the tool and total percent scores were calculated. Studies were given a score of one if the study answered the individual question. Studies that did not answer a particular question were given a score of zero. The percent score for each study is the total score divided by the total number of questions in the tool multiplied by 100. Based on the percent score obtained, each study was rated as follows: 0-33.9% as weak, 34-66.9% as moderate and 67-100% as strong. Results are shown in Table 2.

Q1	Was the research question/objective clearly stated?
Q2	Was the study population clearly specified and defined?
Q3	Were the inclusion and exclusion criteria for being in the study pre-specified and applied uniformly to all study participants?
Q4	Was the sample size justification provided?
Q5	Was loss to follow-up after baseline testing (screening stage 1) 20% or less?
Q6	Was a pass/refer criterion clearly specified?

Table 1. Methodological Quality Appraisal Tool

Author (Year)	Q1	Q2	Q3	Q4	Q5	Q6	% Score
Abdullah et al. (2006)	1	1	0	0	0	0	33.33
Ahmad et al. (2011)	1	1	0	0	0	0	33.33
Arslan et al. (2013)	1	1	0	0	1	1	66.67

Augustine et al. (2014)	1	1	0	0	1	1	66.67
Benito-Orejas et al. (2008)	1	1	0	0	1	1	66.67
Berninger & Westling, (2011)	1	1	0	0	1	1	66.67
Bevilacqua et al. (2010)	1	1	1	0	1	1	83.33
Caluraud et al. (2015)	1	1	1	0	1	1	83.33
G. Chen et al. (2012)	1	1	1	0	0	1	66.67
X. Chen et al. (2017)	1	1	1	0	0	1	66.67
Cianfrone et al. (2018)	1	1	1	0	0	1	66.67
Connolly et al. (2005)	1	1	1	0	1	1	83.33
De Capua et al. (2007)	1	1	1	0	1	1	83.33
Finitzo et al. (1998)	1	1	1	0	0	1	66.67
Gilbey et al. (2013)	1	1	1	0	1	1	83.33
Govaerts et al. (2001)	1	1	1	0	1	1	83.33
Habib & Abdelgaffar (2005)	1	1	1	0	1	1	83.33
Huang et al. (2013)	1	1	1	0	1	1	83.33
Jewel et al. (2013)	1	1	0	0	0	1	50.00
John et al. (2009)	1	1	1	0	1	1	83.33
Khairi et al (2005)	1	1	1	1	0	1	83.33
Kim et al. (2017)	1	1	0	0	1	1	66.67
Korres et al. (2006)	1	1	0	0	0	0	33.33
Lévêque et al. (2007)	1	1	0	0	1	1	66.67
Levit et al. (2015)	1	1	1	0	1	1	83.33
Lin et al. (2002)	1	1	0	0	0	1	50.00
Lin et al. (2005)	1	1	0	0	1	1	66.67
Lin et al. (2007)	1	1	0	0	1	1	66.67
Martines et al. (2007)	1	1	1	0	1	1	83.33
Mathur & Dhawan (2007)	1	1	0	0	1	1	66.67
Molini et al (2016)	1	1	1	0	0	1	66.67
Ng et al. (2004)	1	1	0	0	1	1	66.67
O'Connor et al. (2013)	1	1	0	0	1	1	66.67
Ohl et al. (2009)	1	1	0	0	1	0	50.00
Olusanya et al. (2008)	1	1	0	0	1	1	66.67
Pastorino et al. (2005)	1	1	1	0	1	1	83.33
Pisacane et al. (2013)	1	1	0	0	0	0	33.33
Qi et al. (2013)	1	1	0	0	0	1	50.00
Rojas et al. (2014)	1	1	1	0	0	1	66.67
Rouev et al. (2004)	1	1	0	0	1	1	66.67
Saki et al. (2017)	1	1	1	0	1	1	83.33
Vignesh et al. (2015)	1	1	0	0	1	1	66.67
Vohr et al. (2001)	1	1	1	1	1	1	100.00
Wu et al. (2011)	1	1	1	0	1	1	83.33
Yee-Arellano et al. (2006)	1	1	1	0	1	1	83.33

Table 2. Methodological quality appraisal scores obtained for each study. % score is the total score divided by total number of items in the tool multiplied by 100.

2.6. Overall Quality Assessment

Although randomized control trials (RCT) were not found, the researchers took deliberate measures to create quasi-experimental or cross-sectional one-group and two-group designs. Quasi-experimental designs involved the manipulation of one variable, the implementation of the screening program, without random assignment of participants. Also, it would be unethical to conduct RCTs by assigning certain newborns to go through the hearing screening process and for other newborns to not receive the same services when there is prior evidence of benefit. Therefore, observational studies that assess the feasibility and accuracy of an NHS program were more prevalent. Cross-sectional studies were observational studies that analyze data from a population or representative subset at a specific point in time. Some of the studies looked at historical data retrospectively from an established NHS program while others prospectively planned to collect data from a certain point in a new program.

All studies detailed efforts to ensure reliability and validity of the outcome measures, and all studies clearly specified and defined their study population [Table 2, Question 2]. The majority of studies screened over 90% of the newborns available for their research [Appendix A]. Two studies provided a sample size calculation/power analysis while the remaining stated where the study population originated.

3. Systematic Review Results

The methodological quality rating was carried out for each study. The tool used is shown in Table 1 and the rating for each study appears in Table 2. Based on the percent score obtained, the study was rated as weak, moderate or strong. In total, 17 studies were rated as strong, 24 as moderate, and the remaining 4 as weak.

The studies were conducted in 23 countries from each of the 6 World Health Organization (WHO) classified regions of the world and from three of the four World

Bank income classification groups [Appendix A]. The six WHO regions include the following: African Region, Region of the Americas, South-East Asia Region, European Region, Eastern Mediterranean Region, and Western Pacific Region. The four World Bank income groups are based on the gross national income per capita of the most recent fiscal year. Each country's economy is classified as Low income (GNI per capita of $995 or less), Lower-Middle income ($996 to $3,895), Upper Middle income ($3896 to $12,055) or High income ($12,056 or more). This income group classification is also used for reports by the United Nations.

