Newborn Hearing Screening and Beyond: A Continuing Journey in the Philippines

This keynote lecture is a product of many years of hard work but today this is delivered in honor of Professor and Founding President of ORLIAC, Professor Emeritus Jan Veldman. Thank you for the opportunity to host ORLIAC in Manila way back in March 2018. For those who attended this, I gave a special lecture during the celebration of World Hearing Day on March 3, 2018, when ORLIAC was held in Manila and this gives an update on how we managed in the past four and a half years, with a COVID-19 pandemic in the past two and half years.

The Philippines is an archipelago of more than 7,100 islands during high tide and up to 7,600 when it is low tide. It is situated in the Southeast Asian region. It is one of the countries with the highest population density, with a total population of 110 million spread over 300,000 square kilometers. The crude birthrate is at 19.9 per 1000 – in stark contrast with surrounding Southeast Asian nations now with a decreasing population such as Thailand, Vietnam, and Malaysia among others.

There have been three World Health Assembly (WHA) resolutions that emphasized the prevention of deafness and hearing loss. WHA 38.19 in 1985 then ten years later WHA 48.9 in 1995 and the last WHA 70.13 in 2017. These resolutions from the World Health Assembly emphasized that prevention of deafness and hearing loss should be incorporated in primary health care. As a result of the 2017 WHA 70.13 resolution, a World Hearing Report[1] was formulated and released last year in 2021 (Figure 1).

The WHO Call to Action in 2000 recommended that Universal Newborn Hearing Screening (NHS) be implemented in all countries where rehabilitation services are established.[2] In 1998, we did the first cochlear implantation in my country[3] and established as well the graduate program of Masters in Clinical Audiology at the UP College of Medicine where I now work as Dean. This graduate program is one of only two in the country and is government-subsidized such that slots are limited and entry is quite competitive. It is jointly offered with the College of Allied Medical Professions,[4] We consider this program to be a key element in producing the necessary healthcare workforce needed for the implementation of programs to defeat deafness in my country.

The world hearing report published last year by the World Health Organization (WHO) highlighted the increasing number of people living with hearing loss and in need of services. There is of course the disproportionate burden of hearing loss in low- to middle-income countries like the Philippines.[6]

Our publications from 2003, from a study looking at results of newborn hearing screening in the neonatal intensive unit in the hospital[7,8] then to the community, thus providing the evidence for the eventual policy on UNHS that we proposed to the Department of Health (DOH) and to congress for legislation.

We initially embarked on basic epidemiological studies using otoacoustic emissions testing in the hospital then in the community. We noted the age of referral at our hospital for children with hearing impairment to be at around 34 months.[9] Our focus then was just providing otological clinical services so we decided to work for the establishment of the Philippine National Ear Institute (PNEI) – a research institute formed by Republic Act 9245 and part of the National Institutes of Health of the University of the Philippines – Manila.[10] The PNEI laid down the researches needed for the stable foundation of a national health program focused on newborn hearing. From 2003 to 2008, we conducted several studies to establish the prevalence of bilateral permanent hearing loss in newborns both in the hospital and community settings as a prelude to the enactment of the law, and in order to defend having a program on newborn hearing, we conducted a cost-analysis of hospital-based universal newborn hearing screening.[11] Notably in 2007, a population-based study showed the prevalence of bilateral profound Congenital Hearing Loss at 1.4 per 1000 births.[12] This translates to **more than 3,000 babies** annually or **8 babies** per day who may be profoundly deaf in my country.[13]

If there were proper intervention for a child with hearing impairment, the cost of treating hearing translates to a lifetime savings of about 80,000 dollars or about PhP 4.3M for the patient's family. The government and the family would have spent about PhP 4.3 M to raise, educate, and support a deaf-mute child to adulthood. This was presented to the Senate of the Philippines and highlighted during the Inaugural Congress of the ASEAN Academy of Neuro-Oto-Audiology (AANOA). This was supported by Senator Loren Legarda, PNEI Director Dr. Generoso Abes, AANOA founding member Dr. Helmi Balfas, IFOS Regional Secretary Chong Sun Kim, PSOHNS President Gil Vicente, AANOA President Dato Lokman Saim, and Hearing International Secretary Dr. Norberto Martinez (Figure 2).

