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Population Infant Hearing Screening to Intervention:
Determinants of Outcome from the Parents' Perspectives

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Population Infant Hearing Screening to Intervention: Determinants of Outcome from the Parents' Perspectives

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Thesis submitted to the Faculty of Graduate and Postdoctoral studies in partial fulfillment of the requirements for the PhD degree in Population Health

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However beautiful the strategy, you should occasionally look at the results.

Winston Churchill

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LIST OF ABBREVIATIONS

ASL: American Sign Language
CA: conjoint analysis
CDC: Centers for Disease Control and Prevention
CDI: Child Development Inventory
CHEO: Children's Hospital of Eastern Ontario
CI: confidence interval
CWGCH: Canadian Working Group on Childhood Hearing
dB: decibels
dB HL: decibels hearing level
EHCD: early hearing and communication development
EHDI: early hearing detection and intervention
ENT: Ear Nose and Throat
GFTA: Goldman-Fristoe Test of Articulation
HSC: Hospital for Sick Children
ICF: International Classification of Functioning, Disability, and Health
IHP: infant hearing program
JCIH: Joint Committee on Infant Hearing
LTLF: Learning to Listen Foundation
NICU: neonatal intensive care unit
PCHI: permanent childhood hearing impairment
PLS: Preschool Language Scale
PPVT: Peabody Picture Vocabulary Test
RCT: randomized controlled trial
SD: standard deviation
UNHS: universal newborn hearing screening
USPSTF: United States Preventive Services Task Force
UK: United Kingdom
US: United States of America
WHO: World Health Organization

Abstract

Background

Childhood hearing loss has a negative impact on healthy child development and is associated with poor outcomes in communication, social and academic development. Population-based infant hearing screening has received worldwide attention as an opportunity to improve developmental outcomes for children with hearing loss. Universal newborn hearing screening (UNHS) has become standard protocol in many countries and has recently been implemented in several Canadian provinces and territories. This study concerns population-based infant hearing screening and the potential benefits of this intervention for children and families. While there is evidence that screening can accurately identify babies with hearing loss before three months of age, less information is available on the effectiveness of early identification initiatives and how to maximize the potential opportunities provided by early detection of hearing loss. It is well recognized that UNHS must be part of an early hearing and communication development program.

Objectives

The objectives of this thesis were to contribute to this developing field by: 1) exploring the effectiveness of infant hearing screening through traditional communication outcome measures, 2) identifying process and other outcomes that parents view as important benefits of early diagnosis of hearing loss, 3) examining parents' needs following identification of

hearing loss, and 4) exploring parent preferences for service delivery following the diagnosis of hearing loss.

Methods

Following a comprehensive review of the current state of knowledge, this study used combined quantitative and qualitative methodology approaches to examine these objectives through three interrelated inquiries: 1) an exploration of data from a prospective longitudinal study investigating the impact of early identification, 2) semi-structured interviews with parents whose children were diagnosed through screening and traditional referral routes and 3) a conjoint analysis survey to quantify parent preferences for service delivery.

Conclusions and Implications for Practice and Policy

Applying a population health perspective, this research defined broader outcomes of early identification of childhood hearing loss from the perspective of families and highlighted contextual factors such as access to parent support and coordinated services, which may be important determinants of outcome to consider in program evaluation of screening initiatives. This study can inform the development and implementation of population hearing screening programs as they continue to grow across Canada and elsewhere. This project has the potential to impact health practice and policy by providing evidence-based direction for the delivery of services to young children with hearing loss and their families.

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CHAPTER 1

INTRODUCTION AND LITERATURE REVIEW

Chapter 1

INTRODUCTION AND LITERATURE REVIEW

Statement of the Problem

Population infant hearing screening has become a worldwide initiative. Universal newborn hearing screening (UNHS) has joined numerous other newborn screening programs as the standard of care in many developed countries and more recently has been introduced in several Canadian provinces and terrritories.[1] Advances in technology have made screening of newborn hearing a viable practice. Consequently, childhood hearing impairment has received increasing attention as a public health issue in the last decade. Hearing loss is one of the most common congenital disorders, affecting 1 to 3 per 1000 children[2,3] and has negative consequences for language, social and academic development. Public health interventions are aimed at improving long-term outcomes at a population level. Underlying the advocacy for UNHS are assumptions that screening is accurate, efficient, and leads to improved results in communication development. New developments in amplification and biomedical devices such as cochlear implants have improved treatment options for children with hearing loss. UNHS is a population health intervention that is expected to enhance the potential of children with hearing loss to develop competent oral communication skills and therefore achieve fuller participation in society.

Until the 21st century, Canada had a rather incongruous approach to infant hearing screening with targeted high-risk screening available in a small number of hospitals, based on the

interest of leaders in the field rather than on health policy.[4] Programs varying in scope and complexity were established primarily through local initiatives.[4] An example of one such program was reported in a population-based study of age of diagnosis.[5] In this program, babies in a pediatric neonatal intensive care unit in Ottawa who were systematically screened were identified at a mean age of 5.7 months while physician referral led to identification at a mean age of 2.8 years. Canada first introduced provincially mandated programs in 2002 with the establishment of the Infant Hearing Program (IHP) in the province of Ontario, Canada's most populous province, representing approximately 34% of the country's population. The provinces of Alberta, Prince Edward Island, New Brunswick and British Columbia have also implemented or announced population screening initiatives such that approximately 50% of the country is currently in various stages of program development.[1]

Consistent with the escalation of UNHS initiatives in this country and elsewhere, interest has grown in evaluating the real world effectiveness and value of the intervention. However, there are widely expressed concerns about the costs of population screening and its ability to positively impact the long-term effects for children with hearing loss and their families.[6] Direct measurement of the effectiveness of UNHS is problematic for several reasons. Therefore, proxy measures such as screening yields, age of identification and fitting of hearing devices have been used to quantify the impact of newborn screening. However, the real benefit of screening has been traditionally considered to be the ability to reduce the negative consequences associated with hearing loss.[7,8] These negative consequences have

been primarily defined as poor communication and academic outcomes. Although, a systematic review commissioned by the United States Preventive Services Task Force (USPSTF) concluded that there was good evidence for the early identification and treatment of hearing loss through screening, the USPSTF would not endorse routine newborn hearing screening. The latter decision was made on the basis of insufficient evidence that screening leads to improved speech and language skills.[6]

To date, there have been no randomized controlled trials of the efficacy of UNHS and few controlled studies of the benefits of screening and/or early intervention in reducing communication and other developmental delays associated with childhood hearing impairment. Various factors other than age of identification may influence developmental outcomes in children with hearing loss. These include degree of hearing loss, family involvement, concomitant disorders, use of hearing technology, type of intervention and service delivery models.[9] Disentangling the effects of these multiple influences is complex and explains why there remains considerable uncertainty about the realistic gains in communication development that can be achieved as a direct result of newborn screening programs. Accordingly, advocates of newborn hearing screening have highlighted the importance of examining benefits beyond traditional communication development outcomes.[1,10]

Early research efforts and quality indicators were focused on the effectiveness of UNHS in accurately and efficiently identifying infants with hearing loss. Several key organizations

represented by the Joint Committee on Infant Hearing (JCIH) recommend that hearing loss in infants be identified by three months of age with appropriate intervention initiated by six months of age.[11] The value of screening is linked to the presumed benefits resulting from early intervention, that is, improved communication development which is expected to translate to improved quality of life and employment opportunities. However, screening as the path to earlier identification, is the first step in the process to improve developmental outcomes in children with hearing loss. Proponents agree that intervention and family support are crucial to the success of UNHS.[12] The terms Early Hearing Detection and Intervention (EHDI) and Early Hearing and Communication Development (EHCD) are often used to illustrate that hearing screening is only one step in a complex system of care. The emphasis in newborn hearing screening is shifting from outcomes of the screening process itself to a focus on the essential components of the process after screening, that is, the full process of care.

Children with significant degrees of hearing loss require habilitation to progress in communication development. A comprehensive habilitation service encompasses several components including audiologic assessment/intervention (including amplification devices), family counseling and support, community outreach, communication intervention and documentation of outcomes.[12]

Many elements of the care process for children with hearing loss are lacking a sound science-base. For example, historically, there has been considerable uncertainty around the

most effective therapy methods for developing communication in children with hearing loss. Significant philosophical differences define different intervention approaches. Intervention options range from communication approaches aimed at developing overall communication skills including sign language to those focused exclusively on the development of auditory and spoken language skills. The effectiveness of various approaches in developing communication is largely inconclusive.[13] In addition, service delivery models are varied and largely undefined. Yet, intervention models may have a significant impact on whether the potential benefits of early identification through systematic screening can be realized.

The characteristics of a service model may have a considerable influence on the ability of a family to access and benefit from early intervention programs. For example, it is not known whether families of very early diagnosed children need different information or adjustments in the model of care particularly in the early stages. Families who discover hearing loss through a system-driven model have not experienced the grieving process in the same way as those who suspect their child's hearing problem long before receiving the diagnosis. Moreover, the ability of a family to access services, to "comply" with advice and specific therapy programs at home and to modify the child's learning program is affected by family circumstances.[14]

A new paradigm of health care has evolved in the past decade with a focus on services that are family-centred and designed to meet an individual's medical, educational and psychosocial needs. In this model, there is a greater emphasis on treatments that address

contextual factors as well as the "patient".[15] The notion of family-centred or family friendly care has been advocated by many in the health care literature on children with hearing loss and other disabilities.[12,16-18] Family-centred care is a holistic approach to treatment and necessitates a system of care that is organized to provide a broad range of medical, therapeutic, and social support services.[19] The system of care for children with hearing loss has developed in a patchwork fashion over time frequently with fiduciary and administrative responsibilities in multiple agencies including health, education, and social services. This was identified as an important barrier to the implementation of newborn hearing screening programs in the United Kingdom (UK).[20]

The attention on UNHS has prompted interest in examining the influence of other factors that work in concert with newborn screening to impact developmental outcomes in young children. Considerable attention and research have been accorded to the technical aspects and yield of UNHS programs. In comparison, relatively little attention has been accorded to other aspects of post-screening care such as the appropriateness of service delivery models for families. Thus, the contribution of screening as one component of a hearing health services package aimed at reducing disability may be affected by many family and environmental factors. The abilities of parents to participate in intervention programs may be related to many factors including culture, socio-economic circumstances, geographical disparities, beliefs and supports. These contextual factors may place children at risk for poor outcomes despite the potential advantages of early diagnosis through population screening. Service models have received little attention by way of dedicated scientific

investigation yet this may be one of the most important determinants of outcome in children with hearing impairment.

Purpose and Objectives

This project was undertaken as part of a research program that investigated a variety of issues related to UNHS as programs are implemented across Canada. The indications of continuing growth in the number of UNHS programs suggest the need for a better understanding of the potential benefits of screening as well as a comprehensive approach to implementing services for families of children with hearing loss. In order for UNHS to impact on the health of children, the nature of families' needs and the important components of a care package must be determined such that these can later be carefully assessed through process evaluations. Otherwise, it will be difficult to evaluate the true benefits of universal screening for Canadian children.

The purpose of this study was to better understand the benefits of screening both with respect to measurable communication outcomes in children and parents' perceptions of the utility of screening (i.e., positive and negative aspects of screening). A second goal of the study was to identify the essential components of family-centred care from the families' perspectives and the relative value parents place on the various attributes of programs for young children with permanent hearing loss.

The study was carried out in three inquiries to support the following research questions:

1. Is there an association between age of diagnosis and communication outcomes in children with hearing loss?
2. How do families of children with hearing loss perceive their experiences with the diagnosis and the impact of age of diagnosis on the family in the early stages?
3. What are families' needs following the diagnosis of childhood hearing loss?
4. What are parents' preferences for the various attributes of service following diagnosis of hearing loss?

Present State of Knowledge

Efforts are underway throughout the world to establish universal hearing screening as part of EHDI programs that have been developed using the best available evidence. The proliferation of newborn screening programs in the past 10 years has spurred a tremendous growth in the body of research in infant hearing science. A comprehensive literature search was conducted for this doctoral study.

Study Identification: The search strategy consisted of searches of a variety of electronic databases using the strategy published by the USPSTF team[6] (Appendix 1), which was developed for a systematic review of the effectiveness of universal newborn hearing screening. Initial searching for this thesis covered the period January 1995 to April 2005 and was updated in October 2006. Searching was restricted to all publications in English or French. The search strategy was applied to: Medline, CINAHL and EMBASE, as well as

CENTRAL (the Cochrane Controlled Clinical Trial Register). In addition to the search for primary studies, the Cochrane Database of Systematic Reviews and the Database of Abstracts of Reviews of Effectiveness were searched to identify systematic reviews. References listed in identified studies and consultations with experts in the field provided an additional route to published studies and documents.