Based on the World Bank list of economies: 27 studies were conducted in high income countries, 12 were from upper-middle income countries, and 6 were from lower-middle income countries. No published studies from a low-income country which met criteria had been found.

Goal 1: To identify and examine the reported outcomes of hospital-based newborn hearing screening programs.

All studies reported coverage, referral and LTF rates, though the focus of the research varied across studies. One study compared NHS results to newborn genetic screening for hearing loss results (Wu et al., 2011). Other studies performed a cost-benefit analysis and noted the expenses of the screening equipment (Chen et al., 2017; Lin et al., 2005; Vohr et al., 2001).

Coverage rates ranged from 76.11% to 100% in 44 out of 45 studies. One study by Kim et al. (2017) had an overall coverage rate of 53.63%. They noted that throughout the nine-year duration of their study, the annual coverage rate steadily rose and plateaued to 79.6%.

The overall referral rates ranged from 6.0% to 0.23% in 42 of the 45 studies. Higher rates of 21%, 10.68% and 6.4% were reported in Mathur & Dhawan, (2007), Ohl et al., (2009), and Lin et al., (2002), respectively.

The age at first test varied from less than 24 hours to 7 days for well babies and from 3 days to as close to discharge as possible for NICU babies. The prevalence of PCHL ranged from 0.04% to 0.79% in 43 of 45 studies. One study (Ohl et al., 2009) found a prevalence of 3.15%, though they noted the study population was mostly infants from the NICU. Newborns that require a more than 48-hour stay in the NICU have a higher prevalence of hearing loss. The second and third highest prevalence rates reported (1.5% and 1.25%) were from a randomly selected newborn population (Mathur & Dhawan, 2007) and a small sample size population (Khairi et al., 2005).

The Birth Screening Performance Index ranged from 52.59% to 99.67% with an interquartile range of 89.06% to 95.99% (median 94.05%, mean 90.68%). [Appendix A]. One key factor that played a role in the range of scores is the initial coverage rate within each study. One outlier had a BSPI of 52.89% (Kim et al., 2017). In their study, 53.63% of the newborns were screened, but 98.62% of those screened newborns passed.

Due to the methodological heterogeneity of the available research, a meta-analysis could not be performed. As such, outcomes will be reported generally based on the screening protocol/type of equipment and number of screening stages. Tables 3 and 4 display the average and range of each outcome based on screening protocol and number of screening stages. Seven studies used multiple variable protocols and are not included in either table.

Screening Protocol	**AABR-only**	**OAEs-only**	**Two-Tier**	**Two-Tech**
Number of Studies	6	23	8	1
BSPI	89.28 *(52.9-99.7)*	91.79 *(79.5-99.0)*	94.06 *(74.1-99.4)*	98.75
Coverage Rate	90.98 *(53.6-100)*	96.15 *(80.2-100)*	95.68 *(76.1-100)*	100.00
Referral Rate	1.65 *(0.23-4.07)*	3.36 *(0.3-21.0)*	2.20 *(0.36-4.6)*	1.25
LTF Rate	7.64 *(0.03-18.0)*	18.25 *(0-58.2)*	10.82 *(0-29.9)*	4.23
Prevalence of PCHL	0.28 *(0.16-0.44)*	0.51 *(0.09-3.15)*	0.28 *(0.04-0.6)*	0.24
Number of screenings	2 *(1-3)*	2.5 *(2-4)*	2.34 *(2-3)*	2

Table 3. NHS Outcomes Categorized by Screening Protocol. The average is listed first, and the range is in parentheses.

Number of Screenings	**1 Screen**	**2 Screens**	**3 Screens**	**4 or more screens**
Number of Studies	1	24	12	2
BSPI	99.67	92.16 *(52.9-99.2)*	92.36 *(85.3-99.4)*	94.57 *(93.6-95.5)*
Coverage Rate	99.90	95.04 *(53.6-100)*	96.46 *(89.2-100)*	100.00 *(100-100)*
Referral Rate	0.23	2.33 *(0.29-10.7)*	2.07 *(0.47-6.0)*	13.70 *(6.4-21.0)*
LTF Rate	0.03	14.25	18.12	10.11

		(0-49.7)	*(0-58.2)*	*(0.21-20.0)*
Prevalence of PCHL	0.16	0.46 *(0.09-3.15)*	0.27 *(0.04-0.6)*	1.01 *(0.52-1.5)*

Table 4. NHS Outcomes Organized by Number of Screenings. The average is listed first, and the range is in parentheses.

4. Discussion

All studies noted that universal newborn hearing screening is feasible for their site, though different factors played a role in how each protocol was created and implemented, as well as outcomes.

The variability between studies and the small sample sizes for particular screening protocols (i.e. two-tech, 1 screen and 4+ screens), prevent the possibility of a valid statistical analysis. Instead, overall trends can be seen in Tables 3 and 4.

Another way to measure the success of any NHS program is to assess whether it follows the Joint Committee on Infant Hearing (JCIH) recommendations. The JCIH is a committee formed by audiologists, otolaryngologists, pediatricians and nurses. Their mission is to address issues related to early identification, intervention and follow-up care of infants and young children with hearing loss. Their most recent statement recommended that infants be screened by 1 month of age, diagnosed by 3 months, and fit with hearing technology and/or placed in early intervention services by 6 months of age (Joint Committee on Infant Hearing, 2007). The committee also recommends that an infant who failed the initial screening whether in one ear or both should have a complete second screening with both ears. The majority of studies reported used JCIH recommendations as their benchmarks for outcomes and to set up their protocol.