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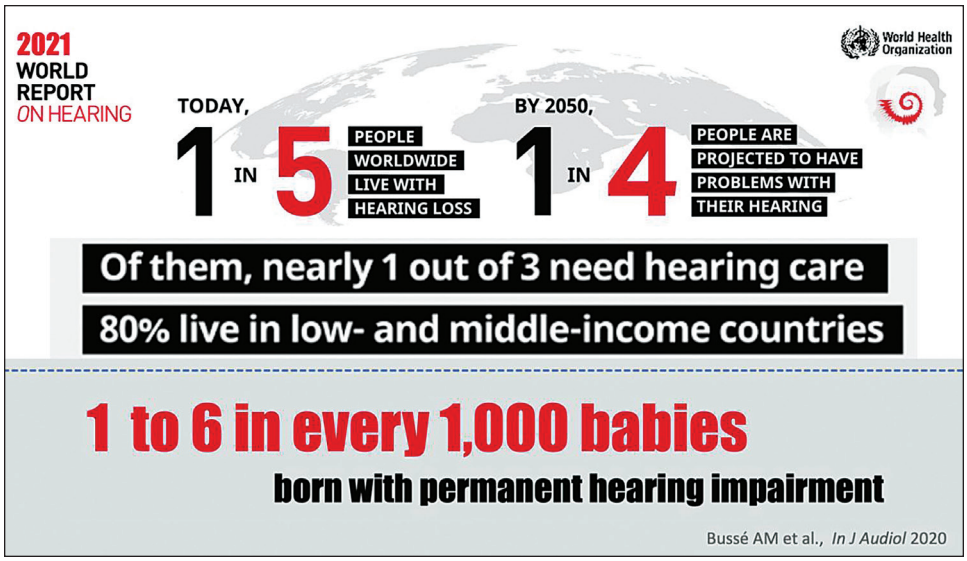

Figure 1. World Hearing Report in 2021.[1,5]

Figure 2. AANOA supported UNHS during the welcome ceremonies on November 2008.

Mandating NHS in the country also entailed involving stakeholders, including otolaryngologists from the different regions who committed to convince their local officials and local hospital administrators the need for instituting these newborn hearing screening programs. After we had the local data at hand, the PSOHNS created a task force on NHS and crafted a position statement on the need for UNHS. As then Vice President, we drafted the position paper that will be presented to Congress while at the same time seeking support from the Department of Health under DOH Secretary Francisco Duque who agreed that Philhealth should be able to support this program when enacted into law. A Technical Working Group was formed, gathering all stakeholders and service providers. Multiple meetings were held, groundwork for launch, and implementation of newborn hearing screening program were instituted with ten Collaboration for Newborn Hearing Screening Advocacy (CONHScA) annual symposia with otolaryngology, audiology, and other hearing screening advocates all over the country.[14] (Figure 3)

On August 12, 2009, President Gloria Macapagal-Arroyo signed RA 9709, an act that established universal newborn hearing screening program for the prevention, early diagnosis, and intervention of hearing loss.[15]

On the modality to be used, reporting, accreditation and training, monitoring and evaluation, and most especially costing of the services and financing for sustainability were also done. We advocated for legislation on UNHS, and once ratified, developed a national program with the Department of Health as lead agency and continued with policy implementation as part of a national technical working group under the DOH. This led to the creation of national Newborn Hearing Screening Reference Center (NHSRC)

Figure 3. CONHScA stands for Collaboration for Newborn Hearing Screening Advocacy which sponsored annual meetings until the full implementation of Universal Newborn Hearing Screening.

Figure 4. National television interview for Newborn Hearing Screening awareness.

that was inaugurated in 2013 at the National Institutes of Health with Professor Cor Cremers of Radboud University Nijmegen as special guest. We also started to increase the awareness on the importance of NHS even on national television (Figure 4).