This search located several systematic and critical reviews that have recently synthesized the evidence for the effectiveness of UNHS in identifying hearing loss and improving communication outcomes. The first major review commissioned by the USPSTF focused on the accuracy of screening and the benefits of early diagnosis for children with hearing loss.[6] The mission of the USPSTF is to evaluate medical procedures and appraise the level of evidence for procedures that affect standards of health care. Since the publication of this review, the Canadian Working Group on Childhood Hearing (CWGCH) has also published a number of critical reviews in a resource document intended to inform guidelines related to early hearing detection or Early Hearing and Communication Development (EHCD) as it is commonly called in Canada.[10] The CWGCH was established in 2000 to review and assess evidence related to newborn hearing screening and management. The CWGCH also supported a recent systematic review to synthesize the literature on the effectiveness of various intervention methods in the management of children with hearing loss. In addition to the USPSTF and the CWGCH reviews, one recent systematic review comparing UNHS with targeted screening was located in the Cochrane database.[21] However, as no published studies met the review's inclusion criteria of randomized controlled trials, the review report

does not illuminate the questions addressed in this study and will not receive further discussion.

Given the extant systematic and critical reviews undertaken by experienced researchers and judged to be of good quality, a systematic review was not conducted in preparation for this thesis. The published reviews establish an important knowledge base for the rationale and scientific evidence for UNHS and the findings that are relevant to this study will be summarized in the appropriate domains. When appropriate, new findings from the current literature search will be included to update the core evidence for the study. In this literature review, an emphasis was placed on publications since 2002, which covers the period since the CWGCH critical reviews. In particular, a series of reports in 2005 and 2006 from an evaluation of the first phase of the newborn screening program in the UK contributes updated information to the evidence-base in this field. Seven subject domains were identified as useful to a review of knowledge in infant hearing and communication development, although there is some overlap between the topics and therefore articles sometimes appear in more than one domain. A separate section has not been dedicated to economic costs as few data are available on the economic costs or cost-effectiveness of hearing screening; where information is available it has been embedded in the section on the effectiveness of newborn screening.

Epidemiology of Childhood Hearing Loss

Permanent hearing loss is reported to be one of the most common of the main pediatric congenital disorders.[22] Consistent with the approach of the CWGCH, only studies from developed countries were considered in this review, given the potential variation in prevalence in different populations. Regional and national data from screening programs in the US and the UK show prevalence estimates ranging from 1 to 3 per 1000 newborns.[2,3,23,24] The variation in prevalence rates reported in the literature can be explained by the complexity and uncertainty in reporting infant hearing loss figures.[10,25] There is considerable variation in studies related to how hearing impairment is defined in relation to severity, frequency range, type of impairment as well as whether the hearing disorder is unilateral or bilateral. Other sources of variation include random sampling error and geographic variation.[10] While no national figures are available for Canada, an estimated 300 children are born with, or acquire, significant permanent hearing loss each year in the province of Ontario.[26] Extrapolating to the Canadian population, approximately 1000 to 1200 new cases of childhood hearing loss will be diagnosed annually.

An examination of the evidence for the prevalence of permanent childhood hearing impairment (sensorineural and permanent conductive impairments) in the age range 0 to 5 years was conducted by the CWGCH. Studies in this review met the following inclusion criteria: 20 or more cases identified in retrospective studies, 20,000 or more babies screened in prospective UNHS cohort studies and 1000 or more babies screened in targeted screening

cohort studies. Studies from non-industrialized countries were excluded on the basis of potentially different prevalence rates and consequently limited applicability to Canada. A total of 6 ascertainment studies (1994 to 2001) and 14 UNHS prevalence studies (1998 to 2002) were retrieved as part of the CWGCH critical review.[10]

The most complete prevalence data from ascertainment studies are provided by two studies from the UK. Fortnum and colleagues[2] ascertained 17,160 cases and reported an adjusted prevalence of 1.07 cases at age 3 years, increasing to 2.05 cases at 9 years of age and over for hearing losses of more than 40 dB HL. These statistics indicate that the prevalence of childhood hearing loss may double after the neonatal period. Similar data were reported from the second UK ascertainment study conducted in Trent region with a prevalence of 1.33 per 1000 in the 5 to 10 year age range for losses of equal or greater than 40 dB HL.[27] Since the CWCGH review, results from an ascertainment study[28] of a 1993 birth cohort of 64,115 babies in Victoria, Australia (population 4.4 million), showed a prevalence of congenital hearing loss of 2.09 per 1000 by age 6 years. The conclusions were derived from the number of children presumed to have congenital loss who were fit with hearing aids by age 6 years, of which 40% were mild losses (20-40 dB HL) and the remainder moderate or greater loss (1.12/1000).

Prevalence data from the 14 prospective screening studies included reports with considerable variation in prevalence rates.[10] This can be accounted for by the wide variation in definition of the target disorder which ranged from more than 20 dB HL in one

ear (unilateral impairment) for one frequency to more than 40 dB HL (pure-tone average) in the better ear (bilateral impairment). Regardless of the definition of the disorder for these studies, the CWGCH report cautions that prevalence rates are limited by the performance characteristics of screening tools and the ability of current audiologic diagnostic techniques to accurately quantify milder degrees of hearing loss, particularly in young children.

The review judged that the most comprehensive data have been provided by a series of reports from the New York State demonstration project.[3] The prevalence reported for 43,311 infants using a target disorder of greater than 20 dB HL at any frequency was 1.96 per 1000 screened. The CWGCH reports an estimate (adjusted for loss to follow-up) of 3.2 cases per 1000 for five UNHS programs addressing the target disorder described above. In particular, the different cut-offs used in determining a clinical case will have a large impact on the prevalence of the disorder. However, as pointed out by other researchers, one limitation of prospective screening studies is that true cases are unknown until children reach an age where the hearing loss can be determined.[29] A recent report of yields from a 5 year retrospective review (1997 to 2001) of newborns screened at a US tertiary care center compared favourably with previously published studies.[30] Of 17,602 newborns screened, 1 in 811 (1.2 per 1,000) low-risk and 1 in 75 (13.3 per 1000) high-risk neonates were identified with hearing loss.

In the systematic review commissioned by the USPSTF,[6] only two studies of the yield of universal screening were rated as good-quality studies. The only controlled trial of UNHS

which was conducted in Wessex, England reported 94 per 100,000 cases of permanent childhood hearing impairment (PCHI) referred by age 6 months.[31] The second study, the New York State UNHS state-wide project, reported a yield of 68 per 100,000 target population.[3] Studies that were rated as fair or poor quality in this systematic review had higher yields which this review concluded to be due to the inclusion of infants with unilateral, mild or unconfirmed hearing loss. These results were corroborated in a recent publication from the first phase of the implementation of the UK newborn screening program which reported a yield of permanent bilateral hearing loss (moderate or greater degree) of 1.0 per 1000 babies screened.[32]

In summary, the CWGCH critical review concluded that there is fair evidence for the prevalence of congenital permanent childhood hearing loss of greater than 20 dB HL at any frequency (from 500 to 4000 kHz) and in any ear of 2 to 3 per 1000 live births.

Individual and Societal Impact of Childhood Hearing Loss

Childhood hearing loss interferes with the typical development of spoken language with far reaching consequences in other domains including, social, academic and eventual quality of life.[20,23,33] Historically, communication outcomes in children with all degrees of hearing impairment have lagged far behind the developmental trajectory of hearing children. However, cochlear implant technology has greatly improved access to hearing for children with severe to profound hearing loss and a large body of literature documents the positive effects on speech and language outcomes[34-36] (Fitzpatrick et al, unpublished manuscript). In

the largest outcome study of 181 children with cochlear implants, children 7 to 9 years of age demonstrated high levels of speech, language, auditory and reading skills.[36] Approximately 50% of the participants achieved speech intelligibility and reading scores comparable to hearing children of the same age. Despite these more encouraging results for children with profound hearing losses, on average, studies show that children with various degrees of hearing loss exhibit lower levels of communicative competence than their normal hearing peers.[37,38]

In particular, two recent studies demonstrated that communication development in children with hearing loss generally remains inferior to that of their normal hearing peers. A large population-based study reported outcome data for 89 of 132 eligible children age 7 to 8 years in the state of Victoria, Australia.[37,39] This population of children demonstrated significant delays in communication functioning compared to normal hearing peer normative data. Children performed 1.3 to 1.7 standard deviations below the normal hearing population on standardized language measures. Mean language scores were inversely related to severity of hearing loss but showed a weak correlation with socioeconomic factors. In addition to language outcomes, parent-reported health-related quality of life measures were significantly lower than normative population scores. A second relevant study in the UK collected communication outcome data on 120 children identified with or without screening (birth cohort 1993 to 1996). The findings showed that while early identified children performed better than later identified children on oral language measures,

children with hearing loss, on average, attained speech and language scores significantly below the normal hearing comparison group.[38]

Few published data are available on the economic impact of hearing loss of varying degrees of severity, but Mohr and colleagues [40] have documented the societal burden of hearing loss in the more than 600,000 people in the US with severe to profound hearing impairment. Fifty-three percent (53%) have a family income of less than $25,000 compared to 35% of the general US population. Other staggering statistics reveal that 44% of this population did not graduate from high school compared to 19% of the general population. Downs[41] estimated the cost to a family in the US of having a child with a severe loss to be 1.5 million dollars over the child's lifetime due to reduced productivity and special educational costs. A recent report of 120 children in England[31] (half screened and half not screened) revealed that the health and education costs for children with hearing loss documented in the year before they reached 7 to 9 years of age were significantly higher (cost difference of £9885.7) than the societal costs for a comparison group of 63 normal hearing children.[42]

The importance of a health problem from a population perspective depends not only on the number of individuals affected but also on the extent to which they are affected. One of the factors which has been cited as being linked to the delays of children with permanent hearing loss is age of diagnosis.[7] The delays associated with permanent childhood hearing loss constitute a sufficiently important health issue that population-based screening has been widely adopted in an effort to improve developmental outcomes.

Impact of Screening on Age of Identification

Age of identification of hearing loss is considered to be a critical factor in the development of a child's speech, language, cognitive and psychosocial abilities and is the underlying premise of infant hearing screening initiatives.[43,44] Two major approaches to screening have been employed: 1) population-based screening and 2) targeted screening for infants identified with risk factors for hearing loss. The most recent guidelines for risk indicators for childhood hearing loss are provided by the JCIH.[11] These include neonatal intensive care for more than 48 hours, a history of familial hearing loss and craniofacial anomalies. Studies suggest 40 to 50% of newborns with hearing loss manifest a risk indicator [45,46] Again, the CWGCH review notes that there is considerable variation in the prevalence estimates for children with risk factors. This is partly accounted for by the changes in proposed risk factors over the years. The review concluded that there is fair evidence that the prevalence of congenital permanent hearing loss is about 2 to 3% in current at-risk infants.

Studies indicate that the goal of diagnosis of hearing loss for all affected infants by 3 months of age as recommended by the JCIH[11] does not appear to be achievable using a targeted high-risk approach to screening.[10] In the absence of systematic screening, late identification is common for the 40-50% of babies presenting with no risk factors.[5,20,46]

A critical review of the age of diagnosis of infant hearing loss with and without systematic screening programs was conducted by the CWGCH. The review identified 23 relevant publications from 1990 to 2002 when the following inclusion criteria were applied: subject inclusion and exclusion criteria clearly defined, risk factors for hearing loss clearly defined and coverage of a wide population base. These studies showed that severity of hearing loss was the main determinant of age of identification when systematic screening was not available. In the absence of systematic screening, children with lesser degrees of hearing loss were diagnosed well beyond 12 months of age, ranging from 20 to 42 months.

Publications retrieved since the CWGCH review report findings consistent with those summarized in the critical review. In a 5 year retrospective study of a US hearing screening program, a mean age of diagnosis of 3.9 months and mean age at intervention of 6.1 months were achieved.[30] Serville and colleagues[47] reported on a sample of 88 children from a Belgium center born between 1990 and 1999. In a center without universal screening, the median age of diagnosis of the population was 45 months, 27 months and 13 months for moderate, severe and profound hearing losses respectively. However, in this sample, progressive hearing loss appeared to account for delayed diagnosis in 14 patients. Consistent with previous literature, a recent study[28] in Australia, where systematic screening was not in practice, found age of diagnosis to be inversely correlated to severity of the hearing impairment. In New Zealand, where an at-risk screening approach has been in place,[48] investigators called for a UNHS service, citing statistics of an overall average age of diagnosis of hearing loss of 35 months for the year 2002. Finally, in a parental survey, the

average age of diagnosis reported by parents, for 77 children (birth cohort 1991-1996) with severe to profound hearing loss was 14.58 months with 55 families reporting a delay of more than 5 months from parental suspicion to diagnosis.[49]

The contribution of UNHS has been directly assessed through one controlled trial in the UK. The screened population consisted of 53,781 infants born in four hospitals in Wessex region from 1993 to 1996. This three year study phase was divided into four periods (four to six months in duration) where neonatal screening was provided and four periods with no screening. In periods with UNHS, detection of hearing loss by 6 months was achieved in 93% of affected babies compared with 17% of babies born in periods without screening.[31] In a recent follow-up study to the Wessex 1993-1996 controlled trial, Kennedy and colleagues reviewed case records of all children identified with hearing loss to evaluate the effectiveness of UNHS in identifying true cases of permanent hearing loss.[29] During periods with screening, 74% of children were referred before age 6 months compared to 31% in periods without screening.