Referral Rates

Referral rates across the screening protocols were fairly close (except for the outliers discussed in the results section). High referral rates after the first hearing screening are often due to the presence of debris in the auditory canal during testing in the first 24 hours of life (Saki et al., 2017). To avoid unnecessary referrals for infants with normal hearing, 44 of the 45 studies reviewed used a protocol of multiple screenings for all infants prior to discharge from the hospital.

Both OAEs and AABR have been shown to be reliable and adequate types of equipment to screen for hearing loss in infants; this review looked to compare the two and find any significant pattern or difference. All studies that answered this part of the research question found significantly lower referral rates for the initial hearing screening using AABR compared to OAEs (Benito-Orejas et al., 2008; Vohr et al., 2001). They found AABR to be more sensitive, particularly in cases were the first screen is conducted shortly after birth.

Coverage Rates

Coverage rates did not appear to be affected by the type of equipment. Five of the NHS programs in the systematic review used a two-tier two-stage system with OAEs for the first screen and AABR for the second screen. Four studies used a two-stage AABR-only approach and 14 used a two-stage OAE-only approach. 21 of these 23 programs had coverage rates at or above 90%, which suggests that other factors besides type of equipment may be affecting coverage rates.

It is important to note that cost to the family is one prevalent factor in coverage rates. Multiple NHS programs addressed this issue. Kim et al. 2017 stated that the National Health Authority in South Korea provided funds for hearing screening and confirmatory tests for newborns from eligible families starting in 2009. The screening rate for their facility, Seoul National University Bundang Hospital, was initially 50%. After the government started covering the cost for low income families, the coverage rate increased significantly to 79.6%.

The training and availability of the tester also played a role in coverage rates. A pilot study in Lagos, Nigeria relied on trained non-specialist staff to address the number of newborns who required hearing screenings. They found that training staff significant contributed to their 98.7% coverage rate and 3.5% referral rate (Olusanya et al., 2008). The study by Lin and colleagues (2002) also found that 9.8% of their newborns were discharged without hearing screenings largely due to the lack of available screeners on the weekends. Training of non-specialist staff to run the screens addressed the shortage of hearing screeners in some locations and significantly improved coverage rates.

LTF Rates

In general, it appears that programs which use an OAEs-only approach or conduct three screens prior to full evaluation tend to have a higher LTF rate. On the other hand, multiple other factors affect the LTF rate. Ravi and colleagues found 27 contributing factors for loss to follow up in their systematic review, as noted in Table 5 below. Further research is needed in order to assess whether the screening protocol and number of stages significantly play a role compared to these other factors.

Table 5. Factors for LTF, in order of prevalence in Ravi and colleagues' systematic review.

1. Educational disparities and lack of knowledge among parents	14. Lack of scheduling a follow-up appointment
2. Distance	15. Lack of understanding of results
3. Work constraints	16. Parental refusal
4. Unfavorable attitudes	17. Lack of service provider's knowledge
5. Less priority given to hearing compared to other medical conditions by parents	18. Overburdened nursing staff used as a screener
6. Rescreened at some other center	19. Lack of family support
7. Inaccurate contact details	20. Lack of service system capacity
8. Change of Address	21. Communication failure, socio-economic barriers, lack of parental reminders
9. Superstitious and cultural beliefs	22. Incomplete test reports
10. Barriers to adequate healthcare	23. Short time to implement NHS
11. Financial constraints	24. Early discharge
12. Lack of health insurance	25. Screening staff on leave
13. Forgot to keep an appointment	26. Reduced maternal education
	27. Anxiety

5. Limitations

The majority of the research included in the present study came from high and upper-middle income countries. The review did not include non-published literature, dissertations, and studies from other sources not included in the databases searched or reference lists assessed. Due to the heterogeneity of studies, it is difficult to parse which specific factors had a significant impact on outcomes.

6. Systematic Review Summary

In summary, this systematic review suggests that screening protocol and type of equipment play a role in referral rates and LTF rates though not necessarily in coverage rates.

Screening coverage rates were affected by multiple confounding factors including time of discharge and availability of screeners. Referral rates appear to be affected by the screening protocol and number of screening stages as well as the age of the newborn at screening. In general, multiple factors affect LTF rates.

Future higher quality studies with well-defined study criteria and experimental/quasi-experimental designs are required to confirm these conclusions with higher confidence. Also, there is a need for better homogeneity regarding the definitions of terms across studies such as 'referral rate' and 'loss to follow up.' In studies where the terminology diverged from those stated in this systematic review, the raw data was used to calculate results based on the systematic review definition. Study findings can therefore be more generalized with standardized terminology and methods for reporting data. Also, more research needs to be done in the areas without established newborn hearing screening programs. The prevalence of hearing loss in most countries in Africa and Southeast Asia are estimates [Appendix B], though concrete data has not been collected. Evidence of the exact prevalence of hearing loss and the burden of undiagnosed hearing loss on local communities can help support the need for audiological services in these areas.

7. Preliminary Results of Guatemala's Program

Guatemala is an Upper Middle Income country in the WHO Region of the Americas (AMR). Prior to early January 2018, there was no universal NHS program in the country. An estimated total number of births throughout the country is 1,125 per day (United Nations Population Div., 2017). It's estimated that 3 out of every 1000 children are born with a hearing loss in the United States (CDC, 2003). If the prevalence is similar across countries, then there is a potential for an average of 3-4 children born in Guatemala every day to have a hearing loss that can be detected through an NHS program.

Two organizations, Rotary Club and Fundación Sonrisas que Eschuchan (Smiles that Listen) have invested in creating a pilot program in Guatemala. They are collaborating with the only practicing audiologist in the country to date, Patricia Castellanos de Muñoz, Au.D., to serve the communities. They started the program in January 2018 in public hospitals in the cities of San Cristóbal, Mazatenango, Quetzaltenango and the capital, Guatemala City [Appendixes C, D, E]. All four cities are populous areas in the country and all except San Cristobal are capital cities of their department, which is the local equivalent to a county or state. By starting in these regions, a large population of newborns throughout the country will be reached [Table 6].

