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## Children At-Risk For Hearing Impairment: A Retrospective Study Of The Ontario Infant Hearing Program Population

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A thesis submitted in partial fulfillment of the requirements for the degree in Master of Science  
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CHILDREN AT-RISK FOR HEARING IMPAIRMENT: A RETROSPECTIVE  
STUDY OF THE ONTARIO INFANT HEARING PROGRAM POPULATION

(Thesis format: Monograph)

by

Katherine Mary Smith

Graduate Program in Health & Rehabilitation Sciences

A thesis submitted in partial fulfillment  
of the requirements for the degree of  
Master of Science

The School of Graduate and Postdoctoral Studies

The University of Western Ontario

London, Ontario, Canada

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## Abstract

There is widespread agreement that infant hearing screening programs are effective but such programs may fail to detect all hearing impairment and children can develop subsequent hearing loss after passing the initial screen. This is the core rationale for surveillance programs that are analyzed in this thesis. Infants with hearing risk factors are followed using surveillance programs that include monitoring by audiological assessment.

The study population in this thesis consists of 2,390 children with normal hearing and 248 children with hearing impairment from different referral routes. The Infant Hearing Program Surveillance group is 1.48% of the number of hearing-impaired children. The thesis methodology identifies children with at least one risk factor and then analyzes the different referral routes and hearing loss features. It concludes that there is a need for further evaluation and improvements in surveillance programs, as well as all parts of screening and habilitation.

## Keywords

Neonatal Screening, Hearing Loss, Surveillance, Referral Route, Risk Factors

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## Chapter 1

### 1 Introduction and Literature Review

#### 1.1 Introduction

Early Hearing Detection and Intervention (EHDI) programs for infants have been implemented with universal newborn hearing screening and treatment for congenital hearing loss in many developed countries. As per Yoshinaga-Itano (2003), there is a critical period for early language development within the first six months of life. This sensitive period derives from behavioral and neurological factors that can affect timelines for aspects of development, such as socio-emotional, auditory, speech, and language (Yoshinaga-Itano, 2004).

Permanent childhood hearing impairment in children affects the development of auditory speech perception, speech production, and English language acquisition (Yoshinaga-Itano, 2003). Permanent childhood hearing impairment has prevalence rates of 1 to 2 per 1000 live births as defined by 40 decibels (moderate) or worse hearing loss in the better ear (Fortnum, 2003). However, when mild hearing loss cases are included, with a definition of loss greater than 25 dB unilaterally, the prevalence is about 2-3/1000 (Hyde, 2005). The severity of hearing impairment varies and there are challenges in identifying some permanent childhood hearing impairments, such as progressive or late onset hearing loss (Hutt & Rhodes, 2008).

Universal newborn hearing screening programs result in earlier diagnosis of hearing loss and early detection enables early intervention services for affected families (Yoshinaga-Itano, 2003). It has been demonstrated that screening in hospitals results in identification of 75% of those with hearing impairment in the first three months of life

versus 75% identified by as late as 30 months in hospitals without screening (Yoshinaga-Itano, 2003). In previous generations, children were identified as late as two and a half years of age, after significant developmental opportunities have passed (Yoshinaga-Itano, 2003).

In the past decade, in most developed countries, early detection with universal infant hearing screening programs has made early intervention a priority, allowing health care professionals to immediately address the child's communication developmental needs (Joint Committee on Infant Hearing, 2007). Screening programs in general are considered ethically fitting in the presence of the appropriate procedures and resources available. This enables timely access to confirmation of impairment, diagnosis, and intervention services that are considered effective, as specified by the World Health Organization (Wilson & Jungner, 1968).

The current standard for EHDI programs is to complete the hearing screening in all infants by one month of age followed by audiological assessment by three months of age in those identified as at-risk for hearing loss based on the physiologic auditory screening outcome (e.g., otoacoustic emissions or automated auditory brainstem response testing); for those with confirmed hearing impairment, intervention should begin by six months of age (JCIH, 2007).

Infants who pass newborn hearing screening but have one or more hearing risk factors, such as low birth weight, as listed in the Joint Committee on Infant Hearing (JCIH) 2007 guidelines, should have at least one diagnostic audiological assessment by 24 to 30 months of age (JCIH, 2007). Additionally, the JCIH recommends that all infants with certain risk factors for other speech or language impairments, like progressive or late

onset hearing loss, as well as auditory neural conduction disorders, such as auditory neuropathy spectrum disorders (ANSD), should be followed using surveillance programs for long-term communication development (JCIH, 2007). These at-risk children are mandated to receive universal neonatal newborn hearing screening, and in addition, ongoing medical, speech and language, and audiological surveillance. This is recommended as per the JCIH (2007) as a standard even if the child passes the newborn hearing screening protocol (JCIH, 2007).

Improved outcomes for children with congenital hearing impairment are associated with confirmation and intervention by six months of age (Fortnum, 2003). Those infants who pass the initial infant screening with risk indicators for hard-to-detect impairments are considered especially susceptible. While on-going monitoring is ideal, few EHDI programs have sufficient resources to implement comprehensive surveillance programs (JCIH, 2007). This makes it difficult to evaluate the effectiveness of different surveillance and assessment protocols for difficult-to-detect hearing impairments, including ANSD and progressive, acquired, or delayed-onset hearing loss. In addition, there is a need for comprehensive program integration of the EHDI data that follows the same protocols to assist in data sharing and data analysis. For example, it would be useful to standardize the Infant Hearing Program protocol requirements nationally.

## 1.2 Literature Review

### 1.2.1 Early Detection

The goal of international EHDI programs is to identify children with permanent auditory impairments in order to improve their communication outcomes in the areas of hearing, speech, and language. It has now been well established that unidentified hearing

loss has a negative impact on communication development and that earlier identification of hearing loss leads to better results in communication, educational achievement, and social development in children (Robinshaw, 1995; Moeller, 2000; Yoshinago-Itano et al., 2000).

Early identification has been associated with better language development regardless of the method of communication (Yoshinago-Itano, 2003). Specifically, mean language quotients were demonstrated to be better with early identification regardless of cognition levels (high or low), as well as vocabulary development, and speech ability. Moreover, early identification was shown to be especially important for those with additional disabilities, such as autism spectrum disorders (Yoshinago-Itano, 2003).

Yoshinago-Itano, Coulter and Sedey (1998) studied 150 deaf and hard-of-hearing infants and children. Children who were identified and put into intervention services by six months of age were found to have considerably improved language development compared to those who did not receive early intervention. The researchers concluded that access to language within this early period is critical for developing language in the developmentally sensitive period (Yoshinago-Itano et al., 1998). Additionally, children with hearing loss who were born in hospitals with newborn hearing screening were 2.6 times more likely to have language development within the normal range than children with hearing loss born in hospitals without screening (Yoshinaga-Itano et al., 2000).

A study by Durieux-Smith, Fitzpatrick, and Whittingham (2008) compared 709 children who were referred for hearing aid fitting and children were either diagnosed by neonatal screening or were diagnosed via medical referral. Children who were screened were identified significantly earlier (mean 6.3 months) than referred children (mean 39.5

months). Earlier identification enables families to obtain information and receive appropriate counseling and support while early intervention allows families to prevent certain language delays from occurring (Yoshinaga-Itano, 2003), supporting the notion that this early period is critical for language development. Specifically, families may experience anxiety and frustration if the child is not attaining developmental milestones and they may attribute this falsely to “natural” development (Hyde, 2005). The current detection tools in countries with mandated screening protocols are the most effective way to reliably detect the required range of hearing impairments in infants below six months of age. These instruments include modern physiologic screening tests, in addition to modern, objective, diagnostic hearing assessment and surveillance in infants who do not pass screening or who are at risk for hearing loss (Hyde, 2005).

### 1.2.2 Universal Newborn Hearing Screening

Historically, prior to the implementation of newborn hearing screening programs, outcomes for children with severe to profound hearing loss were dismal, with adolescents graduating from high school with on average a grade three or four reading level and language levels of a nine to ten year old child with normal hearing (Traxler, 2000). In 1993, the National Institute of Health (NIH), as endorsed by the National Institute of Deafness and Other Communication Disorders (NIDCD) held a Consensus Development Conference. This was a milestone in endorsing screening of all newborns prior to hospital discharge (NIH, 1993). It has been suggested that the main problem with previously used targeted screening of infants with risk factors is that about 50% of infants with permanent childhood hearing impairment were not identified with any of the



targeted risk indicators. Therefore, only around 50% of the total hearing-impaired cases were identified with this method (Hyde, 2005).

As standards continued to increase for early detection, the JCIH 2000 statement recommended universal newborn hearing screening (JCIH, 2000). By 2005, 95% of infants in the United States had been screened, in contrast to 1993 where only 11 hospitals in the United States were screening 90% of infants in their catchment areas (JCIH, 2007). The JCIH principles include hearing screening by one month; screen fails should receive an audiological evaluation by three months and intervention services by six months. The JCIH also recommended the use of risk factors as a supplement to universal newborn hearing physiological screening protocols. The recommendation was that infants from birth to six months should have an evaluation for hearing risk factors and those at risk should receive audiological monitoring for delayed-onset, acquired, or progressive hearing loss. Audiological evaluations for children at risk of hearing impairment should be conducted by 24 to 30 months of age, even if they pass initial screening but possess at least one hearing loss risk factor (JCIH, 2000; JCIH, 2007).

Universal Newborn Hearing Screening is in practice in most developed countries and has dramatically improved outcomes for infants with hearing loss at birth. In the United States, children have demonstrated language growth within the normal range of development at five years of age provided that they are enrolled in early intervention within the first year of life (Calderon et al., 1998; Moeller, 2000). Additionally, UNHS programs have decreased the average age of hearing aid fitting in New South Wales from 22 to 3.8 months, improving language and social outcomes where the prevalence was 0.71-1.52/1000 births with congenital hearing loss (Hutt & Rhodes, 2008).

EHDI databases that include UNHS, diagnostic and habilitation outcomes are extremely valuable resources for data sharing for those involved in the management and research of childhood hearing impairments. This source provides vital data for rates, trends, and features of children with permanent hearing loss (Hutt & Rhodes, 2008). For example, in 2000, the JCIH showed that, of those who fail screening in the United States, only half have appropriate follow up services. There are limitations to access and/or follow up, and it is difficult for state-funded EHDI services in the United States to integrate data and communicate this to the EHDI coordinators and the many different stakeholders involved. For a successful EHDI program, pediatricians, family physicians, and other health care professionals, who are funded through a myriad of different sources, must work in partnership with parents and other professionals such as audiologists, physicians, program managers, family support workers, and educators. And, importantly, there must be collaboration among different institutions that assume responsibility for different aspects of the patient's care (screening, evaluation, intervention) (JCIH, 2007).

A recent study by Moeller, Carr, Seaver, and Stredler-Brown (2013) examined the best practices in EHDI programs, specifically family-centered early intervention for children. The goal was to promote widespread, evidence-based recommendations for family-centered early interventions. The consensus advocated early, timely, and equitable access to services. The main focus was on family involvement and partnerships, along with best practice principles for families to make appropriate decisions and means of communication with their child. This included the use of assistive technologies and supporting means of communication (Moeller et al., 2013).

### 1.2.3 Postnatal Hearing Impairment

It is still the case that not all permanent hearing impairments are identifiable at birth (Fortnum, 2003). The goal and mandate set out by groups such as the NIH and the JCIH are to detect all types of hearing loss, even mild, unilateral, or neural hearing impairments that can have significant developmental effects (JCIH, 2007). Little is understood about the numbers and features of infants who develop significant hearing loss after passing initial newborn hearing screening (Hutt & Rhodes, 2008).

A current challenge in the field of audiology is identifying those children who develop hearing problems after passing the initial newborn screening. Postnatal hearing loss includes acquired, late onset, or progressive hearing loss and these categories are hard to distinguish due to the ambiguous causes and time of onset (Hutt & Rhodes, 2008). Little is known about prevalence or how to predict these cases so that more research in this area is required. Fortnum (2003) reviewed different studies and the incidence of late onset, acquired, or progressive hearing loss ranged from 7.5-25.2% of all hearing-impaired children. Reasons for such a range in prevalence rates could include differences in demographics, or methodology, such as different definitions for each hearing loss group (Fortnum, 2003).

Weichbold, Nekahm-Heis, and Welzl-Mueller (2006) conducted a large Austrian retrospective clinical analysis with 105 cases of permanent childhood hearing loss. All of the cases had received a valid newborn hearing screen. Results showed that 23 of these children had hearing within the normal range at birth. After adjusting for under-ascertainment, the researchers found that around 25% of significant permanent childhood hearing impairments diagnosed by nine years of age were postnatal and undetected by

universal hearing screening (Weichbold et al., 2006). This has implications for future research and implies that UNHS programs may not be sufficient to capture all permanent childhood hearing impairments, where postnatal surveillance programs and improved surveillance protocols might.

An important consideration is that not all prenatal aetiologies lead to an impairment that is detectable at birth by screening (Hutt & Rhodes, 2008). Therefore, postnatal hearing loss could be informed by retrospective analysis of any confirmed cases by evaluating screening databases to determine if there is a need for improved screening or diagnostic procedures (Hutt & Rhodes, 2008). Of vital importance to evaluation of early hearing detection and intervention programs (EHDI) is the prevalence of children who not only have congenital hearing loss, but those who develop an impairment identified after newborn screening (Fortnum, 2003). The data from large databases of EHDI programs could be useful for establishing practice and policy for children with different hearing loss outcomes, including progressive and emergent postnatal hearing loss.

#### 1.2.4 Risk Factors and Surveillance

The JCIH lists 11 risk factors that overlap among risk factors for congenital and acquired, late onset, and progressive hearing loss. JCIH (2007) risk factors include the following: (a) caregiver concern; (b) family history of permanent childhood hearing impairment; (c) neonatal intensive care, based on duration and procedures delivered; (d) in utero infections; (e) craniofacial anomalies; (f) physical findings that are associated with a syndrome known to cause permanent childhood hearing impairment; (g) syndromes associated with permanent childhood hearing impairment; (h)

neurodegenerative disorders; (i) culture-positive postnatal infections associated with sensorineural hearing loss; and (j) head trauma.

The JCIH recommends audiological surveillance of all infants with risk factors in localities where there is no UNHS available by following them for the emergence of postnatal hearing loss, and also for those with risk factors but who pass screening in those areas with comprehensive EHDI programs, (JCIH, 2007). Although the JCIH (2000) guidelines advocated audiological monitoring for all infants with risk indicators at six-month intervals, there have been concerns about the feasibility and costs of such an endeavor (JCIH, 2000).

### 1.3 Challenges and Future Directions

There are only a few EHDI programs that currently have the capacity to report the number of infants screened, assessed, and registered in intervention programs, and the JCIH promotes data pooling and assessment of these programs for future policy and practice (JCIH, 2007). There are many challenges and obstacles to assessing and managing EHDI programs such as the following: tracking outcomes of children after failing screening; a lack of specially trained professionals; need for the integration of data through coordinated management and tracking systems; a lack of timely referrals and surveillance programs; and insufficient funding (JCIH, 2007).

There is a need for evidence-based research to determine the design and cost effectiveness of surveillance programs. In addition, EHDI programs have the difficulty of deciding on protocols regarding the risk factor inclusion criteria used to target surveillance. Risk factors, without being too broad or too narrow, should have high predictive power in terms of identifying hearing loss (Hutt & Rhodes, 2008). Moreover,

there is a lack of parental disclosure of certain risk factors and under-utilization of surveillance program follow up services. Finally, the cost of audiological assessment is a policy factor in determining the feasibility of monitoring services.

EHDI data from comprehensive programs can be used to investigate program issues and provide evidence for economic and social justification for surveillance programs (Hutt & Rhodes, 2008). For example, Weichbold et al. (2006) recommend preschool surveillance, as a secondary screen, a program they consider both advantageous and feasible. Ultimately, the goal of EHDI programs is to find a timely way to identify all children with hearing loss and to assess the program components that are currently in place.

A comparison of these hearing outcomes across populations that have different risk levels is needed to improve screening and assessment, for example, infants who pass or fail newborn screening, and those with and without protocol defined risk factors [Appendix C]. The JCIH has identified the critical need for longitudinal studies and population-based studies to determine the prevalence and natural history of these hard-to-detect hearing disorders and evaluation of risk factors contributing to hearing loss (JCIH, 2007). Establishing this foundation in evidence-based research will help in the design of appropriate prevention and intervention strategies.

## 1.4 Review of the Ontario Infant Hearing Program (IHP)

Neonates born in Ontario receive services through the Ontario Infant Hearing Program (IHP), a comprehensive screening, diagnostic, and habilitation program.

Throughout this thesis, the program will be referred to as the IHP, although different IHP

programs outside Ontario may have different protocols. Children from birth to six years of age, who are at-risk for hearing impairment, are evaluated by the Ontario IHP and receive audiological assessments and related services. All audiological services are delivered by IHP trained pediatric audiologists using standardized protocols and clinical equipment (Hyde, 2008). Hearing assessment objectives are to obtain IHP protocol approved estimates of ear and frequency-specific hearing thresholds, in addition to diagnosing the type of any hearing impairment, if present.

The IHP is one of the few programs worldwide that has standardized (risk factor, questionnaire, behavioural, and physiological) screening and diagnostic protocols within a comprehensive surveillance, assessment, and habilitation program for children at increased risk for hearing loss. The H.A. Leeper Speech and Hearing Clinic IHP, which was implemented in 2002, provides comprehensive physiological and behavioural assessment and intervention programs for children “at-risk” for developing auditory impairments. To note, once hearing loss is diagnosed and an individualized plan is defined, families often transfer to another IHP site for follow-up habilitation and other counseling services more conveniently located in their region.

Children are considered “at risk” and referrals to the IHP are accepted: (a) if a child fails their initial newborn hearing screening; (b) if they are identified with a potential communication disorder and referred by an outside party (parent, physician, teacher); (c) if they are either reactivated after passing their initial screening or if they are transferred within the IHP region; or (d) if a child harbours specific hearing-related risk factors (at least one). Also, the IHP provides comprehensive long-term surveillance for

every child considered to have at least one IHP defined risk factor, but who passed the initial universal screening assessment [Appendix E].