The CWGCH review reported the median age of diagnosis with UNHS to lie between 2 and 3 months regardless of degree of hearing loss. Consistent with the CWGCH report, the USPSTF review also concluded that neonatal screening effectively increases the number of children diagnosed before 6 months of age. In summary, both reviews and the evidence since 2002 provide support that UNHS can effectively reduce the age of diagnosis of hearing loss regardless of the severity of hearing loss. In contrast, studies have consistently

documented differences in age of diagnosis by severity of hearing loss when systematic screening was not available.[5,28,47] Accordingly, population screening may have the greatest impact on outcomes in children with mild to moderate degrees of hearing loss and identification in infancy may actually prevent delay in this population of children.

Taken together the reviews and subsequent findings indicate that there is good evidence that the technological capability exists to reliably screen hearing in infancy. However studies conducted in the US suggest that the potential benefits of screening have been reduced due to issues with poor rates of follow-up for audiological evaluation and commencement of intervention.[50,51] These issues were identified in the 2001 survey of state program coordinators in the US by the National Center for Hearing Assessment and Management.[52] Ensuring that babies referred from screening programs receive timely diagnostic and intervention services remains an important goal according to this survey report. Only an estimated 56% of infants referred from screening received diagnostic assessment before the recommended age of three months. Only an estimated 53% of infants identified with hearing loss entered intervention programs by the recommended six months of age. Furthermore, tracking of 23% to 52% of referred babies was incomplete. Although 97% of parents surveyed rated newborn screening as very important, 23% of them did not return for follow-up testing according to one study.[53] A study investigating screening rates, underscored the impact of family health insurance and other environmental factors on infants' access to screening or re-screening.[54] A recent surveillance report from the Centers for Disease Control and Prevention (CDC)[55] raised alarms that newborn screening programs

in the US are not meeting the goals outlined by the JCIH. The CDC reported that almost half of the infants referred for audiologic assessment in 2001 did not receive an evaluation and approximately one-third of the 1,354 infants identified with a hearing loss in 25 states which reported benchmark data, were not enrolled in intervention programs. Of the 879 who were in intervention, only 626 (71%) were enrolled by 6 months. A review of trends in UNHS programs in the US, identified efficient tracking of children needing follow-up, medical management, coordination and cohesiveness of the follow-up care team as important areas for improvement.[56]

Adverse Effects of Screening

Prior to the widespread implementation of UNHS, concerns were raised regarding the deleterious effects on families due to increased anxiety. Bess and Paradise[57] presented one of the strongest and most influential arguments against UNHS in the 1990s based on several concerns, one of which was the potential for parental anxiety caused by false-positive test results. Using current technology and two-stage screens, approximately 4% of screened babies will be referred for follow-up diagnostic testing.[1] These referral rates and current yields indicate that population screening will identify one case of permanent childhood hearing impairment for every 40 infants referred for follow-up diagnostic testing. Early reports from families of screened children did not confirm a high level of anxiety but rather showed that parents highly valued screening.[58-60] One study using preference techniques, which investigated preferences for universal versus targeted screening in mothers,

pediatricians and hearing specialists, found that all three groups preferred universal screening as the route to early identification of hearing loss.[61]

More recent studies are consistent with these earlier reports and show no harmful effects from hearing screening.[62-64] A study of 90 mothers in Austria[65] reported that there was an association between the information provided to mothers and their attitude towards screening. A recent UK study, examined the effect of newborn hearing screening on maternal anxiety, and found that anxiety levels increased as the number of screening tests (and eventual referral to audiology) increased.[66] The study reported that knowledge of the meaning of a refer result was associated with lower anxiety for mothers whose babies were referred for an audiologic assessment after three screening tests. Similarly, a qualitative study with parents experiencing the screening process in the first phase of the UK evaluation series, reported that parents desired clear information on why a screening result was inconclusive.[67] This study highlighted the need to verify parents' understanding of the screening process. Taken together, the evidence from these studies suggests that overall, parents have a positive attitude toward screening. There is no evidence from studies of parents of hearing or hearing-impaired children to suggest that there are lasting negative effects from screening or that the process interferes with parent-child bonding or later developmental outcomes.

Optimal Period for Language Learning

Newborn screening has gained considerable support from advocates of the theory of optimal or critical periods of language learning. A review of numerous studies over the past 40 years concluded that early sensory experience plays an important role in the organization of sensory information such as language.[68] The authors reported findings of changes in the auditory pathways that suggest sound deprivation during the early months of life affects neurobiological development in both the peripheral auditory pathways and the cortical areas of the brain. Responses obtained with auditory evoked potentials in children with early onset deafness whose hearing was partially restored through cochlear implants were reported to be consistent with immature cortical function.[69] Likewise, current research from cochlear implant studies in children using cortical responses to demonstrate age at implantation as a critical marker in the development of auditory skills, has provided powerful evidence for neuroplasticity.[70,71] Animal studies have provided further evidence that the absence of sound in early infancy can result in significant structural changes in the auditory system.[72] Population screening is predicated on the belief that introducing early auditory stimulation can at least partly reduce the effects of auditory deprivation. This growing body of research provides support for reducing the effects of auditory deprivation on language development in children with hearing loss through early identification and remediation.

In view of the evidence for optimal periods of learning, early auditory stimulation, independent of communication development, should be considered as a potential benefit of early diagnosis of hearing loss. Early access to hearing through early detection was

supported by a critical review (as part of the CWCGH initiative) of the effect of hearing aids on auditory performance[10]. The studies included in the review reported auditory-specific outcomes on children age 5 or older. Studies were limited to this age due to the limitations of the types of measures currently available for the measurement of auditory benefit. This review concluded that there is evidence that properly fitted hearing aids can improve the auditory abilities of children with hearing impairment. Therefore, access to hearing through amplification and/or cochlear implants may be an important positive outcome of early hearing and communication development programs.

Effectiveness of UNHS in Improving Longer-term Communication Outcomes

For many decision-makers, the real success of UNHS depends on the extent to which it is possible to prevent or reduce communication delays in children with hearing loss. While there is evidence that the technological capability exists to screen and diagnose hearing loss as early as 3 months of age, the USPSTF review concluded that there is insufficient evidence supporting the effectiveness of screening in improving communication outcomes.[73] The systematic review commissioned by the USPSTF included 8 studies published between 1995 and 2000 which addressed the impact of early identification on outcomes in children with hearing loss.[6] In a series of studies of children diagnosed through the Colorado screening and intervention program, age of diagnosis emerged as the primary predictor of language outcome. Children identified by 6 months of age were found to have language skills within the normal developmental spectrum.[8] Age at intervention as a predictor of communication development has been corroborated by other studies although less extensive

data are available for children for whom intervention was initiated by 6 months of age.[44,74] However, according to the USPSTF, the available evidence does not provide sufficient support for the effectiveness of early identification of hearing loss in improving speech and language outcomes. This conclusion was reached as a result of the systematic review which judged the quality of the studies as fair to poor on the basis that they included primarily convenience samples and limited information on subject selection and attrition.[6]

Since these reviews, one major controlled study has contributed to the evidence on the impact of early identification of hearing loss[38] by documenting that early identification positively affects communication outcomes. The study conducted in eight districts in the UK (Wessex and Greater London regions) followed 120 children born from 1992-97 and a control group of 63 children matched for age and place of birth. Children with hearing impairment identified by 9 months of age showed significantly better receptive and expressive language abilities at a mean age of 7.9 years than later identified children. Children exposed to newborn hearing screening demonstrated significantly better receptive but not expressive language scores. However, early hearing loss identification or exposure to screening did not affect speech production scores. Although this controlled study provides a higher level of evidence than previous reports noted above, it is important to highlight that there have been significant technological changes since the early developmental years of the children in the study. For example, cochlear implantation has become the standard of care for children with significant hearing losses which did not appear to be the case in this study. In addition, universal screening has been coupled with

improvements in other areas such as hearing aid technology and management and possibly improved intervention opportunities unrelated to age of diagnosis. A related study showed that the economic costs of medical and special education for children with hearing loss in regions with UNHS were not significantly different (although 15% lower) than those in regions without screening. Education costs in the preceding year of the study were calculated to be on average 22% lower for children born in regions with UNHS, however, this benefit was partially offset by a 12% increase in non-educational costs in the UNHS group.[42]

Other relevant research includes two additional studies which did not specifically investigate UNHS but examined age of identification of hearing loss as a predictor of outcomes. One report described in an earlier section, involved a population-based study of oral communication and reading outcomes for 89 children at 7 to 8 years of age in the state of Victoria,[37,39] Australia. The results indicated that language scores were negatively correlated with severity of hearing loss and with lower socio-economic status although the latter was a weaker relationship. As noted by the investigators, few children in this study were identified by age 6 months, however, no trends related to age of identification or intervention of hearing impairment were observed. A third study examined the effects of early auditory experience on spoken language in 76 children with severe to profound hearing loss who had received cochlear implants. In this group of children assessed at 3.5 years of age, pre-implant intervention (i.e., age of identification) was not related to outcomes. However, age at cochlear implantation and degree of hearing impairment (pre-implant pure-

tone-average) were significant predictors of oral language outcomes. This study suggests that age of identification may not have as great an impact on children with severe to profound hearing impairment because cochlear implantation is typically not provided before 12 months of age. Other reasons for delayed implantation may include uncertainty around the boundaries of hearing loss which are more amenable to treatment with a cochlear implant than a hearing aid.

These new studies, which were added to the evidence-base since the reviews were carried out, generated different results with respect to the effect of age of identification of hearing loss.[37,38] Both the USPSTF and the CWGCH reviews as well as subsequent reports[6,9,10,75] highlighted the need for well-controlled studies to document the effectiveness of population hearing screening in improving speech and language outcomes. However, researchers also acknowledge the complexity of conducting such studies given the multiple factors which impact developmental outcomes in children with hearing loss.[9] In summary, the USPSTF[76] has stated that it cannot recommend for or against universal screening given the current state of knowledge. Notwithstanding the limitations in the current science-base the CWGCH has recommended that Canada adopt population screening on the basis that EHCD leads to improved hearing which in turn should be considered the primary benefit of screening.[10]

Type of Intervention and Outcome

A myriad of factors other than age of diagnosis are thought to impact the communication outcomes in children with hearing loss and the task of isolating age from other family, child

and contextual factors is a daunting one. One factor which has attracted considerable attention is the type of intervention provided for a child with hearing loss.

There is no "treatment" for permanent childhood hearing loss but various intervention or educational management programs are aimed at improving function and participation in society. Historically, intervention approaches for children with hearing loss range from those aimed at teaching children to communicate using residual hearing and speech to those focused on developing communication through sign language. In this sense, their theoretical basis may be as much about whether deafness should be treated as pathological or as a cultural difference as about the efficacy of different intervention strategies. Various intervention strategies have evolved from these philosophies under multiple labels such as auditory-verbal, auditory-oral, oral, cued speech, total communication, dual communication and American Sign Language (ASL).[13]

Recognizing the wide diversity of clinical opinion about the contribution of various intervention approaches, the CWGCH supported a systematic review to inform their recommendations for intervention in the context of population-based screening programs. The purpose of the systematic review was to synthesize the evidence on the effectiveness of four current intervention approaches for the management of childhood hearing loss: auditory-verbal, auditory-oral, total communication and ASL.[13] Eligibility criteria for the review were studies of any level of evidence above opinion, on children with permanent congenital hearing loss who received one of the four types of intervention approaches. The

review focused on communication related outcomes. Of the 194 papers meeting the inclusion criteria, only one randomized controlled trial (RCT) was identified and the majority of the remaining publications concerned cohort designs with or without some form of control group.

Due to important gaps in the reporting of relevant data (e.g., sample sizes, age at intervention, duration of intervention) and extreme clinical heterogeneity, the review concluded that neither a meta-analysis nor a full qualitative synthesis was possible. This review highlights the many variables in intervention studies such as parental involvement, therapists' skill, child's cognitive abilities coupled with age of intervention and degree of hearing loss. Accordingly, the review underscores some of the complexities of designing well-controlled studies to examine the effectiveness of intervention approaches.

A second important factor which confounds the effect of early identification on outcomes is the evolution in hearing technology (hearing aids and cochlear implants). In particular, the advent of pediatric cochlear implantation as a routine treatment for severe to profound hearing loss has contributed significantly to the improvement in outcomes in children with significant hearing impairment.[34,36] Studies which have compared oral and total communication (speech and sign language combined) rehabilitation methods point to superior outcomes for children with cochlear implants in oral communication programs.[36,77,78]

Improving the science-base for the effectiveness of various interventions has been highlighted by UK researchers as an important objective to pursue in the context of UNHS programs.[32,79] Our team is currently updating the Health Canada systematic review described above that was commissioned by the CWGCH. Despite the proliferation of studies on intervention for children with hearing disorders, our preliminary review of published articles to September 2005 suggests that research has not further illuminated the question of which intervention method is most effective for children with hearing impairment (Fitzpatrick et al., unpublished data).