City	Pop.	City Characteristics
Guatemala City	1022,000	Capital of the country and most populous city in Guatemala and all of Central America.
San Cristóbal	30,155	One of the biggest housing developments near Guatemala City.
Sololá	430,573	Capital of the Sololá department
Quetzaltenango	661,375	Second largest city in Guatemala and the capital of the Quetzaltenango department.
Mazatenango	43,225	Capital of the Suchitepéquez department

Table 6. Screening sites and characteristics

Rotary Club provided three GSI Corti OAE screeners, one portable ABR machine, salary for one technician, 200 hearing aids and funds for training. Smiles that Listen provided two technicians, one supervisor, transportation to the sites, reports of screening results, follow-up services and contacts with local hospitals. Recently a fourth site, Sololá, was added and data was collected on that site as well. The technicians brought the OAE equipment to their partner hospitals and screened newborns in the nursery and NICU daily. In order to confirm coverage across multiple hospitals in each city, hospital staff would confirm a time slot for the technicians to come screen.

8. Methods

Between January 2018 and March 2019, a total of 2,024 newborns were screened for hearing loss within the first month of life. The program employed two different hearing screening protocols based on whether a newborn was classified as "No Risk" or "High Risk." Both protocols used TEOAEs, though the ABR machine was shared amongst the four locations and used for screening instead of OAEs whenever possible.

For the No Risk group, a two-step screening was used prior to discharge [Fig. 1]. The first screening was performed during the newborn's first month of life, most commonly prior to discharge. A second screen was performed within 1-2 weeks of the first screen in the event of a failed test ("refer") in one or both ears. If the refer result occurred at the second screen in one or both ears, the family was referred to the audiologist for a diagnostic ABR. Families of newborns who passed in both ears after either the first or second screening received an "Orientation to Families" counseling session. The technicians gave the parents a guide of typical development for language and listening skills. The technicians also instructed the family to contact the technicians or audiologist immediately if the baby deviates from those guidelines or falls behind in their auditory/verbal milestones. 46% of those screened had no risk factors.

Newborns with one or more risk factors for hearing loss were classified as High Risk. Risk factors included:

- Low birth weight (14%)
- Prematurity (11%)
- Any exposure to ototoxic medications (5%)
- Use of mechanical ventilation (5%)
- Jaundice (3%)
- Asphyxia (1%)
- Time in an incubator (1%)
- Other factors - family history of hearing loss, syndromes, head/neck abnormalities (0.3%)

Newborns born premature and/or with medical complexities requiring time in the NICU are at higher risk for hearing loss (Bergman et al., 1985). Due to the higher prevalence of hearing loss, only one screening was performed before a referral to a diagnostic ABR [Fig. 2]. This reduced the length of time between the first screening and hearing aid fitting for newborns with PCHL.

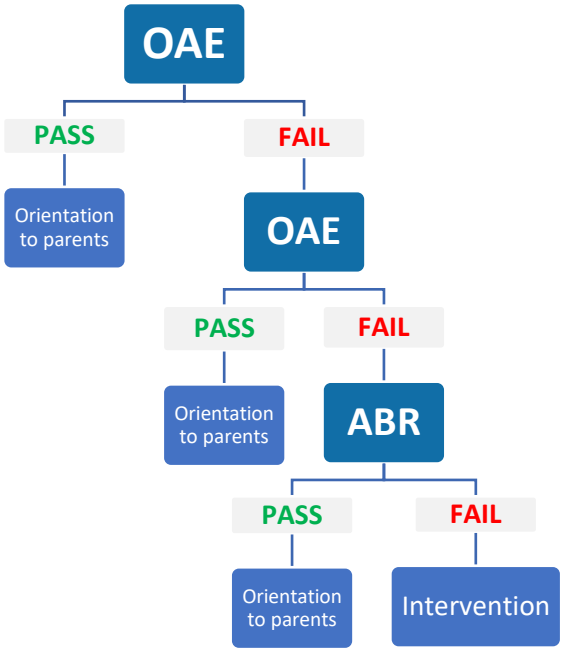

Fig. 1 The protocol for No Risk newborns

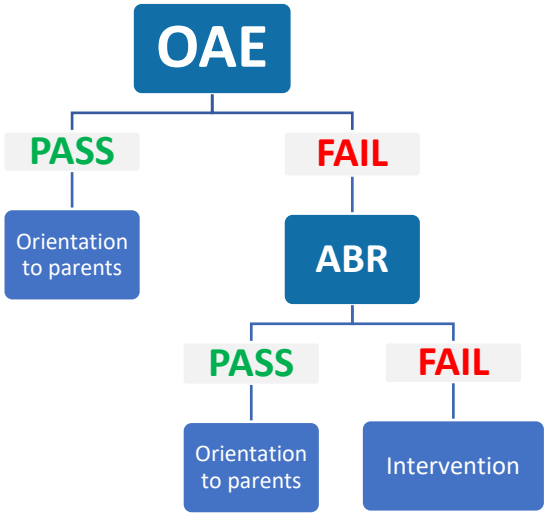

Fig. 2 The protocol for High Risk newborns

9. Results

Goal 2. To examine outcomes relative to the systematic review results.

As noted in the systematic review, the goals for many universal NHS programs are to achieve a high screening rate, a low referral rate and a low lost to follow up rate. Screening rate could not be assessed as the total number of births at these hospitals during this time period collection was not readily available. The referral rate after the first screen was 17.93%, which is within the range for lower and middle income countries assessed in the systematic review (Q1 = 8.08% Q3 = 22.19%, range = 0.23% – 31.94%). Improvement is noted, as previous data from January 2018 to October 2018 showed a first screen referral rate of 24.47%. This was mainly due to the learning curve with use of the screening equipment. When the technicians mastered the technique, the first screening referral rate drastically decreased. The program achieved an overall referral rate of 1.50%, which is near the average for the studies included in the systematic review. A total of 170 newborns (8.40%) were lost to follow up.