The IHP audiological assessment protocol includes age appropriate acoustic immittance with conventional and 1 kHz frequency tympanometry, distortion product otoacoustic emissions (DPOAE), air and bone conduction auditory brainstem response (ABR) using click and frequency-specific tone pips (0.5-4 kHz), visual reinforcement audiometry (VRA), conditioned play audiometry (CPA), and conventional audiometry, while employing the appropriate methodology dictated by the age of the child (Hyde, 2008) by IHP trained audiologists with pediatric expertise [Appendix A]. The audiology assessment follows prescribed equipment and procedures based on the current version of the IHP protocol. These IHP protocols are designed to ensure the validity and reliability of thresholds obtained via physiological and behavioural techniques.

In summary, children from birth to six years of age, who are at-risk for hearing loss according to any of the above criteria, are flagged upon screening through the IHP and followed with assessments and services. As stated earlier, evidence-based research is needed to design appropriate prevention and intervention strategies. This thesis is intended to contribute further to this identified need.

## 1.5 Rationale and Implications of This Thesis

Given the insufficient amount of current evidence, this present retrospective analysis of the comprehensive data set from the Ontario Infant Hearing Program screening and surveillance program at Western University provides unique data for children at-risk for hearing loss. The H.A. Leeper Speech and Hearing Clinic at Western University has extensive experience with infant hearing assessment and surveillance



programs and has a large database of patients. Analysis of these data as a population-based study has the potential to advance knowledge in this important area and to potentially provide a significant contribution to EHDI programs and policies both nationally and internationally.

The main objective of this present retrospective thesis is to evaluate audiological assessment outcomes for all children and infants receiving assessment and surveillance services through one Ontario IHP clinic site located at Western University within the H.A. Leeper Speech and Hearing Clinic.

This data set is of importance due to the large number of children and the unique characteristics of this sample. Few studies have evaluated a clinic sample in which the intake for the clinical population is exclusive to an early hearing detection and intervention program, in this case, the Ontario IHP. Unlike the previous studies of this type, which focus on the outcomes of universal newborn screening (Robinshaw, 1995; Moeller, 2000; Yoshinago-Itano et al., 2000) or a surveillance program for high risk cases (Beswick, Driscoll, Kei, & Glennon, 2012 ), this thesis includes pediatric cases originating from all types of referral sources within this infant hearing program, and compares the outcomes across these different referral groups.

## 1.6 Hypotheses & Research Questions

This study is a descriptive analysis of an Infant Hearing Clinic sample in Ontario, and compares the population characteristics and hearing outcomes across the different intake referral source groups permitted under the Ontario IHP and governed by their protocols. The following five referral group acronyms are used: Newborn hearing screen

fail (NHSF), IHP surveillance group (SURV), external referral (EXT), IHP reactivation (REACT), and IHP transfers (TRANS).

Research Question 1:

*What are the hearing outcomes of infants referred to the Ontario IHP clinic site located at Western University within the H.A. Leeper Speech and Hearing Clinic, a clinic with an intake population exclusive to a comprehensive infant hearing detection and intervention program. Do the hearing outcomes differ, depending on the reason for referral?*

Hypothesis 1:

It is hypothesized that the IHP hearing assessment outcomes, specifically the presence/absence of hearing impairment, will vary as a function of the IHP Referral Source Group (i.e., reason for referral/assessment).

Research Question 2:

*What is the risk factor status, specifically the presence/absence of a hearing risk factor(s), and nature of hearing loss (type, severity, symmetry) in the different Referral Source groups for the University of Western Ontario IHP clinic?*

Hypothesis 2

It is hypothesized that the nature of the hearing loss and risk factor status will vary depending on the reason for referral to the Western University Ontario IHP clinic.

## 1.7 Thesis Format

Chapter 1 provides a brief introduction and a review of the literature and summarizes the rationale and research questions addressed in this thesis. Chapter 2 provides a detailed description of the methods used to address the research aims and objectives in Chapter 1. Chapter 3 presents the results, and Chapter 4 highlights the results and discusses their relevance in the context of the current literature and for potential future policy decisions.

## Chapter 2

### 2 Materials and Methods

#### 2.1 Research Design

This thesis is a descriptive retrospective analysis of data from a cohort of children enrolled in the Ontario IHP and receiving services at the H.A. Leeper Speech and Hearing Clinic site at Western University. In this thesis, the H.A. Leeper Speech and Hearing Clinic will be referred to as the H.A. Leeper Clinic. The study population includes patients referred during the time frame between IHP implementation in February 2002 through August 2011. This thesis assesses the site's population as a descriptive, population-based study to determine the different outcome measures for certain infants, who are at-risk for developing hearing impairment based on specified at-risk groups. This thesis was approved by the Research Ethics Board December 22, 2011 under review #: 17385E.

#### 2.2 Study Population

All subjects were drawn from a patient cohort at risk for having or developing hearing loss, as defined by the IHP, and who were subsequently evaluated at the H.A. Leeper Clinic IHP site. The IHP at the H.A. Leeper Clinic is part of the IHP Southwestern Ontario Regional Centre [Appendix F].

The IHP provides comprehensive physiological and behavioural assessments and intervention programs for children “at-risk” for hearing loss as mandated by the Ontario IHP protocol (Hyde, 2008). Subjects were drawn from the group receiving the following

services: surveillance screening and full diagnostic assessment. Assessment types and timing of initial assessment are described in detail based on the protocol in Appendix E.

This thesis looked at a subset of children receiving IHP services at the H.A. Leeper Clinic, according to the following inclusion criteria: (a) children completing at least one IHP evaluation because they failed their newborn hearing screening (with or without a risk factor); (b) children completing at least one IHP evaluation because they have an IHP designated risk indicator, but passed their newborn hearing screening; (c) children completing at least one IHP evaluation because of a community referral (parent/guardian, teacher, physician concern about communication development) and (d) transfers into the southwestern IHP region.

Candidacy for IHP Audiological Services begins at birth and now includes children up to the age of six years, or entry into Grade 1 (Hyde, 2008). Children initially enrolled for services through the IHP, but who did not complete at least one diagnostic assessment or surveillance screening through the IHP were excluded from this thesis.

Hearing loss can be defined as unilateral or bilateral permanent hearing loss. The IHP definition for permanent childhood hearing loss is: one or more thresholds equal to 30 dB HL or greater (0.5 to 4 kHz), with each ear classified as a conductive, mixed, sensorineural, or auditory neuropathy spectrum disorder type of hearing deficit (Hyde, 2008). Children who were referred to the IHP and who were later either identified with permanent childhood hearing impairment, or normal hearing, were included in this thesis according to the IHP criteria at the H.A. Leeper Clinic site.

## 2.3 Data Collection

### 2.3.1 Electronic IHP Database Review

The H.A. Leeper Clinic IHP electronic database was reviewed for eligible participants. This is a comprehensive site-specific database maintained by the IHP coordinator at Western University. Review of the IHP electronic database in the H.A. Leeper Clinic was completed retrospectively.

#### 2.3.1.1 Electronic Database: Data Extraction and Analyses

Data extracted from the electronic database includes, gender, date of birth, hearing loss status, referral source, and presence of at least one risk factor. The “hearing status” field in the database identified subjects as hearing-impaired or normal hearing, based on IHP criteria. The definition of permanent childhood hearing impairment according to the IHP criteria includes (1) any hearing threshold equivalent to 30 dB HL or greater at any frequency in the range 0.5-4 kHz, in either one or both ears (2) any of the following permanent childhood hearing impairment: conductive impairment associated with structural anomalies of the ear (but not inclusive of non-structural middle ear conditions), mixed, sensorineural, auditory neuropathy spectrum disorder, and retrocochlear disorders, involving the auditory brainstem (3) ages birth-six years (Hyde, 2008, [Appendix B]).

#### 2.3.1.2 Referral Source Groups

Upon exporting data from the IHP database, referral sources were available for nearly all subjects. Referral source describes the route through which children are referred to the H.A. Leeper Clinic IHP site. These data are particularly useful for

examining aspects of the newborn screening and surveillance program, which are discussed later in detail. IHP assessment is available to the following children in a timely matter as mandated by the IHP: neonates and infants who fail IHP UNHS or who fail IHP high risk surveillance; children up to six years with permanent childhood hearing impairment proven by previous IHP assessment or audiometry outside of the IHP; and children up to six years who acquire IHP high risk status extrinsically or through post-natal IHP risk indicator identification (Hyde, 2008). The IHP electronic database documents information about the referral (route and reasons for referral, assessment type) for each child seen for audiological services.

For this thesis, subjects were assigned to five main groups. These five Referral Source groups are defined and will be referred to as:

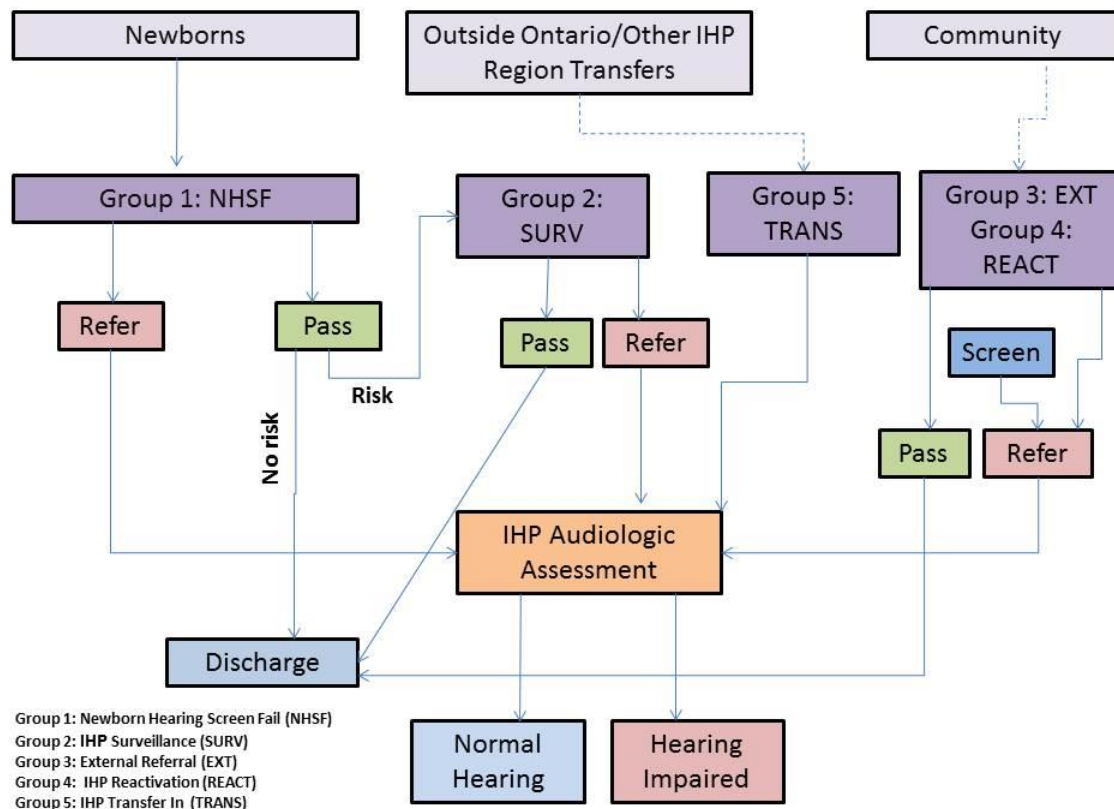
1. Newborn Hearing Screen Fail (NHSF);
2. IHP Surveillance (SURV);
3. External Referral (EXT);
4. IHP Transfer IN (TRANS); and
5. IHP Reactivation (REACT)

NHSF include those babies referred to the IHP with at least one ear failing newborn screening. For the purpose of clarity in this thesis for those readers not in the field of audiology, newborn hearing screen “fail” will be called “fail”, instead of the accepted clinical terminology “refer”, since this terminology may be more familiar to the general healthcare policy audience,. It is clinically more advised to use the term “refer” versus “fail” due to the negative connotations that may be perceived with the latter (Hyde,

2005), but may cause confusion in this thesis because we are studying a variety of different referral routes into the IHP program at the Western University site.

The following summary describes these referral routes and groups included in this thesis. SURV children were referred to the IHP even though they passed newborn screening in both ears, because they had at least one hearing risk factor as per IHP protocol [Appendix C]. EXT includes those children who received a referral from a physician, or from a concerned parent, teacher, or other external source. For some of these cases, hearing screening results may be unknown if the child was missed or did not receive newborn screening (born prior to IHP implementation in 2002). To note, health care referrals were previously accepted into the IHP, whereas all referrals now must be referred from a physician. Cases in the REACT group involve children who originally passed their newborn screening but concerns were raised later and were, therefore, reactivated in the IHP. Finally, children can be transferred into the IHP, either within the IHP region or by another IHP region (Hyde, 2008), and these children were assigned to the TRANS group. Once these categories were assigned, the subjects were then sorted according to their hearing status into normal hearing and hearing-impaired groups. Figure 1 presents an overview of the IHP structure and illustrates the Referral Source groups evaluated in this thesis.





**Figure 1.** An Overview of the IHP Structure and the Referral Categories to the IHP from Newborn Screening.

Note. In this diagram, “refer” means that the child would “fail” the screening test and be referred for a complete IHP Audiologic Assessment.

### 2.3.2 Chart Review

Those identified from the IHP electronic database with a hearing impairment were included in the chart review (n=219). Clinical charts for these subjects were retrieved and data extracted. Following the identification of permanent childhood hearing impairment through the IHP, a child is reassessed every three months during the first year, and at least every six months thereafter, thus providing serial audiograms and/or ABR threshold estimates for the majority of participants.

### 2.3.2.1 Clinical Chart Review: Data Extraction

Data extraction from the clinic charts included the results for newborn hearing screening and high-risk surveillance, risk factor status, community screening outcomes, and audiological assessment measurements. The audiological measurement data included: age in months at time of audiological assessment, hearing test type (Auditory Brainstem Response [ABR], Visual Reinforcement Audiometry [VRA], Conditioned Play Audiometry [CPA], other), hearing loss type (sensorineural, conductive, mixed, auditory neuropathy, none, or unknown) test ear (right or left), pure tone air and bone conduction threshold data (0.5, 1, 2 and 4kHz) [Appendix A]. Tympanometry data were included and immittance data consisted of: date of test, frequency of probe (226 Hz, 1 KHz, other), machine model, middle ear pressure (daPa), peak static admittance (ml), and ear canal volume (ml).

Risk factor data were collected for all subjects in the hearing-impaired group. IHP risk factors are listed in Appendix C (Hyde, 2008). Risk factors were documented in the clinical chart as follows: “Presence of a risk factor?”, where the clinician must indicate “yes”, or “no”. Therefore, it was possible to extract and record whether each subject had at least one IHP defined risk factor present, and if it was indicated “no”, the child was considered to have no known risk factor(s).

## 2.4 Data Analyses

The nature of the hearing loss was studied for each subject defined as hearing-impaired. Only thresholds considered valid and reliable were included in the data analyses. Thresholds were considered valid based on the parameters of the IHP protocol.

Therefore, if there were any questions of reliability, these values were not included based on rules of immittance, ABR, VRA, and CPA guidelines (Hyde, 2008). Threshold values with no response at the limit of the audiometer were entered as the threshold 5 dB greater than the limit of the audiometer so that it could be distinguished between “not tested” and “no response” at the upper limit, thus enabling PTA calculations [Appendix A].

Furthermore, hearing assessment results were excluded from data analyses for any visit where bone conduction thresholds and/or immittance testing indicated temporary middle ear abnormalities, because middle ear status can influence behavioral and physiological outcomes, leading to misinterpretation of the nature and stability of the hearing loss. Threshold immittance values were considered abnormal if they did not meet the IHP criteria for acceptable peak static admittance for the specified age and/or if tympanometric peak pressure was greater or equal to  $\pm 200$  da PA. [Appendix D]. It is not unusual for some pure tone thresholds not to be measured on any given assessment, particularly for young children. Missing thresholds were identified as such. For data analyses, the closest behavioral test date with the most complete threshold information was then used. Subject age was calculated for each visit. Finally, subjects who only received ABR testing were classified as “ABR audiogram only”.

#### 2.4.1 Degree (Severity) of Hearing Loss

Pure tone hearing thresholds were analyzed for each hearing test date, with many subjects having multiple hearing tests. For each test date, air conduction pure tone thresholds were analyzed for each ear separately. Severity of hearing loss was, therefore, classified for each ear separately and only ear-specific results obtained with earphones were used.

The mean threshold for each ear was calculated using both the three and the four frequency pure tone averages: PTA3 (0.5, 1, and 2 kHz) and PTA4 (0.5, 1, 2, and 4 kHz). The results of the pure tone average calculations (PTA3 or PTA4) were categorized as: normal (-10 to 15 dB hearing level [dBHL]), slight (16 to 25 dBHL), moderate (26 to 40 dBHL), moderately severe (56 to 70 dBHL), severe (71 to 90 dBHL), or profound (>90 dBHL) (Clark, 1981). These categories follow Clark's criteria, who used PTA3 only, a definition of pediatric hearing severity recommended by the American Speech-Language-Hearing Association (ASHA) (Clark, 1981).

For the purpose of this thesis, both PTA3 and PTA4, have been included for comparison purposes. Table 2 below demonstrates the commonly accepted classifications of hearing impairment. For defining an individual subject's hearing loss severity, the better ear (BE) threshold was used if hearing loss was bilateral, whereas the worse ear (WE) threshold was used for unilateral hearing loss cases (Clark, 1981). The pure tone average thresholds and hearing loss severity were based on the most recent behavioral audiogram.

**Table 1***Classification of Hearing Impairment*

<b>Hearing Level (dBHL)</b>	<b>Classification</b>
<b>-10 to 15</b>	<b>Normal hearing</b>
<b>16 to 25</b>	<b>Slight hearing loss</b>
<b>26 to 40</b>	<b>Mild hearing loss</b>
<b>41 to 55</b>	<b>Moderate hearing loss</b>
<b>56 to 70</b>	<b>Moderately severe hearing loss</b>
<b>71 to 90</b>	<b>Severe hearing loss</b>
<b>&gt; 90</b>	<b>Profound hearing loss</b>

### 2.4.2 Hearing Loss Type

According to the IHP protocol, the audiologist indicates the type of hearing loss for each hearing test, and includes the identification of permanent conductive hearing components in conductive and mixed losses, as well as auditory neuropathy spectrum disorder in this classification. Specifically, hearing loss type was classified as follows: conductive, sensorineural, mixed, auditory neuropathy, or unknown. As with hearing loss severity, this analysis is determined for each ear, and is based on the most recent behavioral test for which this data (hearing loss type) was available. Cases in which hearing loss type was defined with only ABR testing data available have been grouped as “ABR audiogram only”. Cases that were unclear in regards to hearing loss type were labeled as “unknown”, for the following reasons: no reliable threshold data were

available, incomplete clinical data forms, hearing loss type was queried and/or the test type was contradictory for different test dates.