Aside from the cost-effectiveness study of hospital-based newborn hearing screening program, we also looked at the budget impact of a community-based UNHSP in the Philippines from both the public payer and the societal perspectives. This study of Rivera et al. published in 2017[16] showed that cost effectiveness is sensitive to treatment rate, prevalence, follow-up rate, number of rehabilitation sessions, and coverage of the program. It was not sensitive to cost per rehabilitation session, cost of diagnosis with OAE and ABR, education costs, refer rates, recurrent costs, cost of machines, and sensitivity rates. From the societal perspective, the UNHSP was found to be cost-saving for the full range of parameters tested for cost of screening, amplification,

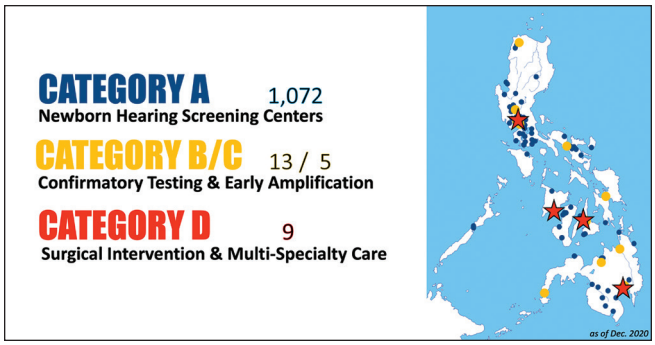

Figure 5. Distribution of the different Newborn Hearing Screening centers in the country.

education, rehabilitation, and fixed program costs. Ensuring treatment of at least 31 percent and follow-up rate of 24% for a community-based newborn hearing would likely be important benchmarks.

The technical arm of the lead agency for this program, the Department of Health, is the Newborn Hearing Screening Reference Center that gives assistance in defining and recommending NHS testing and follow-up protocols which include hearing screening methods, devices used, location, manner, and timing of newborn hearing screening testing. The current protocol uses the 2007 JCIH recommendation of 1-3-6 rule, with screening at 1 month, confirmation of hearing loss at 3 months, and appropriate intervention at 6 months of age.[17]

With respect to the preferred method of screening, the recent study by Neumann K et al. showed OAE as most prevalent in the country.[18] Ten years ago in 2010, an initial web registry for reporting of the OAE results was formulated and up to 2014 it was used in 9 centers which allowed gathering of preliminary data on the NHS program. From the data, there was a registry card that needed manual data encoding and in order to sustain operations, a 1 USD fee was levied per registration and was reimbursable thru Philhealth that paid around 4 USD per hearing screening test done. Personnel training, device, and facility certification standards were implemented for centers that chose to perform newborn hearing screening testing. Tiered categorization of centers was also done with screening centers as Category A, screening and diagnostic centers as category B, screening, diagnostic, and essential intervention with hearing aid amplification as category C, and the highest category D for centers with genetic testing and counselling, cochlear implantation surgery and speech rehabilitation services. Recent data showed there were 1072 category A, 18 category B and C, and 9 category D centers distributed all over the country (Figure 5).

While some services were initially hampered by the COVID-19 pandemic, NHSRC defined safety protocols that needed to be followed. In an updated advisory, first released in April 2020, and is still in effect to date, NHSRC emphasizes that the centers should follow hospital/institution's procedural protocol regarding disinfection and attire. Hearing tests in infants are non-invasive and non-aerosol producing procedures. The advisory also included recommendations in terms of timing, preparation, and testing procedures.

One significant development that was accelerated during the pandemic was the online adaptation of the NHS personnel certifying course that was reported in recent publications by Rozul et al.[19,20] To date, there have been about 3403 trained personnel in 1099 centers with significant increase noted in 2019 compared to previous years. This was not however reflected in the report of Neumann probably due to the time when data for this publication was collected prior to 2019. From the registry alone, the percent screened in the Philippines has been reported to be at least 7-13 percent of live births from the years 2019-2021. The report from Philhealth of 800 claims for NHS from 2018-2019 is still under verification. In 2020, based on manual submission, out of 1099 facilities, 26 percent submitted reports. In 2019, 95% already submitted reports so the pandemic impacted significantly on reporting of results by excel file and paper reports (Figures 6 and 7).