Ten newborns total were identified with permanent congenital hearing loss after a diagnostic audiological evaluation. This was a prevalence of 0.49%. Three had a severe hearing loss and six had profound hearing loss. These nine were fitted bilaterally with hearing aids. One newborn was identified with bilateral microtia and was fitted with a bone conduction sound processor.

As noted in the systematic review, JCIH recommends that infants be screened by 1 month of age, diagnosed by 3 months, and fit with hearing technology and/or placed in early intervention services by 6 months of age (Joint Committee on Infant Hearing, 2007).

The majority of newborns were screened once prior to discharge from the hospital, which meets JCIH recommendations. The ten newborns identified with PCHL were diagnosed on average by 5 months of age. The age at diagnosis ranged from 2 months to 9 months. The babies were fit with hearing technology on average by 8.4 months of age, with a range from 6 months to 12 months. The information can be seen on Figure 3. On average, the wait time between diagnosis and fitting was 3.4 months.

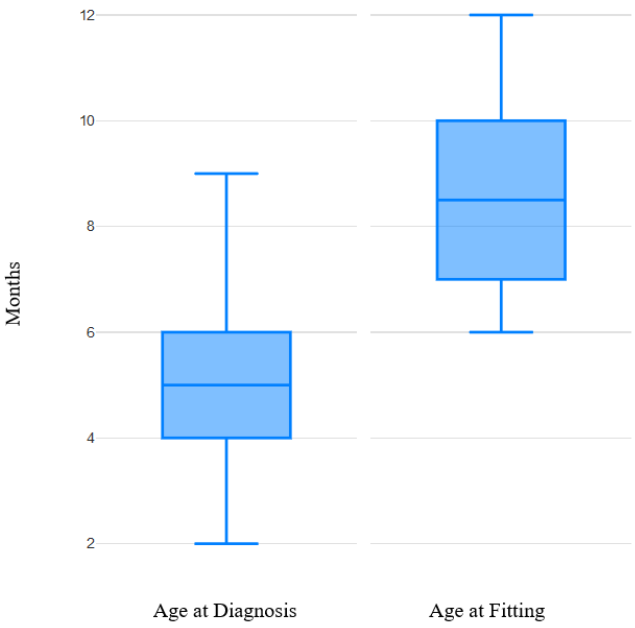

Fig. 3 Age at diagnosis and age at fitting for the ten newborns with PCHL

10. Discussion

Multiple barriers and challenges occurred during the implementation of the program. The challenges generally fit into three categories; the institutions where screenings are performed, family factors, and the screening process.

At the level of the institutions/hospitals, barriers included the bureaucracy for entering equipment. In two locations, the hospital staff was hesitant to let the technicians bring the screening equipment inside their building. Later on, they stated that they could not confirm if the equipment was their property or that of the non-profit organizations involved. They would also impose times that were already assigned to another hospital or inconvenient such as after 5pm. Also, a number of institutions were interested in screening NICU babies or High Risk babies only. High Risk babies are those with any of the hearing loss risk factors listed on page 11.

In regard to the families, some parents provided inaccurate or incomplete contact information. This led to a higher loss to follow-up rate. Some parents were unwilling to have their child screened for hearing loss. Another barrier was that families from distant places are difficult to locate for a re-test. The majority of second screens were conducted after discharge, in the family's home. The geography and logistics for reaching most parts of Guatemala contributed to the difficulty in precisely tracking families. At the national level in 2016, Guatemala's postal agency, Correos y Telégrafos de Guatemala, suspended mail service due to staffing and logistical difficulties. Particularly in small villages and indigenous communities, most families do not have a mailing address. The most common way to locate someone residing outside of major cities is to describe landmarks near them. For example, one family may state that they live near Lake Atitlan, in the village of Panajachel, down the road from Church of St. Francis of Assisi, before one reaches the primary school. The family may also describe the characteristics of the exterior of their home. Incorrect or difficult to follow directions, and the logistics of bringing the equipment to the family's location contributed to loss to follow up.

The screening process also played a role in the screening and referral rates. Some newborns left the hospital less than 24 hours after birth and were not screened. There has also been a learning curve for technicians to use the screening equipment proficiently. Another factor, which other hospital-based programs face, was the noise level of the NICU monitors and other medical equipment. Some hospitals also lacked appropriate space for screening.

The program is continuing to expand and reach more newborns. There are two new testing locations in Guatemala City, Hospital Roosevelt and Hospital San Juan de Dios. These two public hospitals have an average of 30 deliveries per day. Recently a fourth city, Sololá near the area of Lake Atitlan, was added to the program and data from the newborns screened in that location to date were included in this report. With the existing budget, a fourth OAE screener was purchased and Smiles that Listen covered the salaries, transportation and supplies for Sololá.

11. Conclusions and Future Plans

In the future, the program in Guatemala is looking to provide hearing screening in new locations across the country. Training has started for two audiology technicians for the Petén region in the north and two for Izabal in the far east. To implement the program in Izabal, they are in the process of purchasing another OAE device, which should arrive within May 2019.

They are also fundraising for OAE equipment and salaries for two technicians and a program coordinator to

run daily screenings in the newly added public hospitals in Guatemala City. Each hospital has a mean of 30 babies born per day.