Service Delivery Models and Outcome

During the early development of UNHS initiatives, attention was focused on the technical aspects of electro-physiologic screening tests and on sensitivity and specificity of test protocols. However, it has become increasingly clear that screening to improve infant development cannot be detached from intervention services. Therefore, screening is increasingly viewed as a procedure which must be anchored in a context of clinical support services for affected children and families. The effectiveness of newborn hearing screening is intricately linked to the subsequent intervention process which includes audiologic assessment and rehabilitation for confirmed hearing loss.[12]

Gains in communication and social development are contingent on appropriate and effective early intervention.[43] In describing the evaluation of the first stage of the national UNHS initiative in the UK, Young and Tattersall[80] emphasized that evaluating screening

necessarily involves evaluating the early intervention process. The decision to adopt the term Early Detection and Hearing Intervention (EDHI) and the recommendation in Canada to adopt Early Hearing and Communication Development (EHCD) acknowledges the liaison between all components of a system of care with screening as the first step in the care path. Of particular interest to this thesis is the contribution of service delivery models, where screening comprises one component of a total care model for children with hearing loss and their families.

A health technology assessment conducted in the UK in preparation for UNHS identified the lack of coordination within health and between health and education services as presenting a significant barrier to quality of service provision and outcomes for children with hearing loss.[79] As the UK embarked on a national screening program, challenges in service provision were revealed through a national study of services for preschool age children with hearing loss.[81] This study reported that parents identified well-coordinated and high quality services as fundamental to "family friendly" care. In particular, multi-disciplinary work and agency coordination emerged as barriers of particular concern in current service provision. These findings were corroborated by the administration of two indices of service quality and interviews with pediatric audiology services in the UK prior to implementation of the screening program. The findings revealed great variability in the current audiology practices and a lack of understanding of the roles of various agencies and providers.[82,83] In the UK, the evaluation of the first phase of the newborn hearing screening services has also contributed literature on parents' views of their experiences with the screening program.

Professional communication and manner[67] were reported to be the most significant predictors of parents' perspectives of their experiences during the diagnostic process. In both the UK and the US, the lack of trained professionals has been described as a barrier to the timely and effective delivery of intervention services.[81,84]

The implementation of newborn hearing screening represents a paradigm shift where services are moving from a model driven by experts to more family-oriented approaches with greater inclusion of parents in decision-making.[85] A study involving surveys and interviews with families in North Wales,[85] identified a need to focus on broader social and language services for hearing-impaired children and their families. The experiences of 82 parents of children identified early through at-risk neonatal hearing screening services in Australia were examined through a qualitative analysis of parents' written comments.[86] Important themes for service delivery were parents' strong need for support at the time of diagnosis, support during the sometimes lengthy delay before confirmation of the hearing loss and fitting of hearing aids, and communication difficulties with providers. The period immediately following the diagnosis of hearing loss has been reported to be highly stressful for families.[87]

Universal hearing screening has also fuelled an interest in family-centred care in the US. Studies using surveys and qualitative interviews with parents have identified several needs: awareness of the presence of hearing loss as soon as possible, unbiased information about all communication options, support groups, and practical support with hearing aids.[88,89]

Recent work suggests that a family-centred care model reduces stress, improves follow-through at all stages of the process from screening to intervention and results in better outcomes.[16,90] Despite efforts directed at streamlining services subsequent to newborn screening, families continue to report multiple barriers to timely diagnostic and rehabilitative care.[49,91]

Taken together, these studies suggest that the growth in infant hearing screening research and program implementation has encouraged scrutiny of the practice of intervention services following hearing loss identification. Pairing UNHS with quality early intervention in a family friendly context has been described as one of the most important challenges to the success of newborn hearing screening initiatives.[92] With the advent of UNHS, interest in appropriate follow-through services for children with hearing loss is growing. A review of parent perspective data pointed out that parents of children with hearing loss have not been well represented at the decision-making level of programs and services.[93] In a study of outcomes in early and late identified children, parental involvement was identified as an important predictor of communication outcomes.[44]

There is some evidence from other areas of health care to support that certain aspects of the process of receiving care, in addition to the actual intervention outcomes, are important for individuals.[94-96] There is also evidence from pediatric rehabilitation to suggest that patient satisfaction is related to both the actual care process (i.e., technical competence and quality of care) and organizational aspects of the service delivery model.[97]

In summary, the growth of UNHS has been coupled with a growing interest in improving subsequent services for children with hearing loss. Furthermore, interest in consumers' views of appropriate methods of intervention and service delivery models has been intensified because of a cultural/societal interest in giving high priority to informed decision-making and a realization that health services itself may be an important determinant of health outcomes. To our knowledge, no studies of screening or intervention needs have been published in the Canadian context despite the recent implementation of a provincial wide UNHS in Ontario in 2002 and more recent program developments in other provinces and territories. No studies have attempted to examine patients' preferences for the various attributes of health services associated with the screening, diagnosis and management of a child with hearing loss.

Summary of State of Knowledge

Population health hearing screening has been justified on the basis of the prevalence of congenital hearing loss, the devastating effects on the child and family and the premise that early detection leads to improved short and long-term developmental outcomes. To date, evaluations of the benefits of UNHS have focused primarily on whether early diagnosis of hearing leads to improved communication outcomes during the preschool years and beyond. However, the wide variability in results in the literature, the paucity of quality studies and the complexity of conducting such studies, suggest that traditional measures of

communication benefit may not provide the information required to guide decisions about the development of infant hearing programs, at least not in the short-term.

Both the CWGCH and USPSTF reports concluded that newborn screening leads to earlier diagnosis and therefore earlier hearing. However, they identified the need for an examination of other benefits of screening and early intervention for children and their families. They also noted that there is insufficient evidence to draw conclusions about any potential process outcomes or other benefits for families resulting from early diagnosis and intervention. The gains associated with early intervention from the perspective of parents may differ considerably from the perspective of service providers and decision-makers. Accordingly, this thesis privileged parent perspectives in examining the benefits and needs related to early identification of hearing loss. In addition to examining outcomes of early identification initiatives through traditional performance measures, the purpose of the research was to investigate parents' perceptions of benefits, and to identify and quantify their preferences for attributes of care following diagnosis of their child's hearing loss.

Conceptual Framework

Hearing screening is the first step in a system of care encompassing hearing and communication development. The work undertaken in this thesis was guided by a conceptual framework based on a comprehensive review of the literature in infant hearing and the determinants of population health. The conceptual framework was drawn from the World Health Organization's (WHO) model of human functioning and disability.[15] This

International Classification of Functioning, Disability, and Health (ICF) model differs from previous models of disability in that it embraces a new paradigm where health and well-being are seen as an interaction between the individual and his/her environment.

Consistent with a population health perspective, the framework for this research, guided by the ICF model, emphasized contextual factors as contributing to the well-being of the child with hearing loss. The framework in Figure 1 is built on several key pillars which formed the theoretical basis for this research. In this model, there are desired developmental outcomes for the child such as hearing, communication, and social skills targeted through population screening as well as process and quality of life outcomes for the family, all of which eventually lead to fuller participation in society. Drawing on this broader conception of health outcomes, these outcomes are envisioned as a complex interplay between the hearing impairment, child characteristics, family characteristics and environmental or contextual factors. The framework suggests that in addition to reading from left to right, that is, from body structures/function to intervention outcomes and participation, it is important to conceptualize other determinants of outcome that are acting at the child, family and community levels. In the ICF model these are identified as environmental and child factors and expanded in this framework to include family factors. These factors may have implications for the effectiveness of early identification of hearing loss in that certain contexts may provide a better opportunity for positive outcomes from early hearing and communication development programs. The model recognizes the presence of an intricate interaction of causal factors shaping the developmental outcomes in infant hearing loss. The

model represents a starting point for reflecting on the factors that influence the outcomes in childhood hearing loss.

This framework formed the underpinnings of this research project which examined the benefits of neonatal screening and contextual factors through three interrelated inquiries. In particular, as highlighted in the framework, the work in this project centered on two broad domains: 1) the benefits of early identification including traditional communication outcomes and other benefits perceived by parents, and 2) aspects of the care model that are important to parents. Specifically, the first inquiry of this project addressed typical speech and language outcome measures and their contribution to understanding the benefits of early diagnosis of hearing loss. The second inquiry of the research was directed at identifying parents' perspectives of benefits and their needs for support. Finally the third inquiry concerned the characteristics of service delivery models and established parent preferences for these attributes. A focus on the complex interaction between the child with hearing loss and numerous contextual factors provides a more comprehensive perspective of health and well-being for children with hearing loss and their families.

Conceptual Framework for Infant Hearing Loss

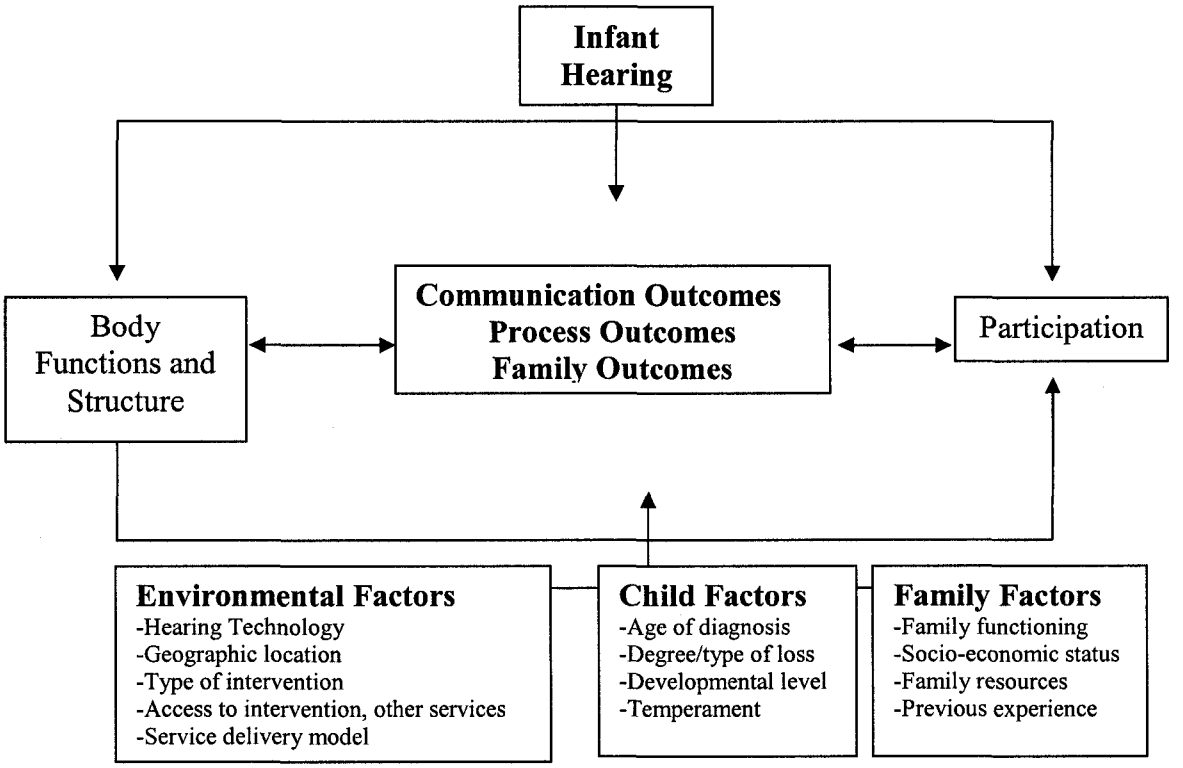

Figure 1. Conceptual framework of factors that influence outcomes in children with hearing loss based on the International Classification of Functioning, Disability, and Health[15]

General Overview of Thesis

Given the proliferation in the number of UNHS programs both in Canada and other countries coupled with the lack of strong evidence for their impact on communication outcomes, there is a need to continue developing the science-base to guide further decisions in early hearing detection and intervention models. This doctoral research incorporated a mixed methodology using both quantitative and qualitative research methods to investigate the process of early identification, intervention and outcomes for young children with hearing loss and their families. The study was designed as a series of three inquiries involving: 1) an examination of the benefits of early identification through traditional communication outcome measures, 2) an exploration of parents' perspectives on the benefits of early identification as well as their needs following identification and, 3) a conjoint analysis to quantify parents' preferences for services. Table 1 provides a précis of the objectives, research questions, methodology and chapters in this thesis associated with each of the three inquiries. The research involved four clinical sites in Ontario. The project received ethics approval from all participating institutions and the University of Ottawa (Appendix 2) and written informed consent was obtained from all participants (Appendix 3).

The methodology and findings for the study are presented as a series of four articles which constitute Chapters 2 through 5 of this dissertation. Detailed methods and results for each inquiry are presented through these chapters, each of which is a stand-alone article, formatted for the appropriate peer-reviewed journal. All manuscripts are under review or

accepted by a scholarly journal (Appendix 4). A brief introduction to each chapter is presented below.

Chapter 2 presents the results of a multi-center prospective study undertaken with a team of researchers at the University of Ottawa, University of Toronto and the Children's Hospital of Eastern Ontario Research Institute to investigate the impact of early identification of hearing loss on several developmental outcomes. The results of communication development in 65 children who had reached the 3 year study point were analyzed as part of this thesis work in order to explore the contribution of traditional outcomes to an understanding of the benefits of newborn hearing screening (see appendices 5 and 6 for study data forms).

Chapter 3 presents the findings of qualitative research (inquiry 2) which explored the benefits of early identification through interviews with 17 parents in four clinical programs in Ontario. A qualitative research approach was adopted for this inquiry to gain a rich and comprehensive understanding of parents' views of outcomes related to early detection of hearing loss. Ten parents of children diagnosed with hearing loss through conventional referral routes and seven parents of children diagnosed through screening shared their experiences of the diagnostic process and their views on the positive or negative aspects of newborn screening (see appendix 7 for interview guide).