The program is still beset with challenges like poor compliance with data submission, loss to follow-up, poor connectivity, sustainable funding of the program from both local and national government, lack of human resources, and a need of much awareness among stakeholders regarding the importance of newborn hearing screening. In a country where 60 percent die without seeing a doctor, there is much work to do. The cost of screening equipment is a major barrier so we conceptualized a biomedical device development project working with engineers in the university to create an AABR screening device which is now on phase two with a TLR 5 early phase rating scale and will now include recruitment of more subjects and refinements for improvements. Harnessing technology and making this more affordable remain as strategies for developing a sustainable hearing screening program and is the subject of HELE, which aims to increase the rates of newborn hearing screening with novel technologies and telehealth. "Hele" stands for lullaby a mother sings to a child. We thought this as very apt for emphasizing the importance of hearing from birth for optimal development. This received a substantial phase 1 grant in 2016 and a phase 2 was launched this year for the premarket development which now stand at technology readiness level 5. It has already spawned many products like the computer-based e-learning training modules, capacity building with provision of basic OAE equipment, and many research publications. It is hoped that a pre-production HELE device can already be produced soon with validated efficacy and reproducibility of all the AABR responses in a clinical setting. With the collaboration of University of California Berkeley and UC Davis, a formidable team has been created. The electronic national newborn hearing screening registry was launched during WORLD HEARING DAY in

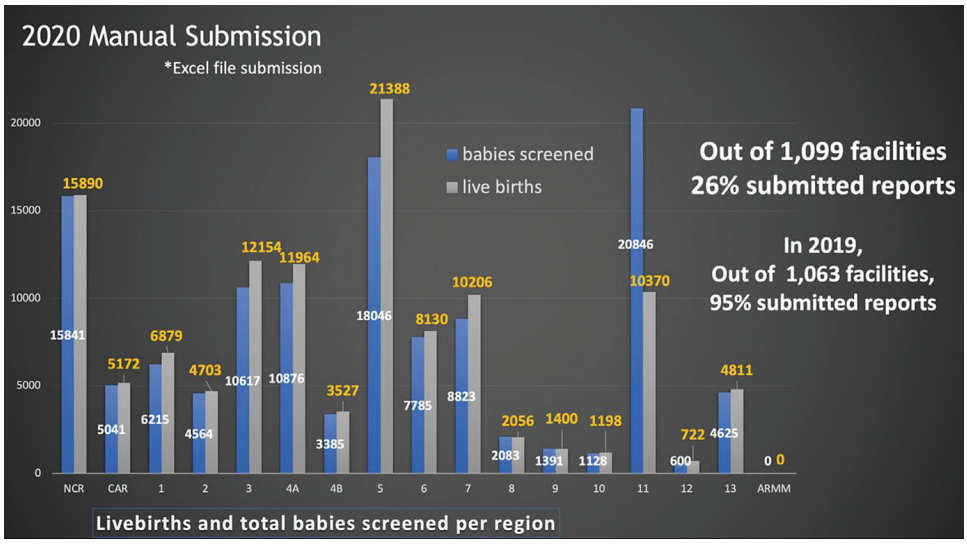

Figure 6. Newborn Hearing Screening registry report.