Around May 2019, a travelling "AABR screening clinic" pilot system will be implemented. The NHS team will visit each of the screening sites at least once every 3 or 4 months. This frequency may be modified over time to fit the needs of the local population and the availability of screeners. During this day-long visit at the local screening site, all of the babies that need a diagnostic ABR in that area will be screened again. The AABR/Natus 2, the only automated ABR system in the country, will be used to conduct this screening. The high specificity and sensitivity of AABR should decrease the referral rate. The aim of this pilot system is to minimize the number of families that are lost prior to diagnostics and to decrease the number of families that need to travel to Guatemala City for a diagnostic ABR. On those same days, the OAE devices will be available to screen new babies.

The NHS program in Guatemala is new and quickly expanding across the country. The rate of congenital hearing loss requiring amplification in the group of newborns screened is 10/2024 (0.49%). This finding is near the interquartile range of the research assessed in the systematic review (Q1=0.14%, Q3=0.4125%). Coverage, referral and loss to follow rates show that a universal NHS program in Guatemala is feasible, and the preliminary results are in line with other pilot programs across the globe. Measures to expand the program should continue.

Author Contributions

All committee members contributed to revision of the paper critically for content and approval for the final version to be submitted. Dr. Patricia Castellanos de Muñoz facilitated the data collection for the Guatemala NHS program and consistently provided updates.

Financial Disclosure

The authors have no financial relationships relevant to this article to disclose.

Conflicts of Interest

The authors have no conflicts of interest to disclose.

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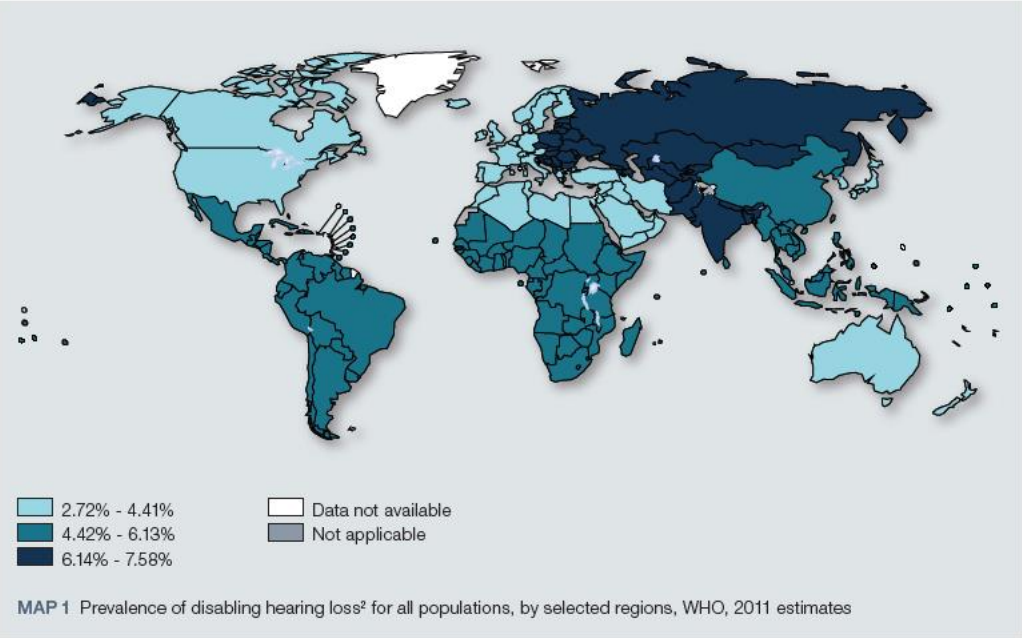
Appendix A – Table of Evidence

Author (Year)	Location	Study Design	WHO Region	World Bank Classification	Total Population	Number Screened	Percent Screened	Total Passed Screening	Percent Passed Screening	Birth Screening Performance Index	Referral Rate Overall (%)	Lost to Follow Up (%)	Prevalence of PCHL (%)
Abdullah et al. (2006)	Malaysia	Prospective Pilot Study	WPR	UMI	4219	3762	89.17%	3598	95.64%	85.28%	0.68	26.7	0.42
Ahmad et al. (2011)	Malaysia	Retrospective	WPR	UMI	16000	16000	100.00%	14406	90.04%	90.04%	0.47	33.87	0.09
Arslan et al. (2013)	Turkey	Prospective	EUR	UMI	2229	2229	100.00%	2207	99.01%	99.01%	1.84	0.64	0.36
Augustine et al. (2014)	India	Descriptive	SEAR	LMI	9671	9448	97.69%	9134	96.68%	94.45%	1.73	17.38	0.41
Benito-Orejas et al. (2008)	Spain	Retrospective pilot study	EUR	HI	5611	5571	99.29%	5502	98.76%	98.06%	2	6	0.54
Berninger & Westling (2011)	Sweden	Prospective	EUR	HI	31727	31092	98.00%	28294	91.00%	89.18%	6	1.1	0.18
Bevilacqua et al. (2010)	Brazil (São Paulo state)	Descriptive	AMR	UMI	12667	11466	90.52%	10581	92.28%	83.53%	3.33	19.75	0.09
Caluraud et al. (2015)	France	Prospective	EUR	HI	101916	101724	99.81%	100979	99.27%	99.08%	1.74	10.87	0.14
G. Chen et al. (2012)	China (rural areas)	Cohort	WPR	UMI	12968	11568	89.20%	10580	91.46%	81.59%	4	26.2	0.5
X. Chen et al. (2017)	China (Shanghai)	Retrospective	WPR	UMI	1682012	1574380	93.60%	1512009	96.04%	89.89%	1.08	31.31	0.17
Cianfrone et al. (2018)	Italy (Rome)	Retrospective	EUR	HI	6200	4719	76.11%	4595	97.37%	74.11%	3.1	29.9	0.14
Connolly et al. (2005)	United States (Mississippi)	Retrospective database review	AMR	HI	17602	17602	100.00%	16886	95.93%	95.93%	4.07	0.4	0.44
De Capua et al. (2007)	Italy (Siena area)	Retrospective	EUR	HI	21125	19700	93.25%	19410	98.53%	91.88%	0.97	16.46	0.18
Finitzo et al. (1998)	United States (Texas)	Cohort	AMR	HI	54228	52508	96.83%	50721	96.60%	93.53%	3.4	31.5	0.22
Gilbey et al.	Israel (Zefat)	Retrospective	EUR	HI	5496	5334	97.05%	5277	98.93%	96.02%	4.59	2.29	0.04