### 2.4.3 Symmetry of Hearing Loss

Symmetrical hearing means that hearing thresholds are similar in each ear, while asymmetrical refers to a between ear difference in these thresholds. In this thesis, the symmetry of pure tone hearing thresholds was analyzed, and was based on the most recent behavioral audiometric test. Hearing loss was defined as unilateral when the hearing thresholds in one ear were within normal limits, defined as a mean three frequency pure tone average,  $PTA_{3^{0.5-2\text{kHz}}}$ , of 20 dB HL or less, with the other (worse) ear below this limit (Pittman & Stelmachowicz, 2003). Bilateral asymmetric hearing loss was present when both ears were below this normal range, and the ears differed by 15 dB or more ( $>20$  dB) at one or more frequencies (thresholds for all 4 frequencies and both PTAs were analyzed) (Pittman & Stelmachowicz, 2003).

When a subject had only ABR audiometric evaluations, this is noted and these were excluded. For the purposes of this thesis, the evaluation of symmetry was based on the most frequently measured pure tone thresholds (IHP protocol dictates measurement and reporting of these frequencies) in our population: 0.5, 1, 2, and 4 kHz. Cases with only unilateral testing and ABR thresholds only were excluded from the analysis.

## Chapter 3

### 3 Results

#### 3.1 Data Retrieved from the Electronic Database

##### 3.1.1 Overview

The subjects of this thesis consist of 2,638 children who were enrolled at the H.A. Leeper Clinic IHP site between 2002 and August 2011. These subjects were referred to the IHP through several different routes, and are classified according to the outcome of their hearing assessment or surveillance testing as either normal hearing, or hearing impaired. Of this total, 2,390 children were determined to have normal hearing. Gender was recorded for the normal hearing population, with 31.9% (n=763) males, 25.4% (n=608) females, and 42.6%, (n=1,019) of unknown gender. Unknown cases were those for which gender data were missing for the “gender” field in the electronic database. Conversely, 248 cases were identified with hearing loss. More than half of the children identified by the IHP database with hearing loss (n=248) were male (62.9%; n=156) and 37.1% (n=92) were female. Table 2 depicts the distribution of gender for both hearing-impaired and normal hearing cases.

**Table 2***Study Population Based on the Electronic Database*

<b>Study Population</b>	<b>Hearing-impaired cases</b>	<b>Normal hearing cases</b>	<b>Total</b>
<b>Total</b>	<b>248</b>	<b>2,390</b>	<b>2,638</b>
<b>Males</b>	<b>156 (62.9%)</b>	<b>763 (31.9%)</b>	<b>919 (34.8%)</b>
<b>Females</b>	<b>92 (37.1%)</b>	<b>608 (25.4%)</b>	<b>700 (26.5%)</b>
<b>Unknown</b>	<b>0 (0.0%)</b>	<b>1,019 (42.6%)</b>	<b>1,019 (38.6%)</b>

*Note.* Percentage totals may not add to 100 due to rounding.

### 3.1.2 Referral Routes

Based on data retrieved from the electronic database, 2,638 children were referred to the IHP through a variety of different routes and subsequently enrolled in the IHP at the H.A. Leeper Clinic during the study period 2002-2011. The study sample was broken down into two groups based on the referral route and also on the outcome of their audiological assessment: Normal Hearing (n=2,390) and Hearing Impaired (n=248). The definition of hearing outcome was based on the IHP protocol definition of normal hearing [Appendix B].

There were five different referral routes to the IHP, and the sample was divided into five groups: Newborn Hearing Screen Fail (NHSF), IHP Surveillance (SURV), IHP Reactivation (REACT), IHP Transfer In (TRANS), and External Referrals (EXT). Table 3 is a summary of the study sample, showing both the referral route into the IHP, and the hearing outcome for each Referral group.



**Table 3***An Overview of the Electronic Database by Referral Group*

<b>Referral Source groups</b>	<b>Hearing-impaired group total</b>	<b>Normal hearing group</b>	<b>Total</b>
<b>NHSF</b>	<b>143</b>	<b>550</b>	<b>693</b>
<b>SURV</b>	<b>23</b>	<b>1,536</b>	<b>1,559</b>
<b>REACT</b>	<b>31</b>	<b>101</b>	<b>132</b>
<b>TRANS</b>	<b>22</b>	<b>56</b>	<b>78</b>
<b>EXT</b>	<b>28</b>	<b>132</b>	<b>160</b>
<b>Missing Data*</b>	<b>1</b>	<b>15</b>	<b>16</b>
<b>Grand Total</b>	<b>248</b>	<b>2,390</b>	<b>2,638</b>

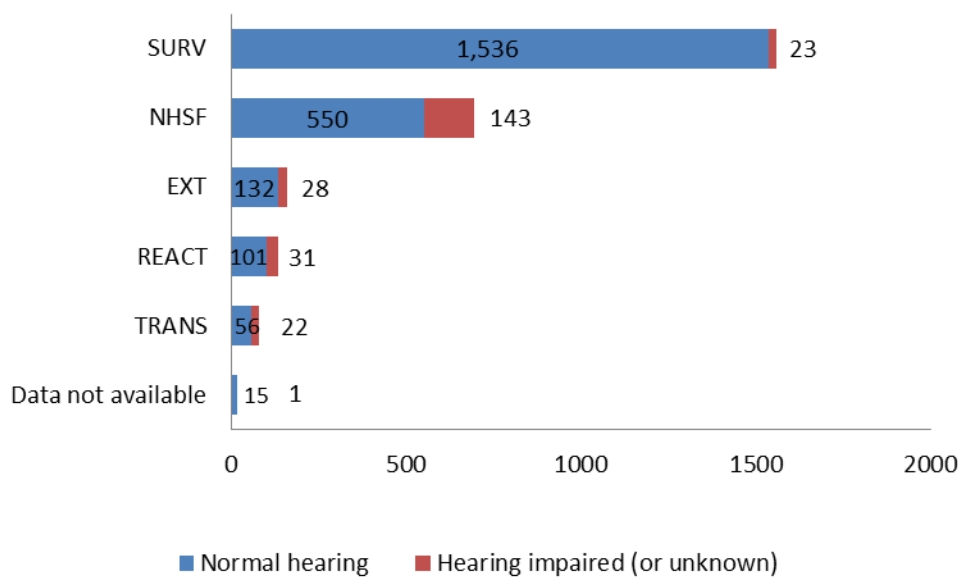
\*Note. Data not entered in the electronic database. Note that percentages may not add up to 100% due to rounding.

The majority of the sample was referred to the IHP for audiological surveillance, with 59.1% (n=1,559/2,638) of the sample in the SURV group. The NHSF group, assessed because they failed their newborn hearing screening, has the second largest number of children referred, with 26.3% (n=693/2,638). Those who entered the IHP by other routes, the EXT, REACT, and TRANS groups constitute the remainder of the study sample: EXT have 6.1% of the total (n=160/2,638), REACT at 5.0% (n=132/2,638), and TRANS with 3.0% (n=78/2,638). Data for referral route and audiological assessment were essentially complete; only 0.6% of the study sample had missing data in the “Referral group” field in the electronic database.

Figure 2 depicts the number of cases identified with hearing loss by the total number referred for each group. The hearing outcome for SURV, the largest Referral group, enrolled in the IHP for audiological surveillance, revealed a low detection rate, with only 1.5% of cases identified with permanent childhood hearing loss (SURV: n=23/1,559). One of the main objectives of this research is to examine the

effectiveness of the IHP Surveillance Program. The data in Table 3, particularly the 23 SURV cases as a fraction of the total (1,559), if extrapolated to a provincial total, could indicate the number of potential beneficiaries of surveillance. For the NHSF Referral group that failed neonatal hearing screening, the audiological testing outcome more frequently revealed a hearing impairment, with 20.6% ( $n=143/693$ ) identified with a permanent childhood hearing impairment. For the EXT and REACT Referral groups, the detection rate for hearing loss was also high at 17.5% (28/160) and 23.5% (31/132) respectively, suggesting that the policy of accepting referral from the community is an appropriate means of detecting cases missed by neonatal screening, or those with emerging postnatal hearing losses.

In summary, these data illustrate that the majority of children referred to the IHP at the H.A. Leeper Clinic site had a normal hearing outcome on audiological assessment (Figure 2). Although the SURV group received the highest number of referrals, the vast majority in this Referral group, were normal hearing, despite having at least one risk factor; 98.5% were discharged from audiological surveillance with a normal hearing outcome. For the second largest Referral group, NHSF, 79.4% ( $n=550/693$ ) did not have hearing loss, based on the outcome of their complete audiological assessment, despite failing the neonatal hearing screening.



**Figure 2.** An Overview of Referral Source Category Data.

### 3.1.3 Risk Factor Data

Hearing risk factor presence/absence data were extracted using this data field in the electronic database and results were analyzed. For those subjects with data available in the electronic database, 46.2% of those children with normal hearing were identified with a risk factor ( $n=1,103/2,390$ ), whereas only 5.2% did not have a known risk factor ( $n=124/2,390$ ). In the hearing-impaired group, 66.1% were identified with at least one risk factor ( $n=164/248$ ), and 27.8% ( $69/248$ ) did not have a known risk factor.

However, data from the risk factor field of the electronic database were missing for a significant proportion of the sample. The normal hearing group was missing 48.7% ( $n=1,163/2,390$ ) of the risk factor data and the hearing-impaired group was missing less with 6.0% ( $n=15/248$ ). In many cases, this data field may have been empty because the hearing risk factor status was inferred from other data fields. For the SURV group ( $n=599$ ), it is required that, based on the IHP protocol, all of these cases have at least one

risk factor present. Furthermore, the 24 cases that were defined as not having a risk factor in the electronic database are presumed to be data entry errors.

## 3.2 Data Retrieved from the Chart Review

### 3.2.1 Overview

Clinical chart review was performed on all cases classified as hearing-impaired in the electronic database (n=248). These charts were reviewed for data pertaining to hearing loss features, including threshold data, and thus provided confirmation of the IHP designation of permanent childhood hearing impairment. Twenty-nine charts were not available, were considered missing data, and were excluded from further analyses, leaving a total sample of 219 hearing-impaired cases for further detailed analyses.

### 3.2.2 Referral Routes

Table 4 data describes the referral route for the cohort with hearing impairment, based on chart review. The NHSF group comprised the majority of the study sample for the hearing-impaired group as hypothesized with 58.4% (n=128). These children failed screening and were subsequently referred to the IHP, and then identified with some type of hearing impairment.

The remaining cases were equally divided among the last three Referral route groups: EXT comprised 12.3% (n=27) of this group, while REACT and TRANS encompassed 11.4% (n=25) and 8.7% (n=19), respectively. Finally, the SURV group, an important group to examine, constituted 9.1% of those identified in the hearing-impaired group based on charts available for review (n=20).

**Table 4***An Overview of the Referral Source Category Data*

<b>Referral Source groups</b>	<b>Hearing impaired-group analyzed</b>	<b>Percentage by Referral group (%)</b>
<b>NHSF</b>	<b>128</b>	<b>58.4</b>
<b>SURV</b>	<b>20</b>	<b>9.1</b>
<b>REACT</b>	<b>25</b>	<b>11.4</b>
<b>TRANS</b>	<b>19</b>	<b>8.4</b>
<b>EXT</b>	<b>27</b>	<b>12.3</b>
<b>Data not available*</b>	<b>0</b>	<b>0</b>
<b>Grand Total</b>	<b>219</b>	<b>100</b>

\*Note. Data not available as the data entry field was blank in the electronic database.

The NHSF group contained the most cases of hearing impairment (n=128) and is, therefore, the group that appears to benefit most from newborn screening programs, as documented in the literature and by the JCIH (JCIH, 2007). EXT, REACT and TRANS children also contributed to the total hearing-impaired sample with close to 32.4% of hearing-impaired cases.

### 3.2.3 Risk Factor Data

Infants who pass hearing screening and have no hearing risk factors according to IHP protocols are not enrolled in the IHP. However, infants who pass their newborn hearing screening but have one of the IHP risk factors are considered “at-risk” and are entered into the audiological Surveillance Program. By nature of their group status, all those in SURV have at least one risk factor. Documentation of risk factor information was performed using the clinical charts to extract new data, and cross-check information derived from the electronic database.

It is important to note that the electronic database and the clinic IHP data forms only capture the presence or absence of a risk factor. Although risk factors were often listed in the chart, the IHP only requires the presence of at least one risk factor, limiting us from examining a comprehensive list of risk factors. Overall, 69.9% (153/219) of the hearing-impaired group had at least one risk factor, whereas 28.3% (62/219) did not have a risk factor. For a small number, the risk factor status was unknown (1.8%; n=4/219). These were cases where the data were not recorded in the chart (n=3) or the actual risk data were unknown because the child was adopted (n=1).

Table 5 shows risk factor data by Referral group for the hearing-impaired group from chart review (n=219). All hearing-impaired children in the SURV group had at least one risk factor identified, as expected, since this is the criterion for enrollment in the IHP Surveillance Program. For those hearing-impaired children in the NHSF group, referred because they failed their newborn hearing screening, the majority (74.2%; n=95/128) also harboured at least one IHP hearing risk factor. The EXT group had just over half (55.6%; n=15/27) identified with at least one risk factor while the TRANS hearing-impaired cohort had (57.9%; n=11/19). Lastly, the REACT group had 48% (n=12/25) cases with at least one risk factor.

**Table 5**

*Referral Source Data and Risk Factor Presence in the Hearing-Impaired Group Referred to the IHP (n=219)*

<b><u>Referral groups (n=219)</u></b>	<b>Risk factor</b>		
	<b>Risk factor present</b>	<b>No known risk factor</b>	<b>Risk factor unknown</b>
<b>NHSF n=128 (58.4%)</b>	<b>95 (74.2%)</b>	<b>33 (25.8%)</b>	<b>0 (0.0%)</b>
<b>SURV n=20 (9.1%)</b>	<b>20 (100%)</b>	<b>0 (0.0%)</b>	<b>0 (0.0%)</b>
<b>REACT n=25 (11.4%)</b>	<b>12 (48%)</b>	<b>12 (48%)</b>	<b>1 (4.0%)</b>
<b>TRANS n=19 (8.7%)</b>	<b>11 (57.9%)</b>	<b>6 (31.6%)</b>	<b>2 (10.5%)</b>
<b>EXT n=27(12.3%)</b>	<b>15 (55.6%)</b>	<b>11 (40.7%)</b>	<b>1 (3.7%)</b>
<b>TOTAL N=219</b>	<b>153 (69.9%)</b>	<b>62 (28.3%)</b>	<b>4 (1.8%)</b>

*Note.* Total n=248. n=29 charts were not available, therefore, n=219 cases were analyzed.

### 3.2.4 Hearing Loss Type

There are three basic types of hearing loss described in the literature: sensorineural, conductive, and mixed. Auditory neuropathy is another type of hearing loss targeted by the IHP, and is categorized as such. For those cases where hearing loss type is not confirmed by the end of this thesis, they are labelled as “unknown”. Unknown cases were examined in detail, as they may be unknown for different reasons.

Tables 6 and 7 and Figures 3-6 summarize bilateral and unilateral hearing loss with a breakdown of hearing loss type in each category, and further subdivided by Referral group. Table 6 (Figures 3 and 4) provides data for all cases with a behavioural

audiogram; the most recent behavioral audiogram with available hearing loss type data was used. Table 7 (Figures 5 and 6) provides data for those with ABR results only.