Overall purpose: To understand the benefits of infant hearing screening and to identify the essential components of family-centred care for children with hearing loss and their families.					
Inquiry	**Objective**	**Research Questions**	**Design**	**Method**	**Thesis Chapter**
One	To explore whether traditional methods of measuring communication outcomes can inform the discourse on the benefits of UNHS.	Is there an association between age of diagnosis of hearing loss and communication outcomes?	Prospective	*Quantitative* Direct measurements	Chapter 2
Two	1) To explore how families of children with hearing loss perceive their experience leading to the diagnosis of hearing loss and the impact on quality of life in the early stages of identifying and managing hearing loss.	1.1) What are the benefits of early diagnosis from the parents' perspectives?	Constant-comparative (Drawn from grounded theory)	*Qualitative* Semi-structured interviews	Chapter 3
	2) To identify the important attributes of assessment and intervention services for families following diagnosis of a child's hearing loss.	2.1) What are parents' needs when a hearing loss is diagnosed? 2.2) From the parents' perspective, what attributes of service provision will best meet their needs?			Chapter 4
Three	To identify and evaluate parent preferences for various attributes of service following diagnosis of hearing loss.	1) What are parents' preferences for the attributes of service delivery? 2) Do the preferences vary with the characteristics of the participants: hearing device, screening status, and region?	Cross-sectional	*Quantitative* Survey	Chapter 5
Integrated Discussion and Conclusions					Chapter 6

Table 1. Description of inquiries, objectives and methodologies

The qualitative interviews conducted in inquiry 2 also generated an in-depth understanding of families' needs which formed the basis of a third article. Chapter 4 presents parents' perspectives of their needs in caring for a child with hearing loss and their views of the important characteristics of care models. The 17 parents shared their observations of the strengths and gaps in the system of care.

The information from the qualitative interviews informed the development of a survey (Appendix 8) to quantify parents' preferences for various attributes of services. Chapter 5 presents the results of inquiry 3 which adopted a quantitative methodology in applying conjoint analysis, as a novel technique in infant hearing research to examine parents' preferences for the characteristics of service models.

The four article-based chapters are followed by an integrated discussion in Chapter 6 to summarize the key findings of the thesis, the contribution to population health and finally to reflect on future research needs in infant hearing science. Finally, the operational aspects of the project including the contributions of the research team and knowledge translation activities are provided.

In summary, as a population health intervention, infant hearing screening has thrust childhood hearing loss under the public health lens. This visibility brings with it a responsibility to ensure that decisions related to hearing screening and subsequent management of the disorder are grounded in the best available evidence. Ultimately, this

thesis was designed to contribute to science by gaining insights into measurable outcomes, as well as the perceived benefits, needs and values of those who use and are most affected by population-based infant hearing screening.

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CHAPTER 2

THE IMPACT OF NEWBORN HEARING SCREENING ON COMMUNICATION DEVELOPMENT[1]

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The Impact of Newborn Hearing Screening on Communication Development

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Running head: Infant hearing screening

Abstract

Objective: Universal newborn hearing screening has become standard practice in many countries. The primary goal of this study was to assess the impact of early identification of permanent childhood hearing loss on oral communication development.

Setting: Participants were recruited from three clinical programs in two cities in the province of Ontario, Canada. This study was undertaken shortly after the introduction of a new provincial population screening initiative; a population-based registry was not in place at the time of the study. All children were enrolled in rehabilitation programs focused on oral language development.

Methods: In this multi-center observational study, 65 children under the age of 5 years with onset of hearing loss before 6 months of age (26 identified through systematic newborn screening and 39 without screening) were assessed with an extensive battery of child and parent-administered speech and language measures. The degree of hearing loss ranged from mild to profound with 22 children in the mild, moderate and moderately severe categories and 43 in the severe and profound categories. Data are reported for the three-year study period.

Results: The screened group of children was identified at a mean age of 6.1 (SD 3.7) months and children referred from sources other than newborn screening were diagnosed at a mean age of 19.3 (SD 11.5) months. Assessment of oral communication development showed no significant difference between the screened and unscreened groups. The communication outcomes for children identified before 12 months of age did not differ from those of later identified children.

Conclusions: Systematic screening of newborn hearing results in earlier identification and intervention for children with permanent hearing loss. The findings suggest that earlier identification may not translate to improvement in short-term language outcomes in all clinicalcontexts.

Background

Newborn hearing screening has become a major theme in pediatric audiology in recent years. Population screening has been widely embraced on the basis that permanent childhood hearing loss is a lifelong condition with significant personal and societal costs.[1] Permanent hearing loss is a relatively common disorder with prevalence estimates ranging from 1 to 3 in 1000 children either affected congenitally or in the first years of life.[2-5] The variation in prevalence rates reported in the literature can be explained by the complexity and uncertainty in reporting infant hearing loss figures. There is considerable variation in studies related to how hearing impairment is defined in relation to severity, frequency range, type of impairment as well as whether the hearing disorder is unilateral or bilateral.[6] Children with hearing impairments are at significant risk for delayed speech and language and subsequently poor academic and social development; consequently society's costs for providing health and educational care for these children is substantial.[1,7-9] In the past two decades, technological advancements in electrophysiological testing have made newborn hearing screening reliable and efficient. This has led to a focus on the development of strategies for the early identification of hearing loss in children. Significant steps have been made in increasing the awareness of pediatric hearing loss as a public health issue such that population based newborn hearing screening has become the new standard in infant hearing care. In fact, universal newborn hearing screening (UNHS) has moved from a desirable concept to standard practice in many countries.

A powerful argument for the early detection and intervention of hearing loss has been the potential to reduce the negative effects of hearing loss and to promote optimal development over time.[10] In particular, delayed communication development as the primary negative effect of permanent hearing loss has been investigated. The underlying rationale for population hearing screening is that identification of hearing loss can be achieved in the first 3 to 6 months of life, therefore resulting in early access to intervention. However, several reports indicate that the average age of diagnosis achieved through typical referral practices exceeds 12 months even for children with profound hearing losses and can be as late as 20 to 42 months for children who exhibit lesser degrees of hearing loss.[7,11,12] When targeted high-risk screening is in place, late identification is common for the 40 to 50% of babies presenting with no risk factors for hearing loss.[11]

There is a considerable body of research in infant communication development and brain plasticity supporting that early sensory experience plays an important role in the organization of sensory information such as language.[13,14] Late diagnosis of permanent hearing loss may place children outside the optimal learning period for language acquisition. However, a specific age threshold for such an optimal period has not been determined, leading different investigators to examine different cut-offs for early and late identification. Unquestionably, early diagnosis should lead to early access to hearing technology and specialized intervention programs. Arguably, this may result in better communication development, and ultimately more positive academic, social and employment outcomes.

Evidence has emerged in the past 10 years to demonstrate that children identified before 6 months of age achieve superior results in language and other developmental areas compared to later identified children.[15,16] Accordingly, UNHS has received considerable support from key organizations such as the American Academy of Pediatrics and the Joint Committee on Infant Hearing.[17,18] In particular, a series of studies from Colorado in the past decade concluded that the early identification and management of hearing loss (during the first 6 months of life) significantly improves several dimensions of communication development as well as social-emotional development.[15,19,20] In summary, these reports suggest that children identified by 6 months of age, independent of other factors (e.g., severity of hearing loss, socio-economic strata, type of intervention programs) achieved higher levels of language skills than children identified after 6 months.[16] Further reinforcement of the positive effects of early identification on communication outcomes has been provided by other studies where intervention age exceeded 6 months of age.[21,22] While a number of these studies have been highly cited as providing good evidence for the effectiveness of early identification and management, the United States Preventive Services Task Force (USPSTF) judged the evidence on enhanced language development to be inconclusive based on a systematic review of the literature.[23] The USPSTF concluded there was insufficient evidence to endorse population screening and highlighted the need for more studies investigating the impact of universal hearing screening.

More recently, a population-based cohort study in the state of Victoria, Australia, which reported outcomes on 88 children at age 7-8 years, showed strikingly different results than

the earlier studies referred to above.[9,24] Age at diagnosis did not correlate with any speech, language or reading outcome measure whereas severity of hearing impairment was strongly related to language scores. Furthermore, the 11 children with hearing loss identified before the age of 6 months did not demonstrate superior speech, language and reading outcomes to the children identified between 6 and 12 months or the 59 children identified after 12 months. Similarly, in another recent study, age at identification and amplification were unrelated to spoken language outcomes at age 3.5 years in a sample of 76 children with cochlear implants. However, younger age at implantation was associated with improved spoken language skills.[25]

In contrast, recent results from a controlled study of 120 children in England demonstrated improved verbal speech and language skills at age 7 to 10 years in children diagnosed by 9 months of age compared to later identified children.[26] In this study, children with hearing loss detected by 9 months of age (or during periods of UNHS), demonstrated better receptive language scores but not speech scores at an average age of 7.9 years.

Given the divergent information on the impact of early diagnosis and the current possibilities offered by early identification and new sensory technologies, there is growing interest in the outcomes of early and later identified children. The aim of the present study was to investigate the effects of systematic screening (children identified through universal or targeted high-risk screening) and early diagnosis of hearing loss. The study was in part motivated by the introduction of a province-wide newborn hearing screening program in

2002 in Ontario, Canada, the first provincially mandated screening initiative in the country. Detailed descriptions of the program have been provided by Hyde and colleagues.[27] Prior to the province-wide screening program, some infants admitted to neonatal intensive care units were screened as part of local initiatives.[11]

This study was undertaken as the provincial program was being implemented, however the intent of the present study was not to evaluate the provincial infant hearing program but to contribute to the science-base by examining the development of children identified with and without screening.

The full research project was conducted to investigate multi-dimensional aspects of development related to screening and early intervention in children with hearing loss. In this paper, we focus on communication skills during the preschool years (to age 5) in children identified with hearing loss through systematic screening and compare the outcomes to children who were not identified through screening. This article summarizes data related to communication outcomes at the end of the three-year study period. In particular, we present three year outcome data from the study to address the following question regarding the benefits of infant hearing screening: "Is there an association between age of identification of hearing loss and communication outcomes?" Consistent with much of the literature on the early identification of children with hearing loss, we hypothesized that children whose hearing disorder was identified early would have superior outcomes compared to children whose loss was identified later.

Method

Design

This study incorporated primarily a prospective cohort study design examining development in children identified with hearing loss through universal or targeted screening and children whose hearing loss was identified without screening through traditional referral practices. The screened group included an inception cohort from a UNHS screening initiative as well as children identified through targeted high-risk screening in neonatal intensive care units prior to the implementation of UNHS in 2002. The referred group consisted of children identified prior to the implementation of UNHS or not screened as part of the UNHS or targeted screening programs. All eligible children were diagnosed between 1999 and 2004 (birth cohort 1998 to 2003).

Data for this multi-center study were collected across three study sites in two different cities. Data collection began at the Ottawa site in October 2002 and at the Toronto sites in 2004. The two study areas represented a population of approximately 5 million people. Both cities have major pediatric hospitals providing universal screening, follow-up, early diagnosis and management. Rehabilitation services are provided in the hospitals or through other clinical, educational, or community settings.

Participants

The study population was drawn from three urban pediatric centers in two cities in the province of Ontario, Canada: Children's Hospital of Eastern Ontario (CHEO) in Ottawa and the Hospital for Sick Children (HSC) and Learning to Listen Foundation (LTLF), both located in Toronto. The CHEO program is the major diagnostic center for the Eastern Ontario region and provides early diagnostic audiology and intervention using an auditory-verbal approach. The HSC is a tertiary care center in Toronto that provides early diagnostic, rehabilitative and therapy services. Children are referred to a variety of rehabilitation programs in the city including the LTLF, an auditory-verbal therapy program for children with hearing loss. Under the guidelines of the provincial infant hearing program, parents are provided with information on all communication development options for children with hearing loss, typically through a parent support worker affiliated with the provincial infant hearing program who meets with families following the diagnosis.[27,28] Prior to 2002, information about rehabilitation options was typically provided by audiologists/therapists in the various centers.

Children were eligible and invited to participate in the study if they met the following inclusion criteria: 1) chronological age less than 5 years, 2) permanent bilateral hearing loss, 3) congenital or early onset loss (before 6 months), 4) consistent use of hearing technology and/or enrolment in an intervention program emphasizing the development of spoken language, 5) intervention services in English, and 6) absence of complex medical and developmental disabilities. Excluded were patients whose developmental condition did not

permit completion of the test protocol. The age at enrollment for the participants therefore varied according to age of diagnosis, and whether hearing loss had been identified prior to the start of the study.