(2013)													
Govaerts et al. (2001)	Belgium	Prospective	EUR	HI	2008	1995	99.35%	1913	95.89%	95.27%	0.35	0.17	0.3
Habib & Abdelgaffar (2005)	Saudi Arabia	Descriptive	EMR	HI	13071	11986	91.70%	11964	99.82%	91.53%	2.5	0	0.18
Huang et al. (2013)	Taiwan	Retrospective cohort	WPR	HI	15930	15790	99.12%	15631	98.99%	98.12%	1.01	5.6	0.29
Jewel et al. (2013)	India (Ludhiana)	Prospective	SEAR	LMI	1000	1000	100.00%	978	97.80%	97.80%	0.4	30	0.4
John et al. (2009)	India	Cross sectional	SEAR	LMI	500	500	100.00%	497	99.40%	99.40%	1.6	0	0.6
Khairi et al. (2005)	Malaysia	Prospective cross-sectional	WPR	UMI	401	401	100.00%	387	96.51%	96.51%	1.24	29.03	1.25
Kim et al. (2017)	South Korea (Rep.)	Retrospective quasi-experimental cohort	WPR	HI	13805	7403	53.63%	7301	98.62%	52.89%	1.4	18	0.2
Korres et al. (2006)	Greece	Retrospective	EUR	HI	25032	25032	100.00%	24662	98.52%	98.52%	0.9	58.2	0.24
Lévêque et al. (2007)	France	Prospective	EUR	HI	36652	33873	92.42%	33828	99.87%	92.30%	1.3	2.5	0.08
Levit et al. (2015)	Israel (Tel Aviv)	Cohort	EUR	HI	17097	17078	99.89%	16955	99.28%	99.17%	0.36	1.17	0.25
Lin et al. (2002)	Taiwan	Quasi-experimental one-group post-test design	WPR	HI	6765	6765	100.00%	6335	93.64%	93.64%	6.4	0.21	0.52
Lin et al. (2005)	Taiwan	Retrospective two-group	WPR	HI	21273	21273	100.00%	20163	94.78%	94.78%	1.8 - 5.8	19.19	0.43
Lin et al. (2007)	Taiwan	Retrospective three-group	WPR	HI	25588	25588	100.00%	24445	95.53%	95.53%	0.82 - 5.78	0.84	0.42
Martines et al. (2007)	Italy	Prospective	EUR	HI	1191	1068	89.67%	1058	99.06%	88.83%	0.94	0	0.19
Mathur & Dhawan (2007)	India	Prospective randomized	SEAR	LMI	1000	1000	100.00%	955	95.50%	95.50%	21	20	1.5
Molini et al.	Italy (Umbra	Cohort	EUR	HI	20841	19557	93.84%	18664	95.43%	89.55%	0.55	29.49	0.36

(2016)	region)												
(Ng et al. (2004)	Hong Kong	Descriptive study & questionnaire	WPR	HI	1076	1064	98.88%	1027	96.52%	95.45%	3.5	0.19	0.38
O'Connor et al. (2013)	Ireland	Descriptive	EUR	HI	11763	11738	99.79%	11213	95.53%	95.32%	4.47	0.3	0.13
Ohl et al. (2009)	France	Cohort	EUR	HI	1464	1461	99.80%	1259	86.17%	86.00%	10.68	19.7	3.15
Olusanya et al. (2008)	Nigeria	Cross-sectional pilot study	AFR	LMI	1347	1330	98.74%	1311	98.57%	97.33%	3.5	12.84	0.53
Pastorino et al. (2005)	Italy (Milan)	Prospective	EUR	HI	19901	19777	99.38%	19636	99.29%	98.67%	3.12	12.77	0.32
Pisacane et al. (2013)	Italy	Retrospective	EUR	HI	182188	146026	80.15%	144803	99.16%	79.48%	1.9	0.75	0.11
Qi et al. (2013)	China (Beijing)	Cohort	WPR	UMI	11104	10983	98.91%	9885	90.00%	89.02%	2.08	43.13	0.32
Rojas et al. (2014)	Colombia	Observational with bivariate analysis	AMR	UMI	56822	56752	99.88%	55875	98.45%	98.33%	0.29	49.68	0.09
Rouev et al. (2004)	Bulgaria	Prospective cohort	EUR	UMI	1750	1672	95.54%	1656	99.04%	94.63%	1.44	4.4	0.17
Saki et al. (2017)	Southwestern Iran	Cross-sectional group design	EMR	UMI	92521	92521	100.00%	91360	98.75%	98.75%	1.25	4.23	0.24
Vignesh et al. (2015)	India (Chennai)	Prospective	SEAR	LMI	1405	1405	100.00%	1374	97.79%	97.79%	2.21	0	0.14
Vohr et al. (2001)	United States (multiple)	Retrospective analysis	AMR	HI	13748	12081	87.87%	9910	82.03%	72.08%	3.22	12	0.2
Wu et al. (2011)	Taiwan	Prospective cohort	WPR	HI	1017	1017	100.00%	979	96.26%	96.26%	3.7	0	0.79
Yee-Arellano et al. (2006)	Mexico	Descriptive	AMR	UMI	3069	3066	99.90%	3059	99.77%	99.67%	0.23	0.03	0.16

Appendix B



MAP 1 Prevalence of disabling hearing loss[2] for all populations, by selected regions, WHO, 2011 estimates