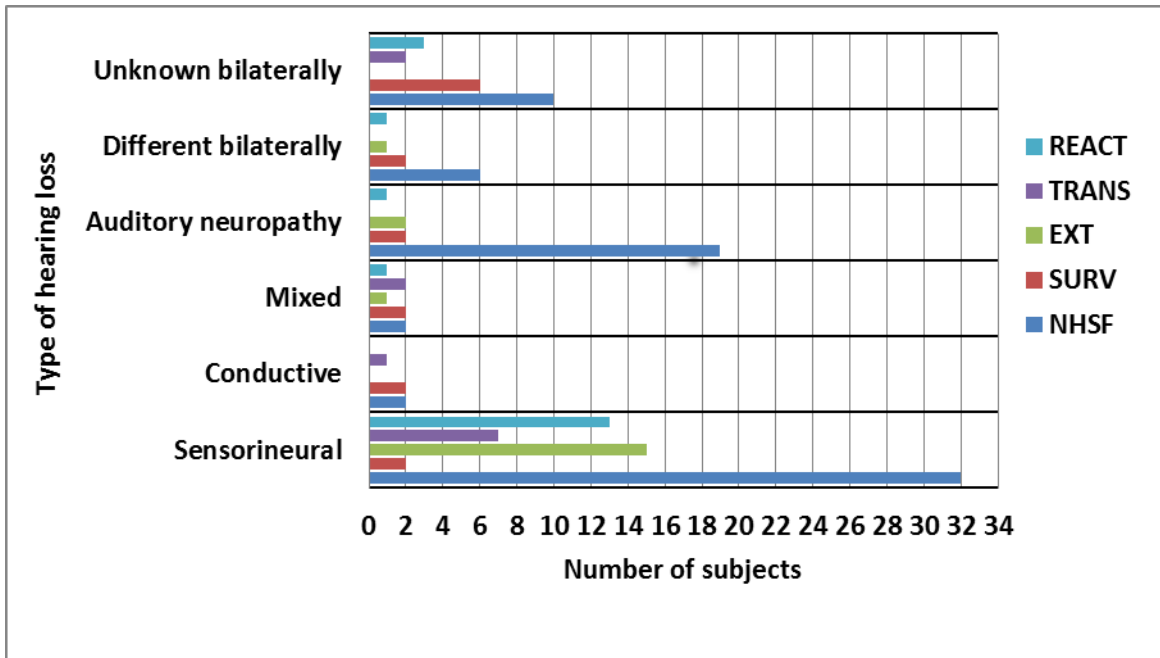
**Table 6**

*Hearing Loss Type by Referral Source – Behavioural Audiogram*

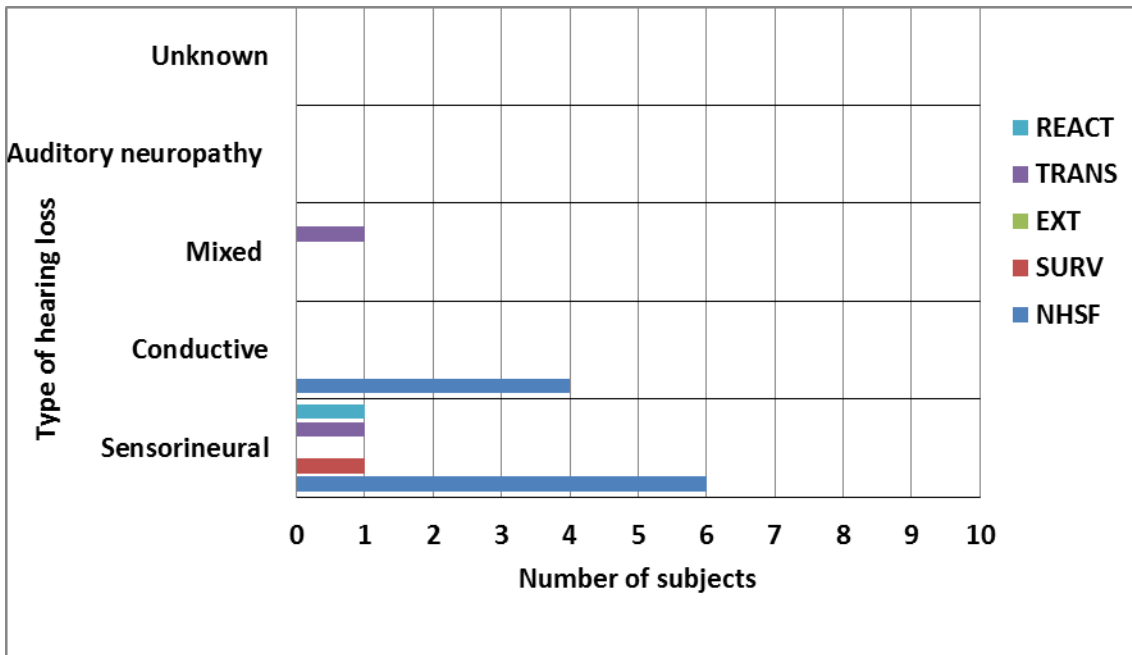
Hearing loss type	NHSF (n=81)	SURV (n=17)	EXT (n= 19)	TRANS (n=14)	REACT (n=20)	Total (n=151)
<b><u>Bilateral</u></b>						
Sensorineural	32 (45.1%)	2 (12.5%)	15 (78.9%)	7 (58.3%)	13 (68.4%)	69 (50.4%)
Conductive	2 (2.8%)	2 (12.5%)	0	1 (8.3%)	0	5 (3.6%)
Mixed	2 (2.8%)	2 (12.5%)	1 (5.3%)	2 (16.7%)	1 (5.3%)	8 (5.8%)
Auditory neuropathy	19 (26.8%)	2 (12.5%)	2 (10.5%)	0	1 (5.3%)	24 (17.5%)
Different bilaterally	6 (8.5%)	2 (12.5%)	1 (5.3%)	0	1 (5.3%)	10 (7.3%)
Unknown bilaterally*	10 (14.1%)	6 (37.5%)	0	2 (16.7%)	3 (15.8%)	21 (15.3%)
<b>Total Bilateral</b>	<b>71</b>	<b>16</b>	<b>19</b>	<b>12</b>	<b>19</b>	<b>137</b>
<b><u>Unilateral</u></b>						
Sensorineural	6 (60%)	1 (100%)	0	1 (50%)	1 (100%)	9 (64.3%)
Conductive	4 (40%)	0	0	0	0	4 (28.6%)
Mixed	0	0	0	1 (50%)	0	1 (7.1%)
Auditory neuropathy	0	0	0	0	0	0
Unknown*	0	0	0	0	0	0
<b>Total Unilateral</b>	<b>10</b>	<b>1</b>	<b>0</b>	<b>2</b>	<b>1</b>	<b>14</b>
<b>Grand total</b>	<b>81</b>	<b>17</b>	<b>19</b>	<b>14</b>	<b>20</b>	<b>151</b>

\*Note. Note that unknown cases may refer to those cases that are not yet determined, have conflicting results, or no data confirming hearing loss type, such as in cases with only one visit.





**Figure 3.** Bilateral Hearing Loss Type: Behavioral Audiogram.



**Figure 4.** Unilateral Hearing Loss: Behavioral Audiogram.

For those cases illustrated in Table 6 (Figures 3 and 4) with behavioural audiometric data, the bilateral hearing loss category includes all cases analyzed with hearing loss in both ears, based on the IHP data forms. For the group with unilateral loss, these were cases where the IHP data form indicated hearing loss in only one ear at the most recent behavioural test. Interestingly, auditory neuropathy cases bilaterally comprise 17.5% of the total bilaterally (24/137).

Conductive (3.6%; n=5/137) and mixed (5.8%; n=8/137) hearing loss had relatively few cases bilaterally, while there were 7.3% of subjects with different hearing loss (type) between ears bilaterally (n=10/137). This includes cases where hearing loss type in one ear is known and is unknown in the other, or, less frequently, when there is a different type of loss by ear. In the bilateral group, 15.3% (n=21/137) had unknown hearing loss (type), at the end of the study period. These unknown cases occur when behavioural testing was incomplete, responses were unreliable over the test period, or the data field was blank on the IHP form, and, therefore, hearing loss type could not be deduced from the available threshold data.

It is clear that the majority of hearing loss cases are from the NHSF group, and are sensorineural, with 51.8% (n=71/137) bilateral cases and 71.4% (n=10/14) unilateral cases. To note, all of the 8.5% (n=6/71) “bilateral different” cases were sensorineural in one ear, and unknown in the other ear. The population with auditory neuropathy bilaterally consisted of 26.8% (n=19/71) children in this group, which is relatively more of this type of hearing loss than any of the other Referral group.

There were few conductive (2.8%; n=2/71) and mixed (2.8%; n=2/71) hearing loss cases bilaterally in the NHSF group. For the “bilateral unknown” cases (14.1%; n=10/71), reasons for the unknown status included: no reliable results, or only one visit where hearing loss type was blank. The NHSF group also had the majority of unilateral cases from the behavioural audiogram data in Table 6 (Figures 3 and 4) with 6/10 sensorineural cases and 4/10 conductive cases.

For the SURV group in Table 6 (Figures 3 and 4), there is an even distribution bilaterally across sensorineural, conductive, mixed, auditory neuropathy, and “different bilaterally”, all with 12.5% (n=2/16). Also, there was one case of sensorineural unilateral loss. The 12.5% (n=2/16) “different bilateral” cases in the SURV group were both sensorineural in one ear and unknown in the other. Of interest are the 37.5% (n=6/16) “unknown bilateral” cases. One of these cases is a child with potential auditory neuropathy, however, there is contradictory data, and no definitive hearing loss type is confirmed at the time of this analysis. The other five cases were “unknown bilaterally” because there were no available thresholds (one visit only), no reliable results, or undetermined status still at the time of analysis.

This led to caution in reviewing available charts for the 20 hearing-impaired cases under SURV, as a more conservative estimate would be 13 or 14 cases (including ABR audiogram data) with confirmed hearing loss type. It is important to note that the hearing-impaired category also includes potentially unknown cases (still not yet determined for hearing loss type).

An assessment of the EXT group in Table 6 (Figures 3 and 4) shows that the majority of bilateral cases in this group are sensorineural (78.9%; n=15/19) by behavioural audiogram. Out of the total of 19 cases, there is one case (5.3%) and two cases (10.5%) with auditory neuropathy. There were no conductive or unknown cases bilaterally. The one “bilateral different” case was one ear with mixed loss, and the other ear was unknown. Unilaterally, there were no identified cases in the EXT group by behavioural audiogram.

The data for the TRANS and REACT groups are similar to the other Referral groups in that the majority of bilateral cases are sensorineural. There were 16.7% (n=2/12) “unknown bilateral” cases in the TRANS group, and 15.8% (n=3/19) in the REACT group. The two cases in the TRANS group appear to have some type of hearing impairment but hearing loss type information is blank for both patients. In the REACT group, the three cases all appear to have hearing loss but the hearing loss type is blank on the data forms, where there is either only one visit, or the patient could not be tested. The TRANS group had two cases of unilateral loss and the REACT group had one case with unilateral loss.

Table 7, and corresponding Figures 5 and 6, all show the data for cases where there was only ABR audiogram data available. Again, the majority of cases are sensorineural (32.4%; n=22/68), and “different bilateral” (29.4%; n=20/68), or “unknown bilaterally” (16.2%; n=11/68). These cases, with ABR threshold data only, should be considered preliminary and hearing loss type remains to be confirmed for these children. Again, the NHSF group comprises the majority of those subjects with ABR only (69.1%; n=47/68).

**Table 7***Hearing Loss Type by Referral Groups- ABR audiogram only*

Hearing loss type	NHFS (n=47)	SURV (n=3)	EXT (n=8)	TRANS (n=5)	REACT (n=5)	Total (n=68)
<b><u>Bilateral</u></b>						
Sensorineural	14 (32.6%)	2 (66.7%)	3 (42.9%)	2 (40%)	1 (20%)	22 (34.9%)
Conductive	1 (2.3%)	0	1 (14.3%)	0	0	2 (3.1%)
Mixed	2 (4.7%)	0	1 (14.3%)	0	1 (20%)	4 (6.3%)
Auditory neuropathy	3 (7.0%)	0	0	0	1 (.20)	4 (6.3%)
Different bilaterally	17 (39.5%)	0	1 (14.3%)	2 (40%)	0	20 (31.7%)
Unknown bilaterally*	6 (14.0%)	1 (33.3%)	1 (14.3%)	1 (20%)	2 (40%)	11 (17.5%)
<b>Total Bilateral</b>	<b>43</b>	<b>3</b>	<b>7</b>	<b>5</b>	<b>5</b>	<b>63</b>
<b><u>Unilateral</u></b>						
Sensorineural	3 (75%)	0	0	0	0	3 (60%)
Conductive	0	0	1 (100%)	0	0	1 (20%)
Mixed	0	0	0	0	0	0
Auditory neuropathy	0	0	0	0	0	0
Unknown*	1 (25%)	0	0	0	0	1 (20%)
<b>Total Unilateral</b>	<b>4</b>	<b>0</b>	<b>1</b>	<b>0</b>	<b>0</b>	<b>5</b>
<b>Grand total</b>	<b>47</b>	<b>3</b>	<b>8</b>	<b>5</b>	<b>5</b>	<b>68</b>

*\*Note.* Note that unknown cases may refer to those cases that are not yet determined, have conflicting results, or no data confirming hearing loss type, such as in cases with only one visit.

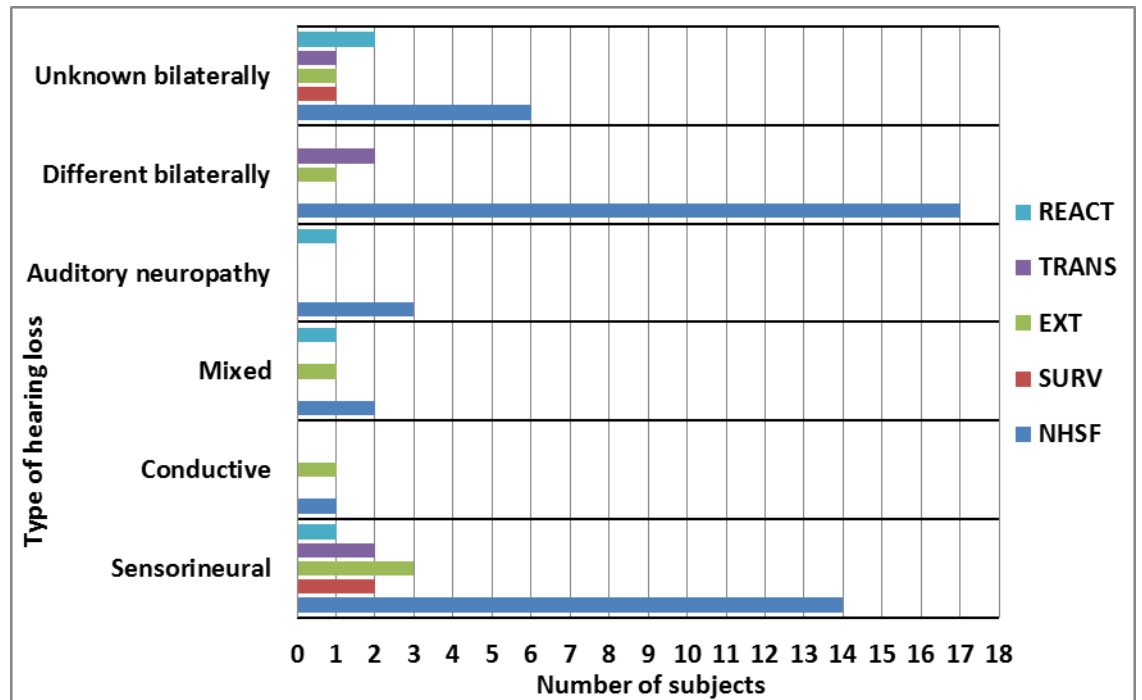


Figure 5. Bilateral Hearing Loss Type: ABR Audiogram Only.

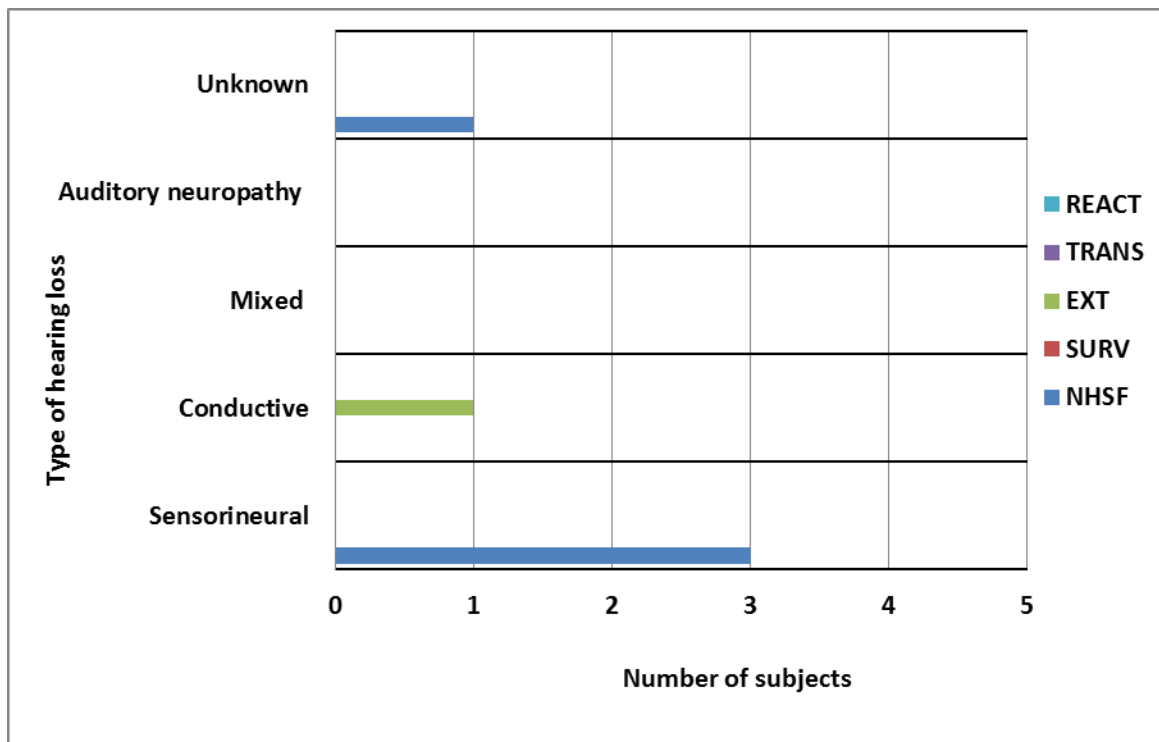


Figure 6. Unilateral Hearing Loss Type: ABR Audiogram Only.

### 3.2.5 Hearing Loss Severity

Table 8 shows the degree of hearing impairment using a common pediatric classification system in which the categories range from normal hearing to profound hearing loss (Clark, 1981). Results are shown using PTA3 according to Clark (1981) and PTA4 for comparison, since this is often used in published studies. Note that “ABR only” audiogram data were not included. The two PTAs provide similar results; the most significant hearing losses (all from moderate to profound categories) account for approximately two-thirds of the hearing-impaired sample. Also, there is one normal hearing case looking at PTA3, however, according to PTA4, the child is classified with slight hearing loss.

**Table 8**

*Degree of Hearing Impairment*

Classification	Hearing level (dBHL)	Number of subjects using PTA 3 (n=93) (%)		Number of subjects using PTA 4 (n=79) (%)	
Normal hearing	-10 to 15	1	1.1	0	0
Slight hearing loss	16 to 25	10	10.8	10	12.7
Mild hearing loss	26 to 40	20	21.5	15	19.0
Moderate hearing loss	41 to 55	23	24.7	22	27.9
Moderately severe hearing loss	56 to 70	19	20.4	16	20.3
Severe hearing loss	71 to 90	12	12.9	12	15.2
Profound hearing loss	> 90	8	8.6	4	5.1

\*Note. Categories are based on the most recent behavioral audiogram pure tone averages, using the better ear if hearing loss is bilateral, and the worse ear if unilateral.