In the absence of a population-based registry of all children, eligible participants in Ottawa were identified through a database maintained at CHEO and through a rigorous chart review at HSC. A total of 74 of 141 children diagnosed with congenital or early onset hearing loss in Ottawa met the study's eligibility criteria. This number is estimated to represent all childhood hearing losses diagnosed in the Eastern Ontario area as the hospital is the only pediatric diagnostic center. The potential participants in Toronto were identified by the study coordinator who applied the study's inclusion criteria to an extensive clinical chart review. Of the 188 children with congenital or early onset hearing loss followed at HSC, 130 met the study criteria. The primary reasons for exclusion of participants at both sites included 1) unilateral or very mild hearing losses without amplification, 2) diagnosed complex disabilities and 3) habilitation in a language other than English. Participants for this study were recruited through clinicians at the three centers. Potential participants completed a brief intake form, which was reviewed by the study coordinator to determine eligibility, applying common inclusion criteria across all sites. The study received full research ethics approval from all three institutional review boards and informed consent was obtained prior to data collection.

Overview of Test Procedures

Following identification of hearing loss, all children and families received intervention through various clinical or educational rehabilitation programs. Only children in programs with a focus on oral communication participated in this research, however, intervention was not controlled in the study. Following extensive pilot testing to determine the most appropriate measures for the age group, a comprehensive developmental assessment battery (comprising auditory, speech-language, general development and cognitive measures) was administered by trained examiners outside the child's clinical program. Children were assessed with this battery of speech-language and other developmental measures beginning at a chronological age of 24 or 36 months age as appropriate for the measure and annually thereafter (36, 48 and 60 months). All language measures were administered according to test protocols (no sign language was used during test administration). Assessments were typically conducted in the home over two to three test sessions. Descriptions of the communication outcome measures analyzed for this paper are provided in the following sections.

1) Direct administration to child: Peabody Picture Vocabulary Test, Third Edition (PPVT-III), the Preschool Language Scale, Fourth Edition (PLS-4) and the Goldman-Fristoe Test of Articulation, Second Edition (GFTA-2). All three speech-language tests administered to the child and reported in this paper have normative data with a standardized score of 100 and a standard deviation of 15.

PPVT-III: This assessment tool is a measure of receptive vocabulary in English.[29] It was developed for use with children aged 2½ to adults aged 90+. A stimulus word is presented to the child who is asked to choose the picture from a four-picture plate in the test book that best represents the stimulus word.

PLS-4: The PLS is a measure of receptive and expressive language development, appropriate for infants and young children ages 2 weeks through 6 years, 11 months.[30] The test has receptive and expressive language tasks, using both pictures and toys as stimuli. The test includes both an Auditory Comprehension and an Expressive Communication subtest.

GFTA-2: The GFTA was developed to provide measures of speech production ability for individuals age 3 to adulthood. The Sound in Words subtest was used in this study to measure articulation in children.[31] In this test, the child is shown a picture and asked to identify it.

2) Parental report: Child Development Inventory (CDI). The CDI is a parent report (for children aged 15 months - 6 years) that provides a profile of the child's development in the areas of receptive language, expressive language, social, self-help, gross motor and fine motor skills.[32] The CDI contains 270 statements that describe developmental skills of children in the first 6.5 years of life that are observable by parents in everyday situations. CDI norms and validity were determined for a community sample of 568 children. The CDI developmental scales correlate closely with age ($r = 0.84$). The receptive and expressive

language subtests are reported as outcomes in this paper. In the absence of another developmental measure at this juncture of the study, the self-help subtest was also used as a covariate in the data analysis. Parents' ratings on the self-help scale were selected rather than the social scale because social development is typically influenced by language skills.

Baseline Characteristics

Other baseline demographic and clinical characteristics were collected directly from parents using questionnaire forms developed for the study. Information known or assumed to influence outcomes such as type of intervention, socio-economic status, and other developmental issues were documented but not controlled in this pragmatic study. In addition, study data related to age of diagnosis, age of amplification and intervention, etiology, status of hearing loss and other health conditions (e.g., time in Neonatal Intensive Care Unit – NICU) were extracted from the clinical charts.

Data Analysis

The primary outcome analyzed for the study was communication development as measured by the speech and language measures described above. All analyses were completed using the Statistical Package for the Social Sciences (Version 13.0). Differences on the main speech and language outcomes between the two groups (screened versus unscreened) were compared using independent samples *t*-tests. The outcomes are reported as standard scores for the PPVT, PLS and GFTA and as Language Quotients for the CDI. Statistical significance was accepted at the P = 0.05 level, all P values are 2-tailed; 95% confidence

intervals were also calculated for means or medians where appropriate. Differences between patient characteristics for the two groups (screened and unscreened) were analyzed descriptively and tested for statistical significance with *t*-test analyses or x^2 techniques as appropriate.

The intent of the study *a priori* was to conduct an analysis according to the two groups of interest, screened and unscreened children. A total of 24 of the 26 screened children (92.3%) were diagnosed prior to 9 months of age. Only 10% of children in the unscreened group were diagnosed before 9 months. Consistent with much of the literature on the effects of early identification of hearing loss, participants were also segmented into early and late identified groups for further exploration of the data. Consideration was given to examining the data for children diagnosed before and after 6 months of age, as this age division corresponds to much of the literature and the goal of the Ontario Infant Hearing Program.[15,27] However, given that only 15 children in this sample were diagnosed before 6 months of age (and that 9 started intervention by 6 months of age), and that the screened and unscreened groups corresponded closely to children diagnosed before or after 9 months of age, we examined outcomes in children identified before 12 months (e.g., n=26 for the PPVT) with those identified after 12 months (e.g., n=27 for the PPVT).

Pearson correlations were conducted to examine the relationship between age of diagnosis and outcome for each speech-language measure. Information was also collected to capture other potential predictors that affect communication outcomes. Using Pearson correlations,

analysis of variance and x^2 techniques as appropriate, relationships were also examined between outcomes and the following potential predictor variables: severity of hearing loss, highest level of parent education (years), scores on the CDI self-help subtest, age at assessment, NICU admission and gender.

Multiple linear regression analyses were performed to assess predictors of communication outcome. All regression models were constructed using the enter method to examine the effects of age of diagnosis on the results for the PPVT, PLS-4, and the GFTA, controlling for the potential effects of several other factors. In addition to age of diagnosis, the following variables were selected: degree of hearing loss (pure tone average in the better ear), highest level of parent education (years) and self-help status (measured from the CDI self-help scale). These variables were selected based on the primary variables of interest in this study and on an examination of relationships between the speech-language outcomes and predictor variables.

Results

Characteristics of the Study Population

A total of 65 children (35 male and 30 female) participated in the study during the 36-month period, October 2002 to October 2005. Thirty-five children (59.4%) had been identified with hearing loss from 1999 to 2002, prior to the start of the study assessments while 30 were identified with hearing loss and entered the study shortly after the start of the project. A total of 26 children were identified through systematic screening, 14 as part of targeted

screening prior to the 2002 UNHS program and 12 as part of the UNHS initiative. The remaining 39 children were diagnosed through traditional referral practices. There was no difference in the early confirmed (screened) and later confirmed (unscreened) groups with regard to the diagnostic period. Sixteen of 26 (61.5%) screened children were identified in the period 2002 to 2004 as were 24 of 39 referred children (61.5%).

Table 1 provides the demographic details for the participants according to the two groups, screened and unscreened. The two groups were similar for several demographic and clinical characteristics including gender, highest level of parent education, time in intervention, pure tone average and age at assessment. The overall age of diagnosis for the 65 participants was 14.0 (SD 11.2) months. As anticipated, there was a significant difference in age at diagnosis of hearing loss ($t=6.68$, $P<0.001$) with the screened group diagnosed at an average age of 6.1 (SD 3.7) months, that is, on average 13.2 months earlier than the mean age of 19.3 (SD 11.5) months for the unscreened group. The 12 children in the UNHS screened group were diagnosed at an average age of 5.3 months and the targeted screened group was diagnosed at a mean age of 6.8 months. Mean age of first hearing aid fitting and mean age of entry into intervention were closely related to age of diagnosis (Pearson's $r = 0.92$ and $r = 0.93$ respectively). Age at intervention was defined as the first chart recorded therapy session. The average (median) delay from diagnosis to intervention was 1.4 months, range, 0 to 26.1). An examination of the data revealed that 6 children entered therapy programs more than 6 months following identification. This delay was accounted for by a diagnosis of auditory neuropathy in 2 children, family reluctance to proceed with hearing aids or therapy

in 3 cases and 1 case of fluctuating hearing loss that was monitored prior to hearing aid fitting. On average, the referred group started rehabilitation intervention programs at 21.7 (SD 12.2) months, that is, 12.4 months later than the screened group who were enrolled in intervention services at an average age of 9.3 (SD 7.2) months. All participants were in oral programs with 59 of the 65 children documented as enrolled in auditory-verbal therapy, reflecting the primary choice of parents in the Ontario system (unpublished data).

Clinically, the screened and unscreened groups did not differ significantly in terms of severity of hearing loss as measured by pure tone average in the better ear ($t=1.33$, $P=0.19$). The mean pure-tone-average for the entire group was 79.4 dB HL (SD 24.3, range 23.3 to 113.3). Twenty-two children had hearing losses in the mild, moderate or severe categories (9 screened, 13 not screened) and forty-three children had losses in the severe and profound categories (17 screened, 26 not screened). Children with severe and profound hearing losses accounted for 66.2% of the total sample. Thirty-six children used hearing aids and 29 participants used cochlear implants at the most recent assessment. Six of the children initially fit with conventional amplification were implanted during the 3-year study period covered by this report. Earlier age of diagnosis was significantly associated with earlier age of cochlear implant surgery (Pearson's $r=0.53$, $p=0.003$). The mean age of cochlear implant surgery for the 29 children was 23.6 (SD 12.3 months).

The clinical profile of the children differed with regards to admission to NICU. Seventeen (65/4%) of the 26 screened children were NICU graduates compared to 6 (15.4%) of the 39

unscreened children. This difference is accounted for by the fact that only children with neonatal indicators were systematically screened for hearing loss prior to the implementation of the provincial UNHS program in 2002. Detailed information was available on 23 and 14 NICU graduates respectively for gestational age and birth weight. The mean gestational age was 30.1 weeks (SD 5.8) and the mean birth weight was 1513.8 grams (SD 1181.7, range 730 to 4450 grams). Children with documented complex medical and developmental disabilities were not enrolled based on the exclusion criteria.

Study inclusion criteria were strictly applied to patient recruitment in Ottawa and Toronto and a post-hoc examination of differences between groups from the two cities did not reveal any differences on several key variables. Applying x^2 analyses or t-tests as appropriate, the 36 children enrolled from Ottawa did not differ significantly from the 29 children in Toronto on the following variables: screening status, NICU admission, family years of education, and severity of hearing loss (either by category of loss or by pure tone average).

Speech and Language Outcomes

Assessments were completed on 65 children at various chronological age intervals. Due to age of entry in the study, scheduling issues, child attention/interest and compliance with study protocol, the number of children completing each assessment measure varied. Figure 1 details the flow of participants through the study protocol.

The results for the direct child assessments and for the parent ratings on the CDI are shown in Table 2, grouped by screening status (screened or unscreened). The mean data are presented for each outcome measure and represent the results obtained at the most recent assessment interval of 24 months (CDI only) 36, 48 or 60 months. An analysis of variance for each speech-language measure revealed no significant difference by age at assessment for either the screened or the unscreened groups. Table 2 summarizes the results as mean standard scores and standard deviations for the speech and language tests: PPVT-III, PLS-4 - Auditory Comprehension and Expressive Communication subtests, the GFTA-2 Sounds-in-Words subtest. For these test measures, a standard score of 100 places a child within the average range with 50% of the normative sample scoring between 90 and 110. The group data are shown as mean language quotients for the CDI language comprehension and expressive language subtests. A language quotient of 100 indicates that a child's language age is equivalent to his/her chronological age, that is, typical language development level.[15] A language quotient below 100 indicates that the child's language is below that of typical hearing peers for the test measure.

As shown in Table 2, the difference in performance between the screened and unscreened groups was not found to be statistically significant for any child administered speech and language outcome measure. Data were also examined dichotomously by age of diagnosis with the group divided into children identified before 12 months and after 12 months of age. Between group performance was not clinically or statistically significant on the PPVT (t=-0.741, P=0.46). Similar results were obtained across all speech-language test measures in

the study. In summary, for the speech and language measures in this study, there was no statistically significant difference in outcomes between screened and unscreened children or between children identified before 12 months and after 12 months of age.

Regression Analysis

Correlation analyses showed no relationship between age of diagnosis and communication performance on any measures (PPVT-III, PLS-4, GFTA-2 and CID) suggesting no trends towards improved language performance as a function of age of diagnosis for any of these test measures. Severity of hearing loss was weakly correlated with outcomes on the child administered tests but not on the parent administered CDI. Outcomes on both PLS-4 subtests were weakly correlated with parent education and self-help quotient. In addition, an examination of outcomes in relation to gender and NICU status using x^2 analyses showed no significant relationship with any outcome measure. There was no theoretical basis or support in the literature for examining NICU as an effect modifier. The self-help quotient which was taken from parents' ratings on the CDI self-help scale was used as a proxy for non-verbal abilities and was available for 43 participants. Multiple linear regression analyses were performed to assess predictors of outcome on the child-administered measures from these variables: age of diagnosis, severity of hearing loss, family education and self-help quotient. Controlling for the possible confounding effects of these indicators, age of diagnosis was not associated with improved outcomes for any of the outcome measures analyzed. The coefficients for the child administered speech-language assessments are provided in Table 3. Better communication outcome was significantly associated with

severity of hearing loss (pure tone average) on all measures except the PLS (AC) (P=0.06). Severity of hearing loss accounted for 11 to 15% of the variance on the various outcome measures. Accordingly, in clinical terms, for every 10 dB decrease in pure tone average in the better ear, an increase in the standard score of 2.9 units could be predicted for the PPVT. Better outcome was also associated with the self-help quotient on all child-administered test measures, accounting for variance ranging from 9 to 15% for the various tests. Family education was associated with higher standard scores (explaining 10 to 21% of variance) on both subscales of the PLS but not on the PPVT or GFTA. The models generated using these four explanatory variables explained from 29% of the total variance in outcome on the PPVT (F=3.82, P=0.01) to 47% of the variance on the PLS (AC) (F=8.32, P<0.001).