Appendix C

Place	1st Screen	Pass	Fail	2nd Screen	Pass	Fail	ABR Full Eval	Lost babies
Guatemala	973	806	167	91	86	5	3	77
San Cristóbal	306	288	18	16	14	2	2	2
Mazatenango	433	332	101	36	30	6	5	66
Sololá	147	114	33	16	13	3	2	18
Quetzaltenango	165	121	44	42	30	12	7	7
TOTAL	**2,024**	**1,661**	**363**	**201**	**173**	**28**	**19**	**170**

Place	Hearing loss	Rate of hearing loss found	% of babies w/ Hearing Loss
Guatemala	2	10/2024 (0.49%)	20%
San Cristóbal	1		10%
Mazatenango	5		50%
Sololá	0		-
Quetzaltenango	2		20%

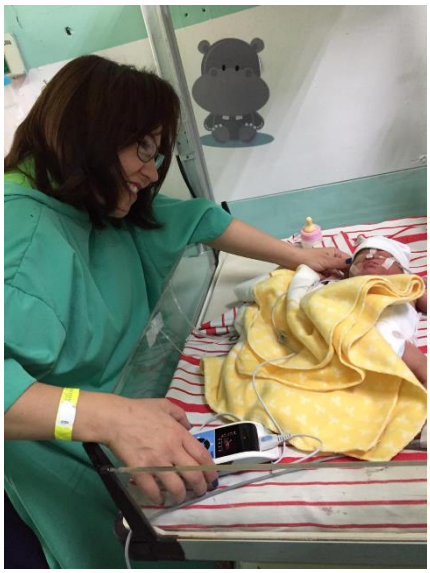
Hearing loss	Degree		Fitted w/ HA
10	Mild	0	
	Moderate	0	
	Severe	3	✓
	Profound	6	✓
	Bilateral microtia	1	✓

Appendix D

The five currently running newborn hearing screening locations. Izabal, the red star, will start May/June 2019.

Appendix E

Mazatenango



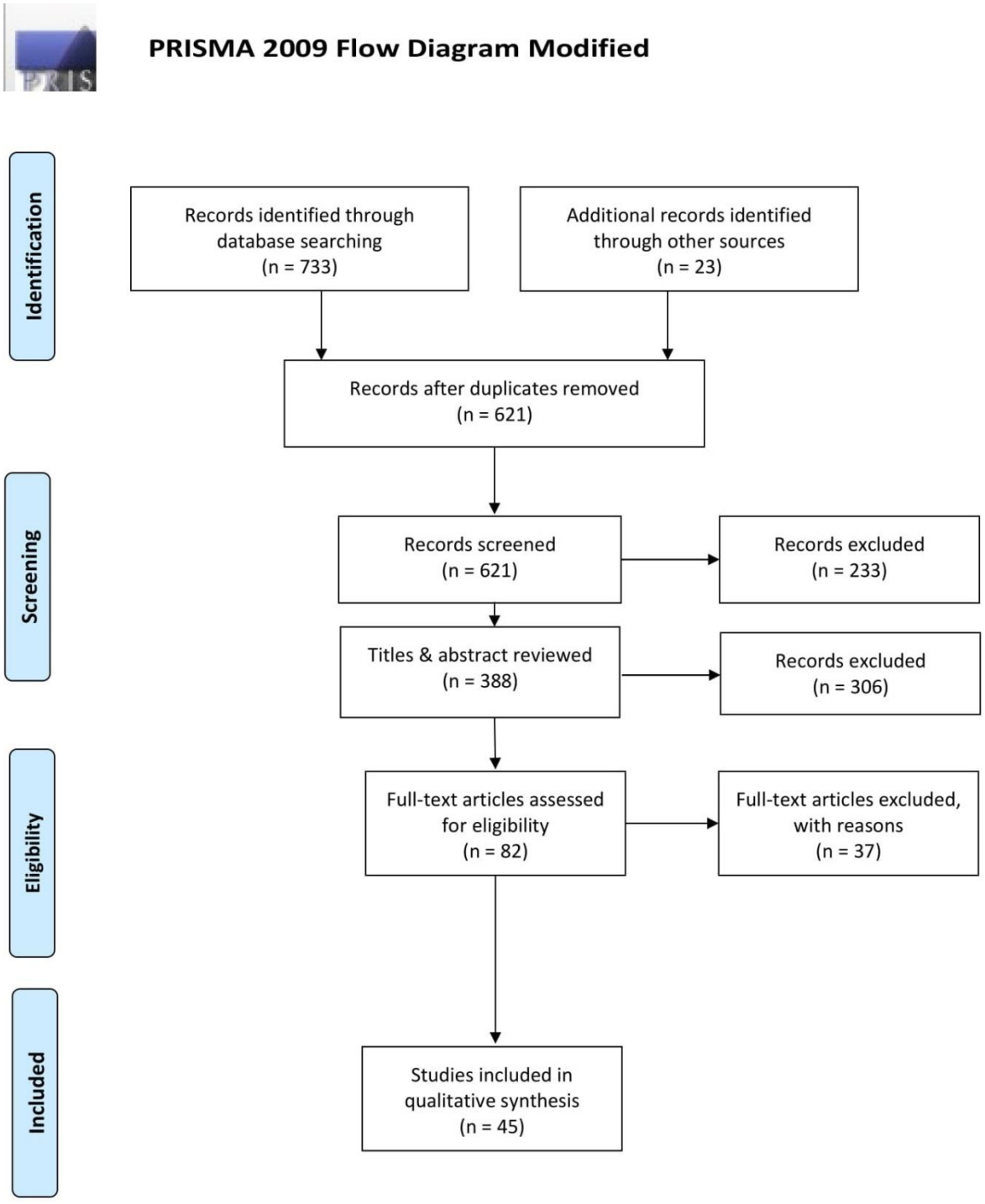
Guatemala City

Sololá

San Cristobal

Quetzaltenango

Appendix F



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