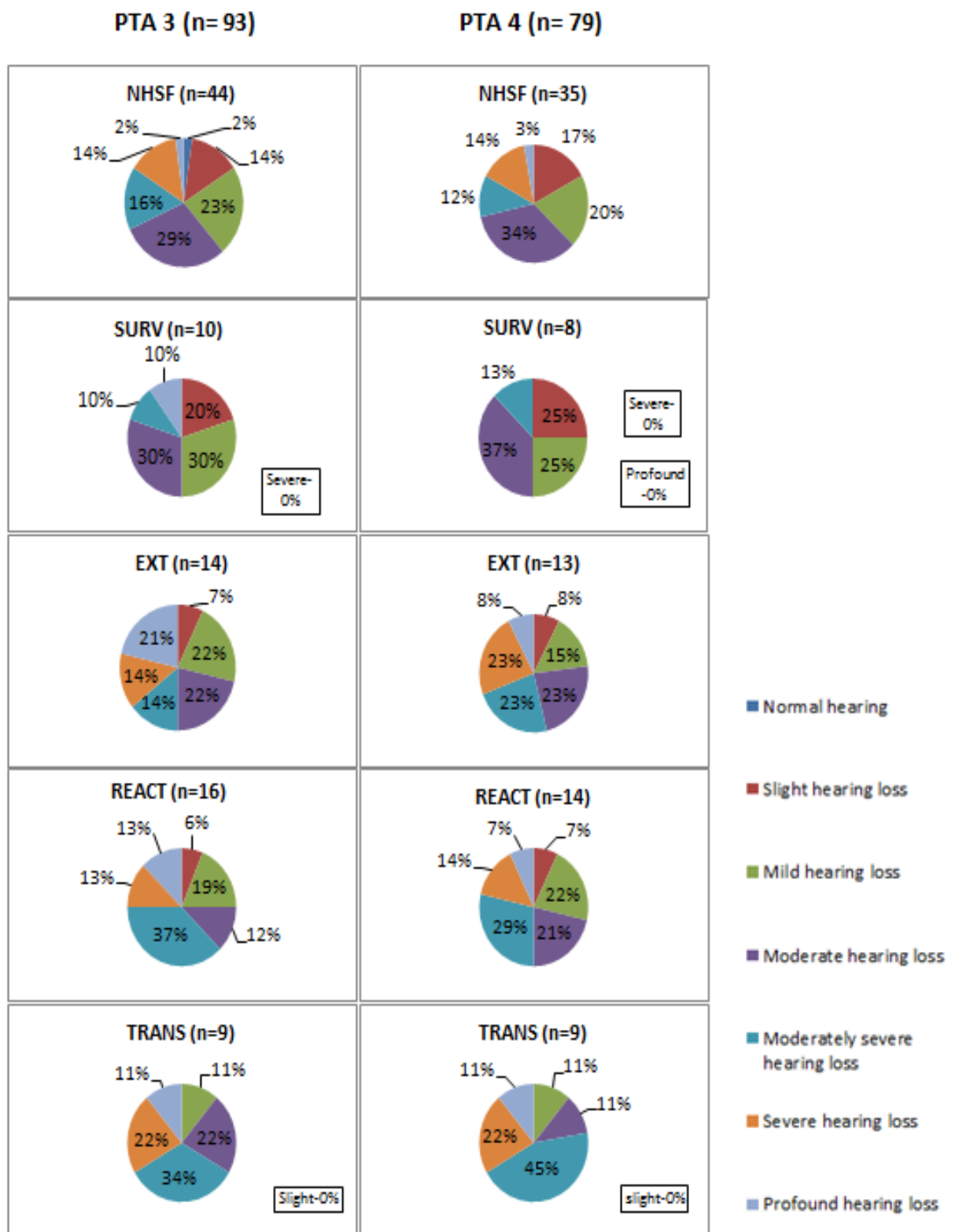
Figure 7 shows the degree of hearing impairment by Referral groups for both PTA3 and PTA4. As noted above, approximately two-thirds of cases fall in the categories of moderate hearing loss or worse. By Referral group, EXT and TRANS have the highest proportion of cases in the moderate-to profound range of hearing loss, while the frequency of this degree of hearing loss in children under audiological surveillance is much lower (SURV: PTA3 at 20%; n= 2/10). It is clear from Figure 7 (and Appendix I) that the NHSF group has the majority of hearing-impaired cases. Within the NHSF group, and based on the PTA3 data (n=44/93), both moderate (29%) and mild (23%) hearing loss are predominant, followed by moderately severe (16%), slight (14%), and profound (14%) hearing loss. Therefore, the degree of hearing loss was distributed evenly in the NHSF group, and likewise for the TRANS and EXT Referral groups. Interestingly, there was one child listed with normal hearing looking at PTA3 data (slight hearing loss when looking at PTA4). This case is based on the most recent visit with PTA data, but the child did return for a subsequent visit, and it appears there is a slight hearing loss but results were not reliable, and no PTA data were available for that visit.

The SURV group is the Referral group with the most cases of mild hearing loss, as well as moderate hearing loss, both at 30% with PTA3 data; results are similar for PTA4. This could indicate that these mild hearing loss cases are not being picked up at newborn screen but would be captured through subsequent surveillance. It is also possible that they were normal hearing at birth and that the hearing loss is emergent, and had progressed to a mild loss at the time of assessment. The results for SURV cases with a slight hearing loss, (10% with PTA3 data; 8% with PTA4) also support this possibility.



Moderately-severe and profound hearing loss degree categories are both 10% of this group of ten children using PTA3 data.

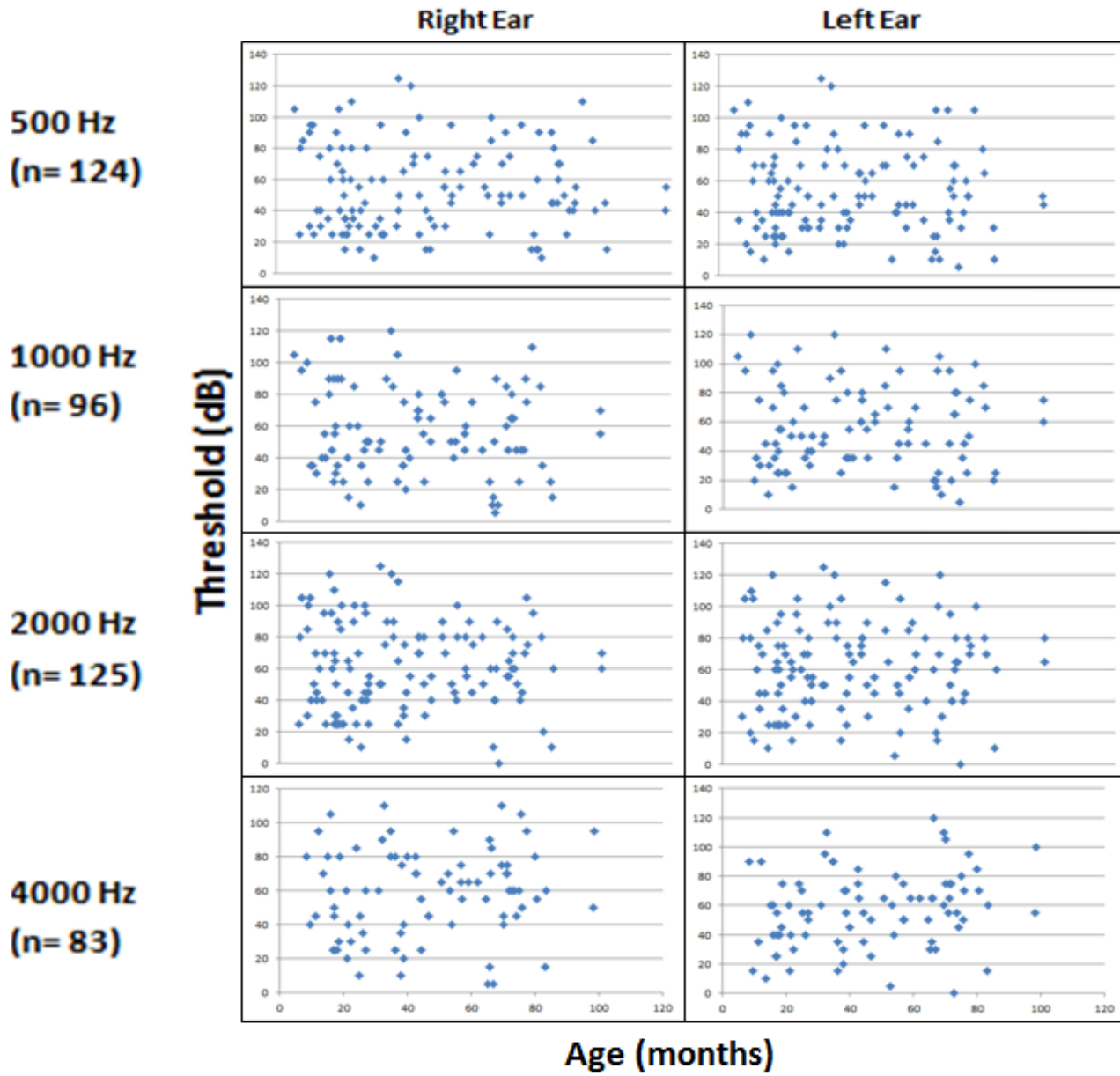
The TRANS group has the majority of their cases looking at PTA3 data with moderately severe hearing loss (34%), followed by moderate and severe hearing loss, both 22%. Profound and mild hearing loss accounted for 11% each. The data with PTA4 show a larger proportion with moderately severe hearing loss (45%) compared to PTA3.



**Figure 7.** Degree of Hearing Loss by Referral Groups.

### 3.2.6 Age

The age (months) of the most recent, complete behavioural audiogram is shown in Figure 7, for the following test frequencies: 0.5 KHz, 1 KHz, 2 KHz, 4 KHz. The sample size varies by frequency because not all cases had all frequencies tested. Cases with only ABR data are not included. The scatterplots show that all degrees of hearing loss are identified, irrespective of age at the final (most recent) behavioural audiogram.



**Figure 8.** Threshold Data by Age for Most Recent Behavioral Audiogram.

### 3.2.7 Symmetry

Ear symmetry is based on comparisons of the left ear versus right ear thresholds, as shown in Tables 9 and 10, and in Figure 9. Scatterplots were analyzed, and correlation coefficients are shown in Figure 9. The scatterplots indicate that most individuals have symmetrical hearing loss, although more variation and a lower correlation coefficient is shown for the 4 KHz threshold data.

Tables 9 and 10 provide insights into how hearing level asymmetry varies by type of test and by Referral group. From Table 9, it is clear that the 2 KHz and 4 KHz test results show a greater proportion of asymmetry cases. The other test types and PTAs are similar in the extent to which they provide lower proportions of asymmetry cases. The numbers in brackets in Table 10 show the number of cases in each category although not all cases have all four frequencies tested. However, 19 cases have 14 such cases (74%), with the second group having 17/25 or 68%. The lowest proportion is for the SURV group, where the 20 cases record only three asymmetries (15%).

**Table 9**

*Cases with Asymmetry at Different Frequencies and PTA3 and PTA4*

Frequency	Cases with asymmetry	Total (n)	% with asymmetry
0.5 KHz	16	124	12.9%
1 KHz	13	96	13.5%
2 KHz	24	125	19.2%
4 KHz	18	83	21.7%
PTA 3	12	93	12.9%
PTA 4	11	79	13.9%

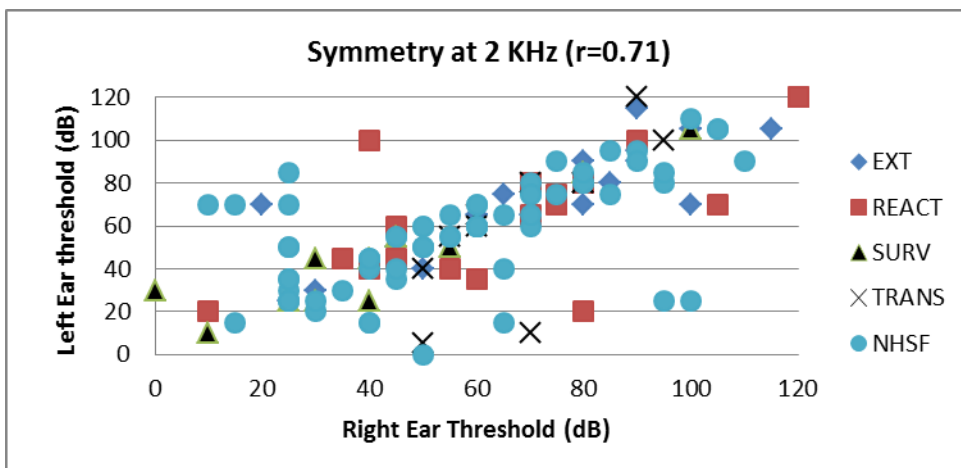
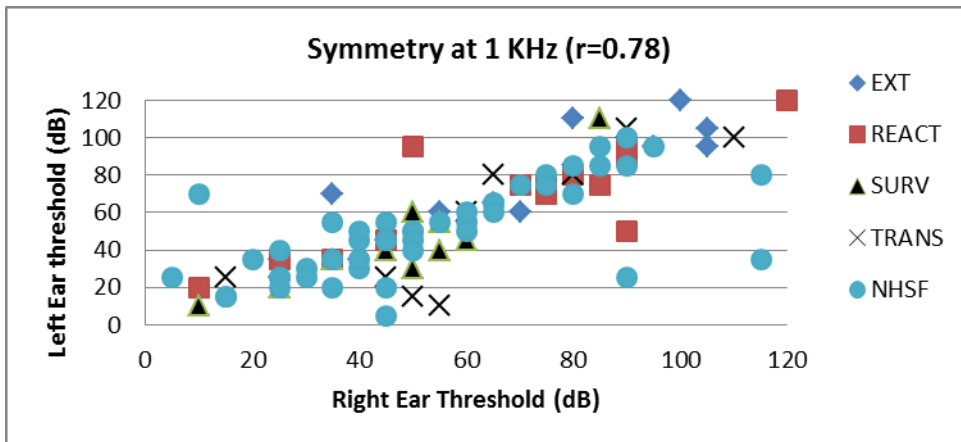
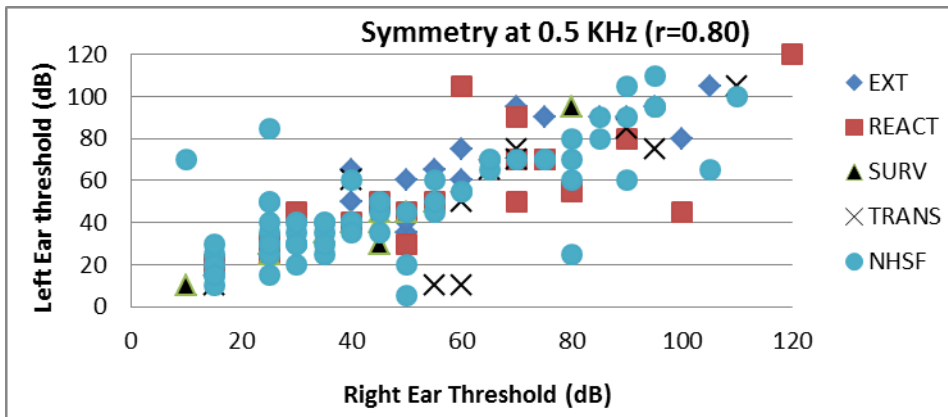
*Note.* Asymmetry is defined as a difference of > 20 dB between both ears. STDEV = 3.79%.

**Table 10**

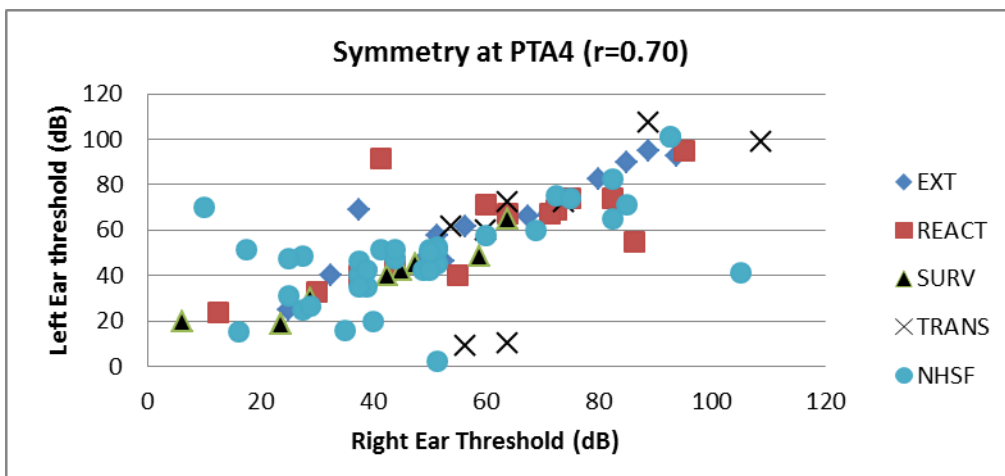
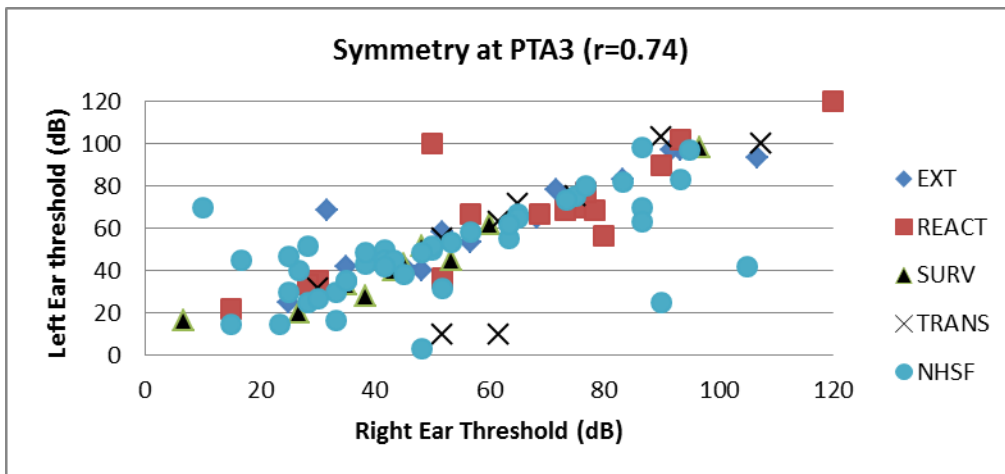
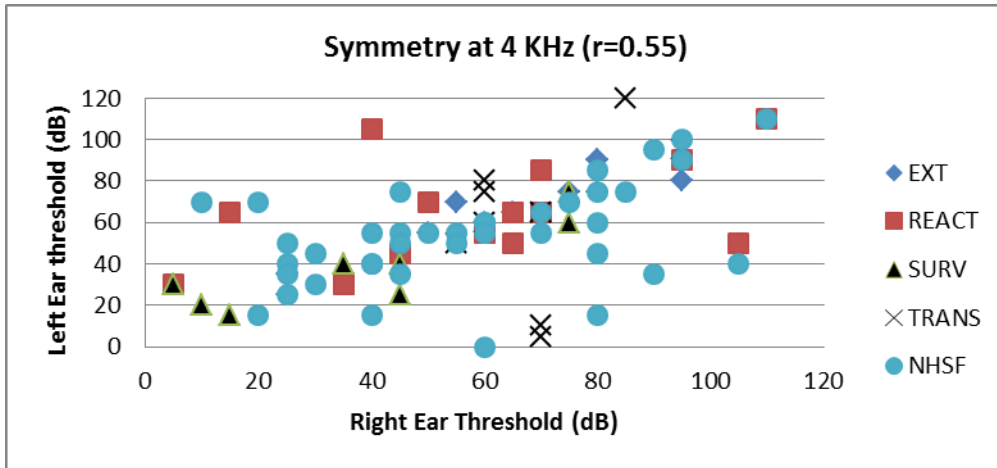
*Asymmetry Cases by Frequency and Referral Type*

Frequency	EXT (n=9)	REACT (n=17)	SURV (n=3)	TRANS (n=14)	NHSF (n=51)
0.5 KHz	2 (22.2%)	3 (17.6%)	0 (0.0%)	2 (14.3%)	9 (17.6%)
1 KHz	2 (22.2%)	2 (11.8%)	1 (33.3%)	2 (14.3%)	6 (11.8%)
2 KHz	3 (33.3%)	4 (23.5%)	1 (33.3%)	3 (21.4%)	13 (25.4%)
4 KHz	0 (0.0%)	4 (23.5%)	1 (33.3%)	3 (21.4%)	10 (19.6%)
PTA 3	1 (11.1%)	2 (11.8%)	0 (0.0%)	2 (14.3%)	7 (13.7%)
PTA 4	1 (11.1%)	2 (11.8%)	0 (0.0%)	2 (14.3%)	6 (11.8%)

Perfect symmetry would consist of scatterplots in which all points fall on the 45 degree line (RE=LE). Greater degrees of dispersion and outliers indicate greater asymmetry. Greater asymmetry is apparent for the cases of IHP reactivation (REACT group) and IHP transfer cases (TRANS).