Discussion

The ultimate purpose of population hearing screening is to allow children to benefit from hearing in their everyday lives. Improved communication outcomes as quantified through traditional clinical measures would provide one source of evidence of the positive effects of UNHS. This study examined the trends in communication development relative to age of diagnosis of hearing loss in a cohort study of 65 children in an effort to contribute to the evidence base for the early identification of childhood hearing loss. In this study, overall, preschool children identified through screening, and therefore on average identified earlier, did not perform better than children identified without systematic screening (later identified) on several measures of communication development. A dichotomous examination of the data by age of diagnosis, before and after 12 months, also did not reveal any trends for

improved outcomes in the earlier diagnosed groups. Furthermore, there was no association between age of diagnosis and outcomes for any of the child or parent administered language measures analyzed in this study. Controlling for potential confounders, we found that severity of hearing loss and self-help quotient as rated by parents on the CDI were the primary predictor variables related to outcome.

We believe this study has demonstrated that age of diagnosis may not emerge to be the strong predictor of outcome in all clinical contexts as suggested in previous literature. There are several possible explanations for our findings. First, in terms of the age of diagnosis, there was only 13 months difference on average between the screened and unscreened groups in this study. Second, the service model of early quality oral intervention may in fact, have compensated for the delay in the later identified children. Children identified "late" at an average age of 19 months may, in fact, be able to catch up when appropriate supports are in place. Third, our sample was characterized by a significant number of children with severe and profound hearing loss; 29 of the 65 children used cochlear implants. However, children did not typically receive a cochlear implant until at least 12 months of age, therefore age of identification may have had less impact on outcomes for this population.[25] In this study, the mean age at implantation of 23.6 (SD 12.3) months, did not differ significantly for the 8 screened (20.7 months) and 21 unscreened (24.7 months) children who received implants. Fourth, later identification may have a lesser impact on families who have higher education levels. It is plausible that these families may be more resourceful in quickly accessing specialized care and adjusting their family circumstances to

reduce the gap in their children's language.[33] Although this study recruited participants through publicly funded health programs and attempted to enroll a range of families in terms of socio-economic status, the families in our study reported an average of 17 years of education, much higher than the average 14.3 years reported by Statistics Canada for Ontario generally (for individuals in the 25 to 44 year age range, the age range of the majority of parents in this study).[34] Our findings support the fact that family characteristics, access to resources, and family involvement may have a protective effect and reduce the negative effects of late identification of hearing loss.

Fifth, due to the timing of the study, the screened group in this study included a large proportion of NICU graduates. It is plausible, although impossible to disentangle in this study, that early identification had a positive effect on children from the NICU. Accordingly, early intervention may have provided these children with the opportunity to catch up with their non-screened, non-NICU peers. Although NICU admission was not significantly associated with any of the communication outcomes in this study, this could have reduced the overall benefits of early identification as infants who graduate from the NICU have been shown to have delays in development.[35]

Finally it is possible that the outcomes we measured in the first few years post diagnosis simply do not reveal the cumulative longer-term benefits of early identification of childhood hearing loss. Assessments of children at later ages involve more complex language skills

(with less visual support) and may expose differences related to earlier identification of hearing loss.

The findings of our study contrast with those of several studies in the past decade that have addressed the impact of early identification of hearing loss on communication outcomes.[16,21,22,26] The discrepancy in findings may be related to unknown and uncontrolled factors in the samples studied, such as service delivery model, therapy approaches (ours focused on oral therapy methods only), and the measures and methods used in evaluating benefits. Our research differed from the British study recently reported by Kennedy and colleagues on one important aspect that may account for some of the differences in findings.[26] Our cohort of children identified from 1999 to 2002 was much younger than the British birth cohort (from 1992 to 1997) and therefore may have had access to newer hearing technology and auditory rehabilitation. For example, cochlear implantation was a standard intervention for all children with severe to profound hearing loss in our study whereas in the British study, only 16 children had cochlear implants (although 26 had hearing loss of ≥ 95 dB), likely representing the era in which this cohort was identified and received early intervention. Furthermore, in our study, we are unable to point to important differences in screened or unscreened children with respect to access to cochlear implant technology either in terms of availability or age at implantation. There is also no evidence to suggest that different rehabilitation techniques or changes in service provision (e.g., therapy) occurred in conjunction with screening in the collaborating centers during the time period of our study. The access to cochlear implants in particular, may have moderated the effect of significant

hearing loss and reduced the impact of early diagnosis on language development.[25] In addition, children in our study were in rehabilitation programs focused on auditory-verbal language development and all test measures were administered using oral communication only. The effects of early identification could be different for children in other rehabilitation programs or in services where the focus is not exclusively on oral communication.

Our study suggests that when children are diagnosed before about one and a half years of age and quality intervention services are in place, age of identification may have less effect on communication development. Our findings are in agreement with those of a population-based Australian study by Wake and colleagues who recently reported no trends towards improved communication skills for school age children.[9,24] A strength of our study is that our findings are based on a comprehensive assessment protocol and rigorous data collected prospectively. Data were collected by examiners who were not involved in the child's clinical care and who were blinded to the extent possible to the child's degree of hearing loss and screening status. Demographic and clinical characteristics were collected through chart records and prospectively from the parents themselves. The multi-center nature of the study leads us to suggest that our findings may apply to other settings, with clinical populations in oral communication programs, where quality early management services are available.

Consistent with most studies in the infant hearing field, our study was dependent on voluntary participation and although we included a control group, we were unable to employ

a randomized control design. A randomized controlled design would not have been feasible as province wide universal screening was already available at the start of this research. Like other studies, we made the assumption that children in the non-screened group had congenital/early onset hearing loss. The advent of organized newborn hearing screening programs with good information systems will be expected to improve accuracy in future studies and may illuminate and eliminate some of the uncertain variables inherent in current studies. Although several variables, such as intervention and service models could not be controlled, the prospective cohort design allowed careful documentation and examination of several potential predictor factors. A limitation of this study is the number of children recruited despite substantial efforts over a three-year period. In addition, completion of the entire test battery at all of the study intervals proved to be a daunting task with preschool children and families. Given the low incidence of childhood hearing loss, the heterogeneity of the population and the potential selection bias due to the apparent unacceptability of controlled interventions, the task of recruiting adequate sample sizes remains a major challenge and barrier to establishing empirical evidence in this research area.[36] Notwithstanding these limitations, we find it striking that this study revealed no trend towards improved communication outcomes for earlier identified children.

This study, along with others, points to the fact that it is difficult to separate the contribution of age of diagnosis to communication development from that of other variables. The impact of new technology such as cochlear implants on early language acquisition should not be underestimated.[25] Other factors such as parental involvement, service models and quality of

therapy and services may also be important determinants of outcome. These latter potential contributors are difficult to quantify and analyze. UNHS research has demonstrated that early identification can be achieved and early intervention can be initiated.[23] This may lead to early and effective access to care. It is probable that comprehensive services, some of which may be related to early identification like the model of care and intervention, rather than age of diagnosis alone, is the most effective way to improve long-term communication and academic outcomes in children with hearing loss. Further research should focus on a better understanding of the contribution of these less measurable factors. Recently Hyde has suggested that the emphasis on improved communication development as an outcome measure for the evidence of the effectiveness of screening and early identification of hearing loss is inappropriate.[37] Our multi-center study raises questions about the advocacy for population hearing screening solely on the basis of improved communication outcomes. Indeed, results from qualitative interviews that we have conducted with parents of children who were screened or referred, suggest that parents value outcomes of hearing screening that extend beyond traditional communication competency measures.[33]

Educators and health care providers have been striving to improve the poor performance levels in communication and academic skills for children with permanent hearing loss. As evidenced by the recent emphasis on universal hearing screening initiatives, many researchers and providers believe that better outcomes can be achieved through implementation of population based hearing screening. In summary, a desirable outcome of an evaluation of the potential advantages of infant hearing screening would be improved

communication development. To date, such results have been difficult to obtain in a rigorous way. The inconclusive findings may be due to an absence of effect of early diagnosis, to the difficulty of quantifying such benefits, to the absence of studies that measure benefits over the long-term and finally to the numerous factors, in addition to early identification, which impact on communication development. Given the severe consequences associated with permanent childhood hearing loss, it is important to continue to scrutinize the effects of newborn hearing screening. The continued evaluation of the longer-term benefits of early identification including multiple dimensions of outcome and parents' perspectives may provide further insights into the value of this population-based intervention.

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Table 1. Demographic characteristics of children (n=65)

	Screened (n=26)	Not Screened (n=39)
Gender - number (%)		
Male	15 (57.7%)	20 (51.3%)
Female	11 (42.3%)	19 (48.7)
NICU admission - number (%)*	17 (65.4%)	6 (15.4%)
Parental education – years (SD)	17.0 (2.4)	17.2 (2.7)
Age diagnosis – months (SD)*	6.1 (3.7)	19.3 (11.5)
Age intervention – months (SD)*	9.3 (7.2)	21.7 (12.2)
Time with hearing device – months(SD)[1]	31.3 (12.2)	28.4 (10.6)
Time in intervention – months (SD)[2]	28.3 (11.7)	25.8 (10.0)
Age at assessment – months (SD)[3]	47.8 (10.3)	51.6 (10.4)
Pure tone average – db HL (SD)[4]	74.5 (25.6)	82.6 (23.1)

*statistically significant $P<0.001$; [1]at most recent assessment; [2]defined as chronological age at most recent assessment minus age of first hearing device fitting (i.e., hearing age); [3] calculated for the tests (PPVT, GFTA, PLS-4 (AC), PLS-4 (EC)) administered beginning at a chronological age of 36 months; [4]Pure-tone average at 500, 1, and 2 kHz in decibels hearing level in the better ear.

Table 2. Results of speech-language assessments for screened and unscreened groups

Measure	**Screened** **Mean (SD) [N]**	***Not Screened*** **Mean (SD) [N]**	**P Value**
PPVT-III	89.3 (18.7) [17]	86.9 (18.9) [36]	0.66
PLS-4 (AC)	99.0 (20.8) [16]	96.7 (15.2) [34]	0.66
PLS-4 (EC)	95.1 (22.1) [16]	89.2 (19.6) [34]	0.35
GFTA-2	87.5 (24.4) [17]	83.1 (19.7) [35]	0.49
CDI-Rec	78.9 (26.1) [20]	84.1 (23.7) [31]	0.46
CDI-Exp	82.5 (28.1) [20]	80.7 (29.5) [31]	0.82

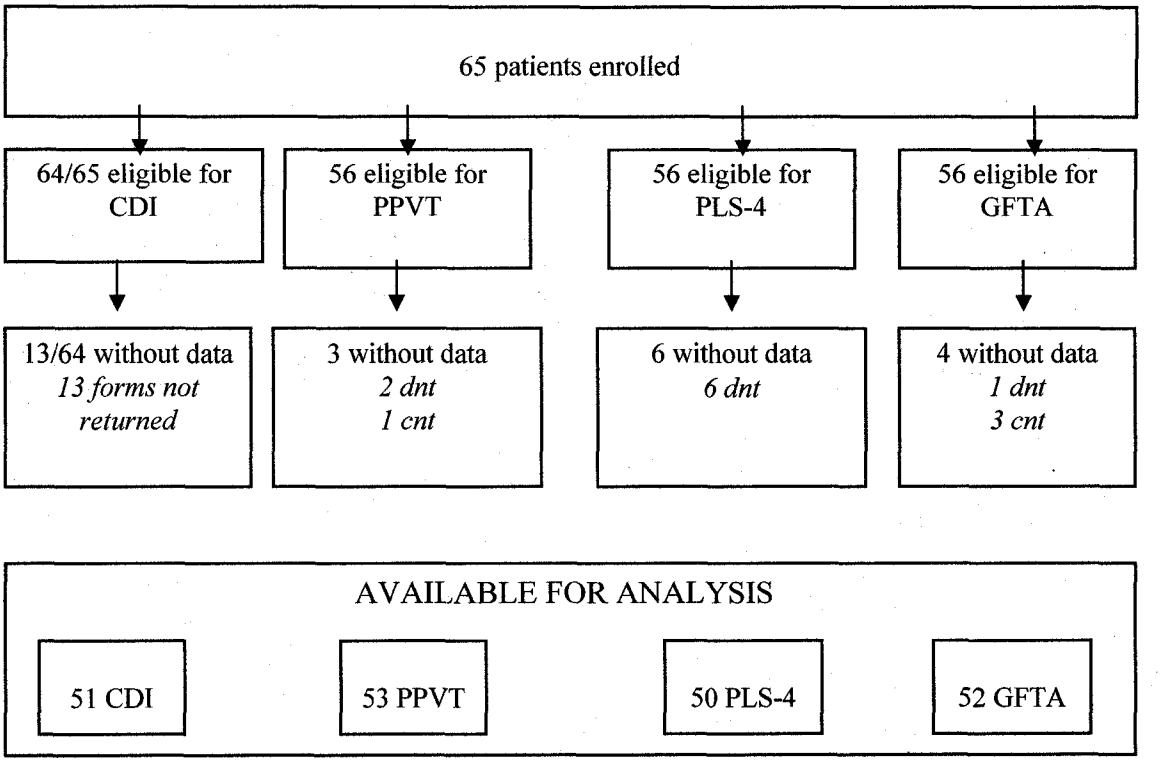
PPVT-III: Peabody Picture Vocabulary Test-Third Edition; PLS-4: Preschool Language Scale-Fourth Edition; AC: Auditory Comprehension; EC: Expressive Communication; GFTA-2: Goldman-Fristoe Test of Articulation-Second Edition; CDI: Child Development Inventory; Rec-Receptive language; Exp: Expressive language