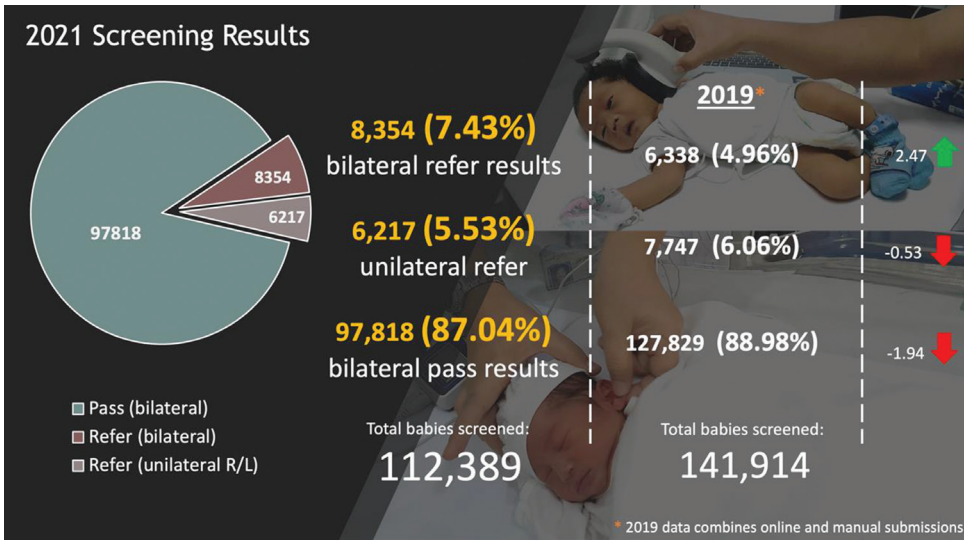

Figure 7. Newborn Hearing Screening results from online and manual submissions.

2022 and the usability testing results have been published by Ricalde et al.[21] In the recent manual of operations and procedures, success indicators were outlined. This is aligned with the WHO standards for monitoring and evaluation, and determining success of programs.

Beyond NHS, what else have we learned? Our studies on the genetic causes of the more common causes of hearing impairment such as otitis media showed a unique mutation A2ML1 that affected protease inhibitors allowing better mucosal defense of the middle ear mucosa. Our findings showed microbiome shifts and when we looked at gingivitis, this was what we found. The genetic counseling we embarked on for this indigenous community taught us clinicians many lessons. Speaking to them in their native language was important and house to house invitations were more effective.

What about genetic basis of congenital hearing loss? We have found that SLC26A4 mutations were more common than GJB2. There were novel mutations seen in our cohort suggesting the need for studies looking at genetic predisposition. We saw commonalities with Indonesia in terms of the prevalence of GJB2 mutations. From our studies on A2ML1 mutations predisposing to otitis media, we are monitoring this with ongoing study on Hispanic populations.

Meanwhile, a seed fund for a national cochlear implant program was approved by Congress and 20 children underwent surgery (Figure 8), while two centers, one in Visayas (Corazon Locsin Montelibano Memorial Regional Hospital in Bacolod) and another in Mindanao (Southern Philippines Medical Center in Davao) were capacitated. Virtual multidisciplinary meetings to discuss the cases were

held and provided a model for optimizing use of resources and outcomes. The initial data from the 20 implanted children are very encouraging.

There are goals of expanding this program to more beneficiaries by having a Z package for Cochlear implantation, funding from DOH medical assistance fund for indigent patients, and development of services for other implantable hearing devices at the Philippine General Hospital.

Moreover, research on hearing need more support given that our studies on genetics of hearing loss among Filipinos yielded very important data on novel genes, uniqueness of our genetic pool as well as mutations predisposing to increased

Figure 8. Cochlear implant surgeries done at Philippine General Hospital as part of the National Cochlear Implant Program.

Figure 9. Dr. Charlotte Chiong as panelist during the UN World Health Assembly Advocacy event on hearing health moderated by Barbara Bush, CEO and co-founder of Global Health corps at the UN Headquarters, New York last September 14, 2017.

Figure 10. Science Policy and Information Forum on Program Development for Hearing Health.

prevalence of otitis media among our marginalized and indigenous population.[22-26] As a low- to middle-income country, these researches will need to be continued and may provide valuable lessons for similarly challenged countries in the works as we highlighted in 2017 during the United Nations World Hearing Assembly Advocacy Event on hearing health (Figure 9).

The recent National Academy of Science and Technology health policy forum where Professor Xing Kuan Bu was featured as keynote lecturer gave important data on their experiences on hearing health program development in China and in the world (Figure 10).

Clearly the sustainable development goals especially 3,4,8 and 10 covered by hearing health ensures equity and should be aspirational goals for national development. This is congruent with the vision of PNEI: "No Filipino shall be deprived of a functioning sense of hearing and balance."

Lastly, as the theme of this congress is East Meets West, the Philippines had a strong history of shipbuilding in an era when the galleon trade was very active and 148 of 200 ships that plied that route were ships built in the Philippines with our hardwood. Tracing therefore the history of the countries in ASEAN in particular with the Dutch, French, British, and Spanish influences, we need to dig deeper and look at genetic markers for both health and disease so that while we develop our friendship and scientific connections, history will prove that we have always been connected in so many ways and further exploration can be done in many dimensions.

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