**Figure 9.** Symmetry Plots at 0.5KHz, 1KHz, 2KHz, 4KHz, PTA3, and PTA4.



**Figure 9.** Symmetry Plots at 0.5KHz, 1KHz, 2KHz, 4KHz, PTA3, and PTA4.



## Chapter 4

### 4 Discussion

#### 4.1 Introduction

Most developed countries, including Canada, have implemented EHDI programs, including universal newborn hearing screening. The literature shows that language development is positively and significantly affected by the age of identification and of intervention services (Yoshinago-Itano, 1998). This critical period for early language development is within the first six months of life (Yoshinago-Itano, 2003).

Universal newborn hearing screening and EHDI programs result in earlier diagnosis of hearing loss and early detection enables early intervention services for affected families. Earlier generations of children could be identified as late as two and a half years of age, after significant developmental opportunities have passed (Yoshinago-Itano et al., 1998).

The current audiological standard is to complete the hearing screening in all infants by one month of age, followed by audiological assessment by three months of age in those identified as at-risk for hearing loss based on the screening outcome, and targeted risk factors, depending on geographical location (JCIH, 2007). For infants with confirmed hearing impairment, intervention should begin by six months of age (Yoshinago-Itano, 2003).

The JCIH recommends that all infants with certain risk factors for other speech or language impairments should be under surveillance for long-term communication development (JCIH, 2007). These at-risk children receive universal neonatal newborn hearing screening, and in addition, should have ongoing medical, speech, language, and

audiological surveillance through their medical “home”. It is considered important that infants who pass neonatal screening, but have one or more risk factors for hearing impairment such as low birth weight, have at least one diagnostic audiological assessment by 24 to 30 months of age. However, the evidence to support these recommendations is generally lacking and a recent study by Beswick et al. (2012) argues that there is a need to examine the effectiveness of newborn screening and surveillance programs through data review and data sharing across infant hearing programs.

## 4.2 Insights from the Literature

The literature reviewed in Chapter 1 is extensive. Key findings in summary form are:

- Early detection is critical. Earlier identification of hearing loss leads to better results in communication, educational achievement, and social development in children.
- UNHS is carried out in Canada and in most developed countries. Its use has dramatically improved outcomes for infants with hearing loss at birth.
- Most, but not all cases of hearing issues are detected by UNHS. Postnatal hearing impairment is complex. Little is understood about the numbers and features of infants who develop significant hearing loss even after passing initial newborn hearing screening.
- Identifying those children who develop hearing problems after passing the initial newborn screening is recognized as a serious challenge in the field of audiology. The data from large databases of

universal neonatal hearing screening programs could be useful for establishing practice and policy for children with different hearing loss outcomes, including progressive and emergent postnatal hearing loss.

There are only a few early detection programs that currently have the capacity to report the number of infants screened, assessed, surveilled, and registered in intervention programs. Moreover, there is a need for evidence-based research to determine the cost effectiveness of dealing with specific surveillance and risk factors because the cost of audiological assessment is a policy factor in determining the feasibility of monitoring services.

A comparison of hearing outcomes across populations that have different risk levels is needed to improve screening and assessment. For example, these populations include infants who pass or fail newborn screening and those with and without protocol defined risk factors. The major contribution of this thesis is in providing a detailed description of the comprehensive data set of children from the Ontario IHP screening and surveillance program at the H.A. Leeper Clinic site.

### **4.3 Research Question 1: Identification of Hearing Impairment**

It was hypothesized that the Referral groups (referral route to IHP) will be related to the hearing assessment outcomes (presence/absence; category and degree of hearing disorder). A specific aim of this study was to look at hearing outcomes in terms of hearing loss present or absent (ie. normal hearing) for the entire population, and for the five different Referral groups. From the total of 2,638 children enrolled at this site, 2,390

were determined to have normal hearing by audiological assessment (full diagnostic and surveillance testing). Of interest, and with potential further consideration, is the considerable difference between the number of hearing impaired cases that were identified as male (62.9%; n=156) versus female (37.1%; n=92) in this thesis.

As per the meta-analysis of Fortnum (2003), the proportion of males to females identified with permanent childhood hearing impairment. This allowed for a calculation of an overall gender ratio for impairment >40dB of 1.16/1.0 (Fortnum, 2003). It is not clear why the proportion (1.70/1.0) of males to females is much higher in this thesis. If gender is consistently predominantly male across research studies for those who are screened and develop hearing loss, this could affect practice and policy in the future.

The detection of hearing loss was also examined within each of the five Referral groups. Infants who failed newborn screening (NHSF), or infants who are recommended for surveillance based on the presence of at least one IHP defined risk factor (SURV), or who are referred externally (EXT), reactivated (REACT), or transferred (TRANS), all are eligible to receive IHP services.

#### 4.3.1.1 NHSF Referral Group Hearing Outcomes

The majority of the cases identified with hearing loss were those children who were NHSF, and therefore had been referred to the IHP with a “refer” result from screening in at least one ear (57.7%; n=143). It is important to note that 15.1% (33/219) of cases labeled “hearing impaired” by manual review were actually cases for which hearing loss type was not yet identified, or unknown. The other 29 charts were not available for manual confirmation and were therefore excluded. Therefore, caution should be exercised in basing conclusions solely on the electronic database.

It was expected that the majority of the cases that developed hearing impairment would have been in the NHSF Referral group, despite the presence or absence of a risk factor. Out of 693 NHSF referrals (failed UNHS), 143 children did develop hearing impairment. However, 79.4% (n=550) children, of the NHSF referred children had normal hearing, which is why it is important to be mindful of how to approach the results of initial screening with parents and caregivers, to alleviate anxiety (Hyde, 2005).

However, considering data that in hospitals with newborn screening compared to hospitals without screening, 84% who received screening were identified prior to six months of age as recommended, while only 8% were detected prior to six months in hospitals without screening (Yoshinago et al., 2001). This demonstrates the benefits of universal screening for developmental outcomes, especially beneficial for those who fail newborn screening (NHSF).

This thesis reported data from the NHSF group, where 693 children failed the screen, and subsequently 20.6% (n=143) were identified with permanent childhood hearing impairment. Another study that examined a group of screened children was conducted by Mason and Herrmann (1998). All of the newborns (n=10,372) were screened in the nursery. In this sample, 415 of these children failed screening, and a subsequent 3.6% (n=15) children were then identified with a permanent childhood hearing impairment (Mason & Herrmann, 1998). Interestingly, a similar study by Barsky-Firsker and Sun (1997) showed results of 10.7% (n=52) from a total of 485 children who failed their screen. It is not clear from the literature why the incidence of permanent hearing impairment is lower in the Mason and Herrmann study.

#### 4.3.1.2 SURV Referral Group Hearing Outcomes

A main point that was noted from the data described in Figure 2 is that the majority of the children referred to the H.A. Leeper Clinic for IHP services from implementation in 2002-2011 were in the SURV group. That means that a total of 1,559 children were assessed who were referred because they had at least one IHP protocol defined risk factor. Out of the 1,559 referred for SURV, 23 SURV cases were identified from the electronic database as hearing-impaired (1.47%; n=23). There were 20 of these 23 charts in the SURV group available for manual review to check for confirmed hearing impairment. There were seven cases upon manual chart review in the SURV group that were still not determined for hearing loss type, however, it is apparent from the threshold data that there is some type of hearing impairment for three of the seven cases. One case was a query for auditory neuropathy, whereas the other two children were queries for sensorineural loss. The remaining four cases had insufficient visit and threshold data, as well as incomplete hearing loss type data from the IHP data forms available.

Therefore, it can be said with more certainty that there are closer to 17/20 SURV children based on chart review with some type of identified hearing impairment. This would give a rate closer to  $17/1,556 = 1.09\%$  detected by surveillance for the entire SURV group referred in the time frame of this thesis.

A study recently published by Beswick et al. (2012) is the only other known large-scale study examining surveillance monitoring outcome data. The goal of the researchers was to describe a targeted surveillance program using risk factors. Beswick et al. (2012) had available a comprehensive risk factor registry, whereas this thesis was only able to account for the presence or absence of at least one risk factor. During their

study period from September 2004-December 2009, 7,320 children were referred for targeted surveillance after passing newborn screening. They found that 0.77% (56/7,320) were identified with postnatal hearing loss (this is the rate of postnatal loss) (Beswick et al., 2012).

This thesis has a similar time frame, from 2002-2011, although our catchment area was smaller, and we had a total of 1,559 children referred to the H.A. Leeper Clinic for surveillance monitoring, although we examined all Referral groups. While comparing the results of this thesis with those of Beswick et al. (2012), this thesis had a postnatal hearing loss rate of 1.48% (23/1,559). These data are from the electronic database and, therefore, may be a slight overestimate of the total confirmed cases of permanent childhood hearing impairment, if some of these cases were still not fully determined.

Another interesting study by Johnson et al. (2005) concluded that of 21 infants who had failed the otoacoustic emissions (OAE) testing but had passed their automated brainstem response (ABR) audiometry during newborn screening developed either bilateral, or unilateral permanent hearing impairment, identified around nine months of age. These researchers show that many of these cases are milder hearing loss and that many of the machines used are designed to identify moderate or greater hearing loss (Johnson et al., 2005). They recommend more surveillance during childhood.

The results from this thesis, which has a higher rate of detected postnatal hearing impairment than found by Beswick et al. (2002) suggest that ongoing surveillance and improved surveillance program evaluation would be useful for improving hearing outcomes.

#### 4.3.1.3 EXT, TRANS, and REACT Referral Group Hearing Outcomes

The EXT group included 11.3% (n=28), TRANS with 8.9% (n=22), and REACT had 12.5% (n=31) of the hearing-impaired sample from the electronic database. These groups are relatively evenly distributed and have more variability in their definition of referral routes. For example, a REACT case could include a child who originally passed UNHS without a risk factor, or who originally passed, with a risk factor, and was cleared after surveillance. The child could be later reactivated due to concern. Conversely, a TRANS case is transferred within the IHP region but identified by another IHP region, and therefore, there is limited knowledge of these cases in terms of their original referral information. Finally, the EXT group could be anyone with screening results unknown, or who moved from outside the country. Other situations for the EXT group could include cases in which the child was born prior to newborn screening implementation. These EXT cases are referred due to some kind of concern, or due to the absence of any initial screening. Therefore, EXT, REACT, and TRANS groups in this thesis are less generalizable than the NHSF, or the SURV group.

#### 4.3.2 Hearing Loss Presence/Absence and Hearing Risk Factor

The risk factor data results were not the main focus of the research questions but are of interest because it is useful information, especially in looking at incidence with the hearing-impaired population. For example, a study by Mehl and Thomson (2002) identified 291 children with congenital hearing loss in The Colorado Newborn Hearing Screening Project from 1992-1999. In this study, 47% of the 291 children were identified with at least one risk factor. Additionally, the study also looked at cases from the year



1999 specifically. In this year, 86 children were identified with hearing loss, and 32 of the 86 children were identified with having at least one risk factor, showing the importance of non-targeted universal screening (Mehl & Thomson, 2002). This thesis found that 66.1% of the hearing-impaired group was identified with at least one risk factor ( $n=164/248$ ), which is slightly higher than the described study by Mehl & Thomson (2002).

A limitation of this thesis was the fact that hearing risk factor data were based on the definition of presence or absence of at least one risk factor, with no specific details provided. However, given this caveat, in the hearing-impaired group, 66.1% had at least one risk factor ( $n=164/248$ ), in contrast to the normal-hearing group, where only 46.2% had the presence of at least one risk factor, based on the electronic database. This thesis also shows that, as expected, there is substantial variation in the presence of risk factors by Referral group. For those children referred from NHSF, a large majority (74.2%) were identified with at least one risk factor. This is again a much larger number than seen in the study by Mehl and Thomson (2002). It would be helpful to know more detailed risk factor data, such as a comprehensive list of risk factors as a necessary component of the IHP protocol, and to have this documented in the IHP database.

Also of importance is that every child in the SURV group had at least one IHP protocol risk factor, which is why they were referred to the IHP Surveillance Program. The risk factor data are incomplete for the normal hearing group based on the electronic database, and those cases were listed as unknown. This was 48.7% of the total sample with risk factor data incomplete from the electronic database. An article by Mason, Gaffney, Green, and Grosse (2008) shows that there is a need for standards for reporting

results in EHDI programs overall. It was taken into account that those cases in the IHP SURV group should have been considered to have at least one risk factor by definition, despite the large number of unknown cases (incomplete data entry), for the normal hearing group, based on the electronic database.

The EXT group had just over half (55.6%) identified with at least one risk factor while the IHP transfer hearing-impaired cohort had 57.9%. For the IHP group that was reactivated, 47% of the cases had at least one risk factor. These three Referral groups are again more difficult to generalize because of the various different ways that the children are transferred or referred. It is therefore expected that there will be more variability than with the NHSF and SURV groups. There is also no known study that has broken down the Referral groups in this manner.

## 4.4 Research Question 2: Nature of Hearing Loss

Research Question 2 focuses on the details of the hearing impairment for the hearing-impaired subjects, and the analysis evaluates these results according to the referral route into the IHP. Among the 248 hearing-impaired children identified via the electronic database, a detailed analysis of the clinical charts was conducted in order to evaluate the nature of the hearing impairment in each subject, and is discussed in the context of the Referral groups.

### 4.4.1 Hearing Loss Type

The literature describes three main types of hearing loss: sensorineural, conductive, and mixed. In addition, this thesis presents data on auditory neuropathy, which has been included in targeted permanent childhood hearing loss since the JCIH

2007 guidelines (JCIH, 2007). This thesis measures the extent of variation in hearing loss by Referral group. To briefly summarize the results shown in the thesis, for all groups, the “bilateral same” category for sensorineural hearing loss has the largest number of cases and conveys the best overall picture when comparing hearing loss across categories. Most neonatal hearing loss is sensorineural, and a known genetic cause is found in 50% of these children, as described by Patel and Feldman (2011), and our results are similar.

Additionally, it is noted that there are substantially more bilateral cases (n=200) compared to unilateral (n=19) and this is clear across Referral group categories, and most pronounced in the NHSF, as expected. Additionally, there is a relatively high proportion of conductive hearing loss unilaterally, which could be due to structural abnormalities, such as microtia. Auditory neuropathy is most common in the NHSF group with 23 cases out of the 29 identified. It would be interesting to examine how many of these cases were Newborn Intensive Care Unit (NICU) cases, although these specific risk data were not available.

There were a relatively large number of cases where the data from the IHP form in the charts were “unknown” bilaterally. There was a total of 32 cases with “unknown bilaterally”. This can be misleading because there are different scenarios where there is threshold data that indicate hearing impairment, but the hearing loss type is not yet confirmed, or the IHP form was left blank, and perhaps the type was, in fact, known. Other reasons include conflicting data, one visit only data, a blank field for hearing loss type, or something is being queried, however, not yet confirmed. For the “different

bilaterally” cases, the majority are where one ear is defined, while the other is unknown.

The same reasons as above apply for why this may be unknown.

Using the “Bilateral same: Behavioral Audiogram” category, the lowest incidence of (sensorineural +conductive+ mixed) divided by total category cases is recorded for the NHSF and for the SURV group (0.28 and 0.30). The other three categories are much higher, all falling in the range from 0.53 to 0.59. When including auditory neuropathy as a fourth hearing loss category, the incidence by referral category is:

- NHSF; (55/128, 0.43)
- TRANS (10/19, 0.53)
- SURV (8/20, 0.40)
- REACT (15/25, 0.60)
- EXT (18/27, 0.67).

This shows that NHSF and SURV continue to have the lowest incidence rates but the difference is smaller.

#### 4.4.2 Severity of Hearing Loss

Although incidence data for hearing loss are important, the severity of the loss is likely a more important indicator of impact. This thesis shows the degree of such loss in total and by Referral group using both PTA3 and PTA4 data and using Clark’s (1981) criteria. Although PTA3 is used more commonly, PTA4 data were also included for comparison with PTA3. Overall, both PTA3 and PTA4 show a significant hearing loss (all from moderate to profound) for approximately two-thirds of the cases.

Overall, the majority of cases were moderate hearing loss, followed by moderately-severe. Of importance, the mild and slight hearing loss groups comprise

32.3% of the overall sample here with PTA3 data. The SURV group included 10 cases with PTA3 data and 8 cases with PTA4 data. The SURV group is the Referral group that had the most cases of mild hearing loss, as well as moderate hearing loss, both at 30% with PTA3 data; results are similar for PTA4. This could indicate that these mild hearing loss cases are not being picked up at newborn screen but would be captured through subsequent surveillance. It is also possible that they were normal hearing at birth and that the hearing loss is emergent, and had progressed to a mild loss at the time of assessment. There is significant evidence that both slight and mild hearing loss can have large effects on language development as well, and this group could benefit from improved efforts in identification and management of hearing loss (Bess, Dodd-Murphy, & Parker, 1998).

In assessing the degree of hearing impairment by Referral groups for both PTA3 and PTA4 data, this thesis shows that both EXT and TRANS Referral groups have the highest proportions in these categories with 0.71 and 0.77 (EXT PTA3 and PTA4) and with 0.89 and 0.89 for TRANS. The lowest severity (at 0.5 for both PTA3 and PTA4) is in the SURV group. If the focus is only on the two most severe categories (severe and profound), the same two categories have relatively more of these cases.

Based on Referral groups and by looking at PTA3, it is apparent that there are more moderate hearing loss cases in general across groups. An interesting point is that compared to PTA4, there are many fewer profound hearing loss cases looking at PTA3. This could have clinical implications for deciding upon severity criteria, as PTA3 data is commonly used as a standard, but perhaps there should be more investigation into the differences between PTA data .

Another puzzling finding is that the greatest degree of profound loss by Referral group is in the EXT group with PTA3 data (21%). This is considerably more than any other group looking at PTA3 or PTA4. However, the EXT group with profound loss looking at PTA4 drops to 8%.

Both the TRANS and REACT groups have a larger proportion of moderately severe loss, with 34% and 37%, respectively. However, the NHSF group trended towards a higher proportion with moderate (29%, PTA3; 34% PTA4) hearing loss and mild hearing loss (23% PTA3; 20% PTA4).

#### 4.4.3 Hearing Loss Symmetry

Hearing loss symmetry is based on comparisons of left ear versus right ear, with asymmetry defined as a difference of >20dB between both ears (Pittman & Stelmachowicz, 2003). This thesis provides important insights into this issue using both tables and scatterplots. Asymmetry is of importance clinically with binaural amplification implications.

Symmetry scatterplots (RE vs. LE) show a very high association in terms of symmetry, with more variation at 2 and 4 KHz. The 2 KHz and 4 KHz test results show a greater proportion of asymmetry cases, and is consistent with other research (Pittman & Stelmachowicz, 2003).

The other four test types are similar in the extent to which they provide lower proportions of asymmetry cases. By Referral group, it is clear that the TRANS group (74%) and the REACT (68%) have the greatest degrees of asymmetry. The lowest

proportion is for the SURV where there were only three asymmetries (15%). These findings are shown clearly and consistently in the tabulations and scatterplots.

The REACT group might have been cases of progressive or mild hearing loss in one ear, and therefore, originally passed screen. This might be a reason for more asymmetry seen in this Referral group. There are, however, no consistent data in the TRANS group and due to sample sizes, it is difficult to make any concrete conclusions.

#### 4.4.4 Hearing Loss Age

This thesis also examined the impact of age of the child on the various hearing frequencies. Age (in months) was then plotted against frequencies and provides age range data for referencing the data. Only the most recent (and most complete) behavioral audiogram data were used for each of the hearing-impaired cases. ABR only audiogram cases were excluded. Scatterplots and simple correlation coefficients show that the degree of association was extremely low in all cases. As a result, this thesis does not focus on age issues which appear to not be a statistically significant factor.

### 4.5 Limitations of Study

This study is based on data from an extensive database at the H.A. Leeper Speech and Hearing Clinic. The database at this IHP site is manually entered. The presence of blank data for the normal hearing group, especially for gender and presence or absence of a risk factor limited the usefulness of these data upon review from the electronic database. This is a common problem with electronic databases and data entry compliance has been similarly cited by Beswick et al. (2012) and the need for standards is stated by Mason et al. (2008). Also, despite best efforts, there were 29 charts out of 248 that could

not be located in the archives. That left 219 charts with hearing impairment that were available for review. It is recognized that any unknown data will affect attrition bias, and it is assumed there will be some human error from data extraction to data entry, although this is not viewed as a significant limitation.

There were also limitations in analyzing risk factor data, due to the definition which can only reliably say whether there was the presence of at least one risk factor. A future goal for the IHP would be to incorporate a comprehensive list that must be checked off based on protocol defined risk factors and that all of these are entered in the electronic database. Data were manually extracted for risk factors for the hearing loss patients through chart review (confirmation of at least one risk factor), although there were many unknown risk data from the electronic database. The charts for the normal hearing cases were also archived and there were 2,390 cases, which were out of the scope of this thesis for manual review. However, referral groups were captured almost entirely for the entire study sample.

In addition, the IHP data forms were not always consistently checked off in every visit for different fields, such as hearing loss type. This is why it was chosen to use the most recent behavioral test when available to define hearing loss type or for choosing PTA data for hearing loss severity and symmetry. Conversely, ABR audiogram only data limited our analysis of the data and were thus kept separately as to not imply that more reliable testing was performed. ABR only data were thus not used in determining the hearing loss severity and symmetry, and kept separate for hearing loss type.



Hearing loss features have many different classification systems, and depending on the chosen methodology, this can affect the interpretation of results. Therefore, Clark's (1981) ASHA accepted criteria were chosen for hearing severity and comparing PTA3 and PTA4 data for contrast, and the NIH (2008) Pittman & Stelmachowicz (2003) criteria were chosen for asymmetry. It is our hope that further research using these data will provide more insight into other features of hearing loss in this sample.

Finally, it is noted that the electronic database showed that there were 23 hearing-impaired cases for the SURV group. However, this does not take into account those cases that are still not yet determined for hearing loss type, and are still being assessed. After further manual review as described in Chapter 3, it is estimated that it is more likely that there were 17 cases with determined PCHI in the SURV group. This difference has an effect on how the Surveillance Program is evaluated and its impact on policy.

## 4.6 Insights for Policy

The different routes that children take to enter the IHP have an important impact on how we look at program evaluation and its effects on policy makers. This thesis is one of the largest scale studies that examines targeted Surveillance Programming, however, the recent study by Beswick et al. (2012) also described a targeted surveillance program. In their findings, they found a lower rate of hearing loss detection through surveillance at 0.77% (56/7,329). In this thesis 1.48% 23/1,559 SURV children were confirmed with hearing loss by the electronic database.

Among their conclusions, Beswick et al. (2012) state that the limitations of the program bring into question its usefulness. They suggest that better time frames are

needed for assessment, assessments performed, and that discharge criteria all need to be revisited (Beswick et al., 2012).

Similarly, this thesis may have also experienced the same challenges and, in addition, the majority of cases that were assessed in the IHP at the H.A. Leeper Clinic were surveillance and normal hearing cases. This thesis did find a slightly higher rate of hearing impairment in our surveillance group compared to the findings of Beswick et al. (2012).

A cost benefit perspective may be useful in considering the current Surveillance Program for the OIHP, with further studies looking at the effectiveness of this program. One possible option might be to move towards a two stage screening process, which would reduce the cost elements of the Surveillance Program. Weichbold et al. (2006) concluded from their study that because some children do not have the targeted risk factors or any at all, another screening around preschool may capture these postnatal hearing loss cases best. However, this change to the IHP would be costly requiring another entire round of screening. In terms of improving community awareness, research programs investigating compliance, standardizing reporting results, and program awareness by physicians, families, and appropriate timepoints in surveillance would also be useful.

The overall conclusion of this thesis is that the IHP electronic database at the H.A. Leeper Clinic provides a unique insight into the effectiveness of the Surveillance Program. However, one recommendation is that the data capture process at the IHP clinic level at Western University should include all information in the clinic charts, and

that the data should be digitized. Electronic management of these data would also ensure that complete referral data, in addition to all details regarding risk factor were captured. This would allow a more feasible way to measure program impact and facilitate research into the epidemiology of permanent childhood hearing impairments as well as hearing intervention outcomes.

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## Appendices

### **Appendix A: IHP Assessment Criteria**

#### **Auditory Brainstem Response (ABR)-based Assessment**

1. Compliance with IHP test parameters.
2. Selection and sequencing of stimulus type, frequency and intensity.
3. Branching to 1 kHz, 4 kHz and AN sub-protocol where indicated.
4. Size and replication of averages.
5. Accuracy of response detection decisions.
6. Appropriateness of EHL estimates.
7. Appropriateness of hearing loss type and severity inferences.
8. Consistency between records and IHP report form.
9. Consistency between records and any textual report.
10. Appropriateness of test strategy across multiple test sessions (if applicable).
11. Timeliness of multiple test sessions (if applicable).

#### **Visual Reinforcement Audiometry (VRA)-based Assessment**

12. Compliance with IHP Protocol for VRA such as the following:
  - a. Evidence of 2 consecutive conditioning trials to establish that the infant is conditioned prior to initiating threshold search
  - b. Evidence of "bracketing" e.g. at least one (-) below MRL when MRL is considered to be established at an elevated (greater than 30dBHL) level.
  - c. Evidence of bone conduction threshold attempts and intervening frequency threshold attempts where indicated.
  - d. Evidence of MRL established for at least 0.5 KHz and 2 KHz for both ears (i.e. assessment is 'finished')
  - e. Evidence of a control trial strategy to ensure a reliability of at least 70%
  - f. A reliability score of 70% or better evident in each complete assessment
  - g. If the reliability score is less than 70%, documentation of an attempt at reassessment should be present (i.e. assessment is 'not finished').

13. Appropriateness of hearing loss type and severity inferences.
14. Consistency between records and IHP report form.
15. Consistency between records and any textual report.
16. Appropriateness of test strategy across multiple test sessions (if applicable).
17. Timeliness of multiple test sessions (if applicable).

#### **Distortion Product Otoacoustic Emissions (DPOAE)**

18. Compliance with IHP test parameters
19. Constancy of autocalibrated stimulus levels.
20. Replication where indicated.
21. Consistency between records and IHP report.
22. Consistency between records and any textual report.

#### **Middle Ear Audiometry (MEA)**

23. Correct probe frequency.
23. Repetition of tympanogram where indicated.
24. Appropriate reflex stimulus levels and repetitions.
25. Consistency between records and IHP report.
26. Consistency between records and any textual report.

## **Appendix B: IHP Definitions of Normal Hearing and Permanent Childhood Hearing Impairment**

### **IHP Normal Hearing Definition**

From the IHP perspective, hearing is 'normal' when the target disorder is deemed not to be present. This is not the same thing as the conventional, clinical meaning of 'normal hearing'. In ABR-based Assessments, clear and reproducible ABRs by air conduction at 0.5 kHz and 2kHz in each ear at the mandatory minimum levels are sufficient to define 'normal' hearing from the IHP perspective. If any other frequency is tested for any reason, a similar result is required. In VRA-based and CPA-based Assessments, a similar inference applies, but only if the VRA thresholds obtained are ear-specific.

Because there are many causes of absent or depressed DPOAEs, normality of OAEs at all frequencies is not necessary for an overall conclusion of IHP 'normal hearing'.

When a 'normal hearing' determination is made, the family should be counseled fully about what exactly is meant by such a result and about the need for continued vigilance. The family should be provided with standard IHP documentation covering issues such as risk indicators, communication development milestones and actions if a concern develops. This information should be provided in the most relevant language available from the IHP.

### **Permanent Childhood Hearing Impairment (PCHI) Present**

The infant is defined to have the target PCHI by any elevation of BC tonepip ABR threshold or VRA MRL of 10 dB or more above the required minimum test levels at 500 Hz or 2 kHz, in either or both ears. In the event that BC testing has proved unfeasible or inconclusive, AC threshold measurements may serve to define sensorineural hearing levels provisionally, provided that immittance results are clearly normal. PCHI is also deemed to be present if AC thresholds are clearly higher than those that could be attributed to purely conductive impairment. PCHI is also deemed to be present if test results indicate the presence of AD.

## **Appendix C: IHP High-Risk Indicators for Permanent Childhood Hearing Impairment**

### *Perinatal*

The following indicators a-l are usually associated with attendance in a special care nursery, whereas indicators m-o may arise from any nursery. ANY ONE of the indicators is sufficient to place the baby at risk. Perinatal indicators are sought by screening personnel, other hospital staff, and the child's physician(s).

At initial Assessment, audiologists should review risk status and should seek risk indicators in children presenting as not at risk. New risk information may arise at any time throughout the child's progression through IHP services.

- a. Birthweight less than 1200 grams
- b. Five-minute APGAR score less than or equal to 3
- c. Congenital Diaphragmatic Hernia (CDH)
- d. Persistent Pulmonary Hypertension of the Newborn (PPHN)
- e. Hypoxic-Ischemic Encephalopathy (HIE), Sarnat II or III
- f. Intra-ventricular Hemorrhage (IVH), Grade III or IV
- g. Peri-ventricular Leukomalacia (PVL)
- h. Extra-Corporeal Membrane Oxygenation (ECMO) or inhaled Nitrous Oxide (iNO) or High-Frequency Oscillatory (HFO) or Jet (HFJ) ventilation
- i. Hyperbilirubinemia  $\geq 400\mu\text{M}$  OR meeting any standard criteria for exchange
- j. Serologically proven cytomegalovirus (CMV) infection
- k. Other proven perinatal TORCHES infection (toxoplasmosis, rubella, herpes, syphilis)
- l. Serologically proven meningitis, irrespective of the pathogen
- m. Familial Permanent Childhood Hearing Impairment
- n. Craniofacial anomaly
- o. Other high risk indicator specified by baby's treating physician

### *Infant (0-24 months)*

All of the above, plus:

1. Parent/Caregiver concern about hearing/speech/language.
2. Postnatal infections associated with sensorineural hearing loss (e.g. bacterial meningitis).
3. Syndromes associated with progressive hearing loss (NFII, Stickler, Usher, etc).
4. Neurodegenerative disorders (e.g. Hunter syndrome) and sensory motor neuropathies (e.g. Friedreich's ataxia, Charcot-Marie-Tooth syndrome).
5. Head trauma sufficient to cause unconsciousness or skull fracture

Newborns known to be at risk on any Perinatal indicator shall be screened only by AABR. Infants who manifest any indicator are targeted for surveillance procedures through **the first two years**. Infants with meningitis may proceed upon recovery directly to fast-tracked Assessment, with subsequent surveillance in the event of a normal initial Assessment.

## Appendix D: IHP Middle Ear Analysis (MEA) Technical Criteria

### Tympanometry

The current IHP protocol is based on discussions in 2003 with Dr Robert Margolis, University of Minnesota, and on normative data kindly provided by him and published later in JAAA.

**For infants under six months corrected age:** Tympanometry shall be done using a 1kHz probe frequency, with repetition as necessary and feasible, to improve reliability. The key abnormality criterion is a compensated peak static admittance of  $\leq 0.6$  mmho, compensated from the negative tail at -400 daPa.

**For infants six months and over corrected age:** Tympanometry shall be done using a 226 Hz probe frequency, with repetition as necessary and feasible, to improve reliability. The key abnormality criterion in the age range 7-12 months is a compensated peak static admittance of **0.1 mmho**, compensated from the positive tail at +200 daPa. From 13-18 months, the criterion is **0.15 mmho**. From 19 months on, the criterion is **0.2 mmho**.

### Middle-Ear Muscle Reflexes

Irrespective of age, acoustic reflexes shall be elicited with a 1 kHz stimulus and measured ipsilaterally, using a 1 kHz probe frequency. Stimulus level shall start at 90 dB and increase in 5 dB steps up to no greater than 100 dB. Note that for a given nominal level, real-ear SPLs in young infants may be up to 20 dB greater than in adults. Reflex presence is defined by a clear, negative deflection, repeatable at any stimulus level.

### Comments

Tympanometry criteria are set at the 5th percentiles of age-specific normative distributions. In the case of double peaks, the large peak is used. Admittance change without development of a genuine peak is abnormal regardless of change size. Caution is required in applying these criteria to young neonates, in whom canal wall collapse may lead to steep negative tails. The clinical utility of other measures such as peak pressure, width and gradient is unclear in infants. Reported 90% range boundaries for TPP are from approximately (-150 to -100) up to (0 to 50) daPa.

## **Appendix E: Types of Assessment and Timing of Initial Assessments**

### **1. Types of Assessment**

Assessments are ABR-based or Behaviour-based. The latter includes Visual Reinforcement Audiometry (VRA), conditioned play audiometry (CPA), or conventional audiometry. The choice of approach is at the discretion of the IHP audiologist, taking account of the individual characteristics of the child and the context and purpose of the Assessment. Assessment may be of Initial, Follow-up or Surveillance types. This protocol shall apply to all types, but test selection and direction of testing effort in the context of follow-up is at the discretion of the IHP audiologist. For Initial Assessments funded by the IHP, the full complement of tests as specified in this protocol is mandatory.

### **2. Timing of Initial Assessments**

Where not medically contra-indicated, Initial Assessments of infants referred from IHP screening shall be targeted at a corrected age of 6-8 weeks. For NICU graduates after extended hospital stays, Initial Assessment shall be targeted within 4 weeks of discharge home, subject to appropriate health status.

Initial ABR-based Assessment shall follow any abnormal result at the IHP Surveillance ABR targeted at 4-6 months corrected age in high-risk infants who pass the AABR screen. For infants at risk who refer on the screen but are normal at Assessment, at least 3 months shall elapse between Assessment and the next Surveillance test, which will usually be VRA-based at 10-12 months. Initial Assessment by other age-appropriate techniques may be indicated by an abnormal finding at any high-risk Surveillance event, up to and including the 30-month family interview. See Appendix C for IHP risk indicators for permanent childhood hearing impairment, which govern eligibility for IHP Surveillance.

Abnormal findings on any IHP Surveillance event shall lead to a full Assessment of the appropriate type, as soon as possible, even at the same visit if test conditions and scheduling permit.

For any infant with a meningitis risk indicator, Assessment is indicated as soon as possible after recovery, if there is a referral into IHP. For this risk indicator specifically, IHP Screening or Surveillance testing prior to full Assessment are NOT appropriate. Special,

non-IHP, fast-track protocols for follow-up of meningitis may be in place locally. Optimal Assessment procedures and timing above and beyond the above specification are currently under review of evidence.

There is accumulating evidence that specific risk indicators other than meningitis may warrant direct eligibility for Assessment and may render screening irrelevant or even inappropriate. Examples may include ear canal atresia and proven cytomegalovirus infection. This matter is also under evidence review.

### **3. Surveillance Assessments**

Surveillance Assessments shall be conducted on all IHP registrants who are determined as at risk by IHP risk indicators. They shall be conducted without regard to passing UNHS or determination of normality at any prior Assessment. At a corrected age of 4-6 months, Surveillance Assessment shall include manual ABR measurement by air conduction at the IHP minimum levels for 2 kHz and 4 kHz bilaterally. DPOAE and MEA testing are discretionary. ABR absence at any minimum level shall lead to prompt, full diagnostic Assessment, which may be a separate appointment or may be initiated at the Surveillance if test conditions and schedules permit.

Surveillance testing at 4-6 months shall NOT be replaced by telephone interview, except in cases of persistent inability or refusal to attend for testing. AABR screening shall NOT be substituted for manual ABR testing.

At a corrected age of 10-12 months, Surveillance Assessment shall include VRA Minimum Response Level (MRL) determination at 2 kHz and 4 kHz bilaterally. DPOAE and MEA testing are discretionary. Any MRL greater than the IHP minimum level shall lead to prompt, full diagnostic Assessment, which may be a separate appointment or may be initiated at the Surveillance, if test conditions and schedules permit.

Surveillance testing at 10-12 months shall NOT be replaced by telephone interview, except in cases of persistent inability or refusal to attend for testing.

Families shall be contacted at a corrected age of as close as possible to 18 months and administered appropriate questioning about auditory responsiveness and early language milestones. Any substantive, questionable finding or parental concern shall lead to prompt,

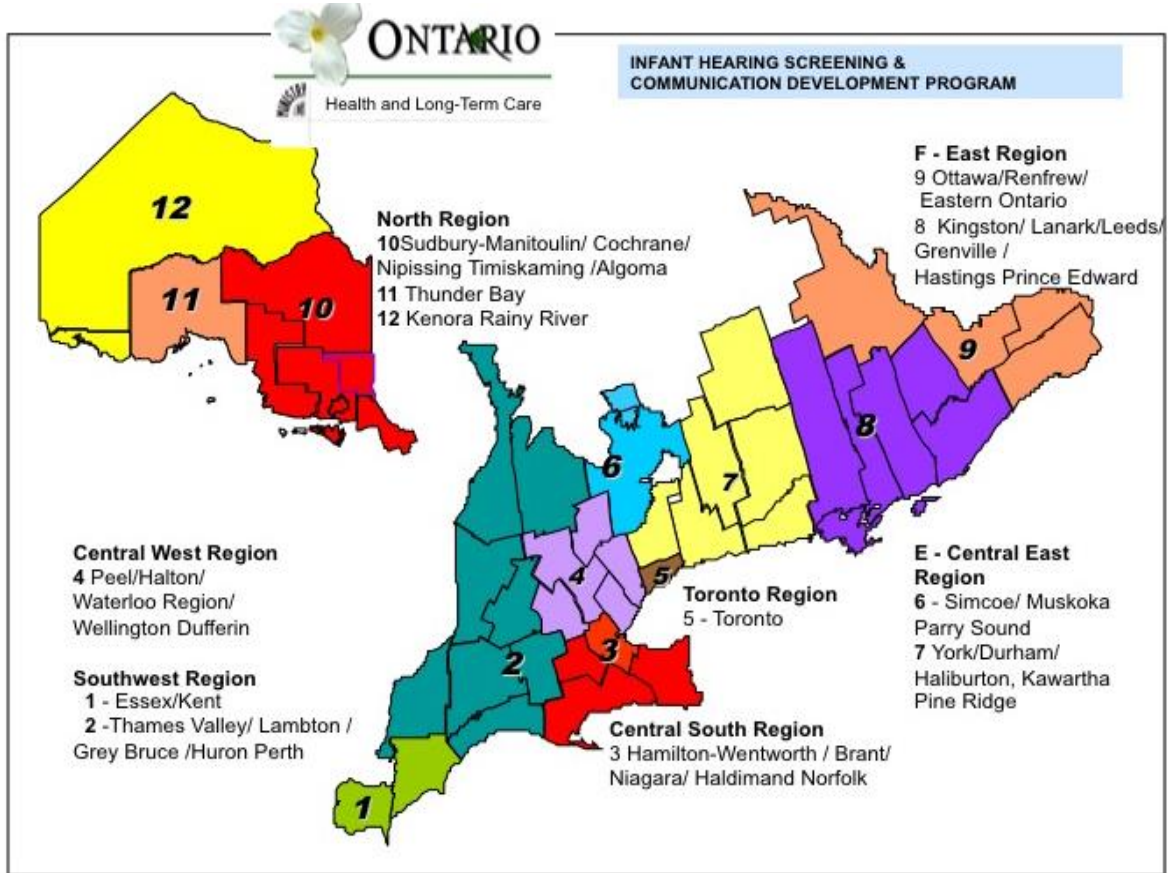


full diagnostic Assessment.

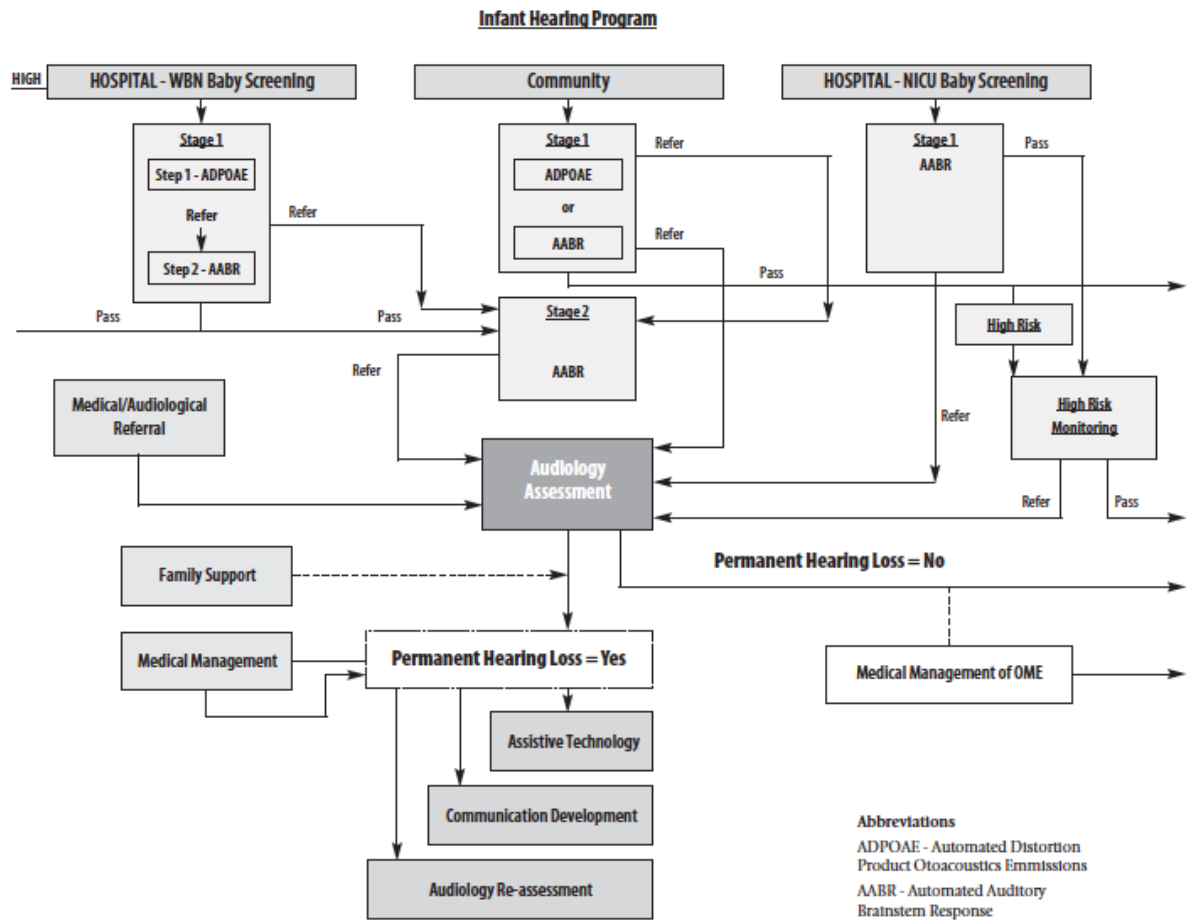
Families shall be contacted at a corrected age of as close as possible to 30 months and administered appropriate questioning about auditory responsiveness and early language milestones. Any substantive, questionable finding or parental concern shall lead to prompt, full diagnostic Assessment.

Infants who pass all the above Surveillance events shall be discharged from the IHP. They may be re-admitted to the IHP only if audiometry outside of IHP and by an audiologist registered with CASLPO has identified probable PCHI. Such audiometry shall not constitute IHP Assessment, but shall be deemed to establish sufficient PCHI risk to justify referral into the IHP for diagnostic Assessment.

### Appendix F: Ontario IHP Regions



## Appendix G: Ontario IHP Schematic Flowchart Protocol



## Appendix H: Hearing Loss Type Tables in Detail

### Hearing Loss Type- EXT (n=27)

Hearing loss type <u>External referral</u>	<u>Unilateral</u> <i>Behavioral audiogram</i>		<u>Bilateral same</u> <i>Behavioral audiogram</i>	<u>Bilateral different</u> <i>Behavioral audiogram</i>		<u>Unilateral</u> <i>ABR audiogram ONLY</i>		<u>Bilateral same</u> <i>ABR audiogram ONLY</i>	<u>Bilateral different</u> <i>ABR audiogram ONLY</i>	
	Right ear	Left ear		Right ear	Left ear	Right ear	Left ear		Right ear	Left ear
Sensorineural	-	-	15	-	-	-	-	3	-	-
Conductive	-	-	-	-	-	-	1	1	1	-
Mixed	-	-	1	1	-	-	-	1	-	-
Auditory Neuropathy	-	-	2	-	-	-	-	-	-	-
Unknown	-	-	-	-	1	-	-	1	-	1

Note. Hearing loss cases with behavioral audiogram (n=19). Hearing loss with ABR audiogram only (n=8). Total with unilateral loss (n=1); total with bilateral loss (n=27), including n=2 cases with one ear "unknown" for hearing impairment. Totalling these numbers does not equal 27 because children appear more than once in the "bilateral different" (columns four and seven).

### Hearing Loss Type- NHSF (n=128)

Hearing loss type <u>UNHS Newborn Refer</u>	<u>Unilateral</u> <i>Behavioral audiogram</i>		<u>Bilateral same</u> <i>Behavioral audiogram</i>	<u>Bilateral different</u> <i>Behavioral audiogram</i>		<u>Unilateral</u> <i>ABR audiogram ONLY</i>		<u>Bilateral same</u> <i>ABR audiogram ONLY</i>	<u>Bilateral different</u> <i>ABR audiogram ONLY</i>	
	Right ear	Left ear		Right ear	Left ear	Right ear	Left ear		Right ear	Left ear
Sensorineural	4	2	32	1	5	-	3	14	6	7
Conductive	4	-	2	-	-	-	-	1	2	2
Mixed	-	-	2	-	-	-	-	2	2	2
Auditory Neuropathy	-	-	19	-	-	-	-	3	-	1
Unknown	-	-	10	5	1	1	-	6	7	5

Note. Hearing loss cases with behavioral audiogram (n=81). Hearing loss with ABR audiogram only (n=47). Total with unilateral loss (n=14); total with bilateral loss (n=115), including n=23 cases with one ear "unknown" for hearing impairment. Totalling these numbers does not equal 128 because children appear more than once in the "bilateral different" (columns four and seven).

*Hearing Loss Type- SURV Group (n=20)*

Hearing loss type IHP Surveillance Group	Unilateral Behavioral audiogram		Bilateral same Behavioral audiogram	Bilateral different Behavioral audiogram		Unilateral ABR audiogram ONLY		Bilateral same ABR audiogram ONLY	Bilateral different ABR audiogram ONLY	
	Right ear	Left ear		Right ear	Left ear	Right ear	Left ear		Right ear	Left ear
Sensorineural	-	1	2	1	1	-	-	2	-	-
Conductive	-	-	2	-	-	-	-	-	-	-
Mixed	-	-	2	-	-	-	-	-	-	-
Auditory Neuropathy	-	-	2	-	-	-	-	-	-	-
Unknown	-	-	6	1	1	-	-	1	-	-

Note. Hearing loss cases with behavioral audiogram (n=17). Hearing loss with ABR audiogram only (n=3). Total with unilateral loss (n=1); total with bilateral loss (n=20), including n=2 cases with one ear “unknown” for hearing impairment. Totalling these numbers does not equal 20 because children appear more than once in the “bilateral different” (columns four and seven).

*Hearing Loss Type- REACT (n=25)*

Hearing loss type IHP reactivation	Unilateral Behavioral audiogram		Bilateral same Behavioral audiogram	Bilateral different Behavioral audiogram		Unilateral ABR audiogram ONLY		Bilateral same ABR audiogram ONLY	Bilateral different ABR audiogram ONLY	
	Right ear	Left ear		Right ear	Left ear	Right ear	Left ear		Right ear	Left ear
Sensorineural	-	-	13	1	-	-	-	1	-	-
Conductive	-	-	-	-	-	-	-	-	-	-
Mixed	-	-	1	-	-	-	-	1	-	-
Auditory Neuropathy	-	-	1	-	-	-	-	1	-	-
Unknown	-	1	3	-	1	-	-	2	-	-

Note. Hearing loss cases with behavioral audiogram (n=20). Hearing loss with ABR audiogram only (n=5). Total with unilateral loss (n=1); total with bilateral loss (n=24), including n=1 cases with one ear “unknown” for hearing impairment. Totalling these numbers does not equal 25 because children appear more than once in the “bilateral different” (columns four and seven).

*Hearing Loss Type- TRANS (n=19)*

<u>Hearing loss type</u> IHP transfer in	<u>Unilateral</u> <i>Behavioral audiogram</i>		<u>Bilateral same</u> <i>Behavioral audiogram</i>	<u>Bilateral different</u> <i>Behavioral audiogram</i>		<u>Unilateral</u> <i>ABR audiogram ONLY</i>		<u>Bilateral same</u> <i>ABR audiogram ONLY</i>	<u>Bilateral different</u> <i>ABR audiogram ONLY</i>	
	Right ear	Left ear		Right ear	Left ear	Right ear	Left ear		Right ear	Left ear
Sensorineural	1	-	7	-	-	-	-	2	1	1
Conductive		-	1	-	-	-	-	-	-	-
Mixed	-	1	2	-	-	-	-	-	-	-
Auditory Neuropathy	-	-	-	-	-	-	-	-	-	-
Unknown	-	-	2	-	-	-	-	1	1	1

*Note.* Hearing loss cases with behavioral audiogram (n=14). Hearing loss with ABR audiogram only (n=5). Total with unilateral loss (n=2; total with bilateral loss (n=17), including n=2 cases with one ear "unknown" for hearing impairment. Totalling these numbers does not equal 19 because children appear more than once in the "bilateral different" (columns four and seven).

## Appendix I: Severity of Hearing Loss by Referral Group Tables (PTA3 and PTA4)

*Degree of Hearing Impairment by Referral Group – PTA3*

Classification	Hearing Level (dBHL)	Referral Group				
		EXT (n= 14)	NHSF (n= 44)	REACT (n= 16)	TRANS (n= 9)	SURV (n= 10)
Normal hearing	-10 to 15	0	1 (2.3%)	0	0	0
Slight hearing loss	16 to 25	1 (7.1%)	6 (13.6%)	1 (6.3%)	0	2 (20%)
Mild hearing loss	26 to 40	3 (21.4%)	10 (22.7%)	3 (18.8%)	1 (11.1%)	3 (30%)
Moderate hearing loss	41 to 55	3 (21.4%)	13 (29.5%)	2 (12.5%)	2 (22.2%)	3 (30%)
Moderately severe hearing loss	56 to 70	2 (14.3%)	7 (15.9%)	6 (37.5%)	3 (33.3%)	1 (10%)
Severe hearing loss	71 to 90	2 (14.3%)	6 (13.6%)	2 (12.5%)	2 (22.2%)	0
Profound hearing loss	> 90	3 (21.4%)	1 (2.3%)	2 (12.5%)	1 (11.1%)	1 (10%)

*Degree of Hearing Impairment by Referral Group – PTA4*

Classification	Hearing Level (dBHL)	Referral Group				
		EXT (n= 13)	NHSF (n= 35)	REACT (n= 14)	TRANS (n= 9)	SURV (n= 8)
Normal hearing	-10 to 15	0	0	0	0	0
Slight hearing loss	16 to 25	1 (7.7%)	6 (17.1%)	1 (7.1%)	0	2 (25%)
Mild hearing loss	26 to 40	2 (15.4%)	7 (20%)	3 (21.4%)	1 (11.1%)	2 (25%)
Moderate hearing loss	41 to 55	3 (23.1%)	12 (34.3%)	3 (21.4%)	1 (11.1%)	3 (37.5%)
Moderately severe hearing loss	56 to 70	3 (23.1%)	4 (11.4%)	4 (28.6%)	4 (44.4%)	1 (12.5%)
Severe hearing loss	71 to 90	3 (23.1%)	5 (14.3%)	2 (14.3%)	2 (22.2%)	0
Profound hearing loss	> 90	1 (7.7%)	1 (2.3%)	1 (7.1%)	1 (11.1%)	0

## Curriculum Vitae

**Name:** Katherine Smith

**Post-secondary Education and Degrees:** McGill University  
Montreal, Quebec, Canada  
2000-2004 H.Bsc.

The University of Western Ontario  
London, Ontario, Canada  
2010-2013 M.Sc.

**Honours and Awards** Scholarship valued at \$10,000 over two years with Teacher Assistant responsibilities.  
2010-2012

Certified Clinical Research Professional (C.C.R.P)- 2008-Present

**Related Work Experience** Teaching Assistant  
The University of Western Ontario  
2010-2012

Intern at the World Health Organization, Health Policy Research in Social Equity and Human Rights, Geneva, Switzerland  
2012 (four months)

Clinical Research (Pediatric Oncology), London Health Sciences Centre, London, Ontario. 2010-2012

Clinical Research Associate, The Hospital for Sick Children, Toronto, Ontario. 2007-2010

Senior Project Coordinator, MDS Pharma Services, Project Management, Mississauga, Ontario. 2006-2007

Project Coordinator, MDS Pharma Services, Project Management, Mississauga, Ontario. 2004-2006

Office Administrator, Health Canada, Pest Management Regulatory Agency, Ottawa, Ontario, Sept. 2004-Nov. 2004



- Posters** “Disseminating Major Treatment Change Information to Study Patients- The Need for an SOP & Checklist”. – Presented at Annual Children’s Oncology Group Conference, October 2008
- Article** Children’s Oncology Group Clinical Research Associate Bulletin, “Targeted Toxicity Reporting”, February 2010
- Committees** Children’s Oncology Group Full Membership  
Society of Clinical Research Associates Accreditation