Table 3. Multiple linear regression of four independent variables to predict outcome

Measure	Coefficient	P-Value	95% CI
PPVT-III			
Age diagnosis	-0.04	0.88	-0.51 to 0.44
Pure tone average	-0.29	0.01	-0.50 to -0.07
Family education (yrs)	1.65	0.14	-0.55 to 3.85
Self-Help scale quotient	0.22	0.02	0.03 to 0.41
PLS-4 (AC)			
Age diagnosis	-0.08	0.70	-0.46 to 0.31
Pure tone average	-0.17	0.06	-0.34 to 0.01
Family education (yrs)	3.46	<0.001	1.66 to 5.26
Self-Help scale quotient	0.27	0.003	0.10 to 0.44
PLS-4 (EC)			
Age diagnosis	-0.42	0.08	-0.88 to 0.05
Pure tone average	-0.29	0.008	-0.50 to -0.08
Family education (yrs)	2.81	0.01	0.67 to 4.95
Self-Help scale quotient	0.33	0.002	0.13 to 0.54
GFTA-2 (Sounds-in- Words)			
Age diagnosis	-0.32	0.24	-0.87 to 0.22
Pure tone average	-0.32	0.01	-0.57 to -0.08
Family education (yrs)	-0.54	0.67	-3.06 to 1.98
Self-Help scale quotient	0.22	0.05	0.001 to 0.43

CI: confidence interval; PPVT-III: Peabody Picture Vocabulary Test-Third Edition; PLS-4: Preschool Language Scale-Fourth Edition; AC: Auditory Comprehension; EC: Expressive Communication; GFTA-2: Goldman-Fristoe Test of Articulation-Second Edition

Figure 1. Flow of Participants through Test



Key: dnt-did not test; cnt-could not test

CHAPTER 3

PARENTS' PERSPECTIVES ON THE IMPACT OF THE EARLY DIAGNOSIS OF CHILDHOOD HEARING LOSS[1]

[1] Formatted for the International Journal of Audiology

Parents' Perspectives on the Impact of the Early Diagnosis of Childhood Hearing Loss

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Key Words: hearing loss, hearing screening, children, benefits, parents' perspectives

Abbreviations: Universal Newborn Hearing Screening (UNHS); Canadian Working Group on Childhood Hearing (CWGCH); United States Preventive Services Task Force (USPSTF); Infant Hearing Program (IHP)

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Abstract

Newborn hearing screening has been widely implemented to improve outcomes for children with permanent hearing loss. This study examined benefits beyond those typically measured clinically, by exploring parents' perceptions of the effects of early/late identification of hearing loss. The parents of 17 children in Ontario, Canada, participated in the study. Seven children were identified through systematic screening and ten through traditional referral practices. All children were in oral rehabilitation programs. The study adopted a qualitative approach, examining parent's views through individual interviews. Purposive sampling was used to select a diverse group of parents to allow a broad range of perspectives to emerge. Benefits of early identification included improved communication development and early access to hearing. Negative aspects of late identification included regret for the family and family stress around the child's language gap. Although, screening programs may offer a seamless transition to audiology services, the transition to intervention services appeared less fluid for some families. Overall parents strongly support infant hearing screening and identify benefits that are not easily quantifiable through traditional clinical measures.

Recent years have marked an unprecedented interest in the early identification of childhood hearing loss. The consequences of permanent childhood hearing loss, a disorder affecting 1 to 4 per 1000 children typically include significant delays in communication, academic and social development (Yoshinaga-Itano, 2003a;Wake et al, 2004; Davis et al, 1997). One factor believed to contribute to these poor outcomes is the late detection of hearing impairment (Moeller, 2000; Yoshinaga-Itano, 2003b). Consequently, advances in hearing screening technology have led to the implementation of population-based hearing screening in much of the developed world in an effort to alleviate the deficits associated with childhood hearing loss.

Universal newborn hearing screening (UNHS) is predicated on the fact that earlier diagnosis translates to early intervention and improved developmental outcomes particularly in the areas of speech and language acquisition. There is a substantial body of evidence supporting newborn hearing screening as a means to early identification of hearing loss (Canadian Working Group on Childhood Hearing, 2005). In recent years, there has also been considerable interest in rationalizing UNHS and therefore the early identification of childhood hearing loss as a means to improved communication and subsequent literacy, academic, and quality of life outcomes. The value of UNHS has been investigated primarily through research approaches that attempt to quantify the benefits of early detection and management of hearing loss in terms of later communication development. During the 1990s, several studies provided support for the benefits of early diagnosis in achieving better developmental outcomes. In essence, these studies concluded that early diagnosis leads to

improved outcomes in communication and provided the impetus for a strong advocacy for universal screening programs. (Yoshinaga-Itano et al, 1998; Moeller, 2000; Calderon & Naidu, 2000; Yoshinaga-Itano, 2003a). However, the conclusion that early identification of hearing loss results in improved communication outcomes has been called into question in recent years. In particular, a systematic review commissioned by the United States Preventive Services Task Force (USPSTF) judged the evidence supporting the positive impact of early identification on communication development to be poor to fair. This rating was based on the quality of studies retrieved that drew conclusions on the basis of primarily convenience samples and weak methodological designs. The same review ascertained that there was fair to good evidence that UNHS leads to earlier diagnosis of hearing loss than does the traditional referral system. (Thompson et al, 2001).

The USPSTF systematic review and subsequent critical reviews undertaken by the Canadian Working Group on Childhood Hearing (CWGCH) acknowledged and underscored the difficulty of documenting the benefits of universal screening and early identification of hearing loss (Thompson et al, 2001; Canadian Working Group on Childhood Hearing, 2005). The USPSTF review pointed out that there were no studies addressing quality of life outcomes despite the fact that a key assumption underlying UNHS is that delay in diagnosis and treatment create anxiety and stress for the family and child. The CWGCH also called attention to the notion that exploration of other outcomes that extend beyond communication development is important in understanding the potential benefits of early identification of hearing loss. In fact, previous research into the benefits of early identification and

intervention have made an important contribution to the field in that they illuminate the difficulties of quantifying the advantages of early intervention in an area characterized by small heterogeneous populations and multiple child, family and environmental factors that potentially impact developmental outcomes. UNHS is a public health initiative embedded within a complex social, economic and environmental context. Accordingly, traditional research addressing communication development outcomes may have limitations in demonstrating the value of early identification of childhood hearing loss. Traditional outcome measures complemented by qualitative research approaches may more fully inform the discussion on the effectiveness of newborn hearing screening. It is perhaps an opportune time to involve those individuals most affected, that is, parents of children with hearing loss, in defining the important variables that should be addressed in Early Hearing Detection and Intervention (EHDI) outcome research.

Positive attitudes towards screening have been reported in studies addressing the perceptions of both parents of children with hearing loss and those with normal hearing (Watkin et al, 1998; Magnuson & Hergils, 1999; Watkin et al, 1995; Clemens et al, 2000). Furthermore, in an investigation of preferences for a screening approach, mothers, pediatricians and hearing specialists all favored a universal hearing screening system over a high-risk or targeted approach (Wall et al, 2001). Taken together, these studies suggest that parents value the implementation of UNHS programs. More recently, interest has moved from investigating parents' support for UNHS to including their views and experiences as consumers regarding the practices of system-driven screening and identification. Reports

from England, where the implementation of a country-wide screening program is being evaluated, identified factors such as professional communication and manner as having an important effect on parents' experiences with screening and early identification (Young & Tattersall, 2005; Tattersall & Young, 2005). However, to our knowledge, no studies have been conducted with parents from regions with recently implemented newborn hearing screening programs to understand the effects or important outcomes of early identification programs from parents' perspectives.

This study was undertaken to explore the effects of early or late identification of childhood hearing loss from the perspective of parents, valuing both the views of parents who had experienced identification of their child's hearing loss through a systematic screening process (universal or high-risk) and those who had learned of the hearing loss through traditional referral patterns, for example, referral through family physician.

Context of the Current Study and Study Objectives

A province wide population infant hearing screening program was introduced in 2002 in Ontario, Canada's largest province of 11 million people, to replace the previous patchwork of services that either included local targeted screening initiatives or no systematic screening at all. This program involves not only universal screening as a core service, but key to the initiative is the inclusion of services focused on early hearing and communication development (Hyde et al, 2004).

In the context of this new program, the aim of this study was to gain a greater understanding of the benefits and experiences of early diagnosis for parents of children with hearing loss. The objectives of this study were to gain insights into 1) parents' perspectives of the impact of hearing screening and early intervention, 2) parents' needs in the early stages of learning about and managing their child's hearing loss, and 3) the important elements of a service delivery model for families following diagnosis of hearing impairment. Through the narratives of families of young children with hearing impairment, this paper reports the findings for the first objective, that is, parents' perceptions of the effects of the early identification of hearing loss.

Method

Sampling Frame and Recruitment

The participants for the qualitative research inquiry were recruited primarily from the 65 families currently participating in an ongoing longitudinal study at three Ontario clinics investigating the impact of early identification of hearing loss on developmental outcomes (Durieux-Smith et al, 2002). In addition, parents were identified through clinicians at a fourth Ontario clinic.

The inclusion criteria from the original study were applied to this research inquiry, therefore parents of children with the following characteristics were considered eligible: 1) chronological age less than 5 years, 2) permanent bilateral hearing loss, 3) congenital or early onset hearing loss, 4) consistent use of hearing technology and/or enrolment in an

intervention program that emphasizes the development of spoken language, 5) English as the language of intervention. Children who were not enrolled in programs focused on the development of oral communication skills and children with hearing losses and additional complex disabilities were excluded as the programmatic needs of these two populations may differ significantly. These criteria allowed us to aim for homogeneity amongst participants with regards to characteristics assumed to be important to the study.

Participants were then selected through purposive sampling to include two primary groups of parents (mother or father), whose children were identified with hearing loss through a systematic screening process and those who did not experience screening. This sampling strategy was selected to allow us to examine the perceptions of parents who have and have not experienced the screening process as screening will likely result in early diagnosis of hearing loss compared to later diagnosis for unscreened children. In addition, maximum variation sampling was used to include within these two groups, parents of children with different characteristics, to allow the broadest range of perspectives and important common patterns on the topic to emerge. Accordingly, to obtain diverse views and experiences, we maximized the differences amongst the participants in the group and therefore based the selection of parents on the age of the child, degree of child's hearing loss (mild to profound), type of hearing device used (hearing aid or cochlear implant) and geographical location.

It was anticipated that 15 to 20 participants would constitute a sufficient number to reach data saturation for this study. However, sampling in qualitative inquiry is generally flexible

and was left to evolve as the study progressed. The parents selected from those who were already enrolled in the longitudinal study were contacted by letter and the families from the fourth clinic (who are not part of the longitudinal study), were informed of the study through the providers in their audiology clinic. Research Ethics Board approval was obtained from all institutions involved and written informed consent was obtained from the participants prior to the initiation of data collection.

Procedures

Data were collected through individual semi-structured interviews with parents. An individual interview format was selected to facilitate participation for parents so that they could more comfortably express their feelings about the process of learning about their child's hearing loss. The meetings were conducted in the parents' home in all but one case where it took place at a parent's office. The context of people's personal lives and the environments and situations they encounter as they parent a child with hearing loss influences their responses. It was anticipated that collecting data in the home environment whenever possible, would immerse the researcher in the current world and experiences of the participants, thereby enhancing understanding of the context. All data were collected by one interviewer, a clinician-researcher with several years experience in pediatric audiology and family-centred rehabilitation programs. Other members of the research team who were involved in debriefing sessions included professionals in sociology, audiology and health economics. This inquiry was undertaken as part of doctoral research in Population Health studies.

The interviewer asked open-ended questions to encourage participants to share their experiences. An interview guide was developed to structure the session and ensure that the major topics of interest were covered (Table I). The interview was partly guided by specific questions to facilitate probing of specific areas of interest for this research. However, the researcher also formulated other questions as the interview proceeded depending on the responses of the parents. The questions were piloted for clarity with the first two participants in the study. The interviews lasted an average of approximately 45 minutes. All interviews were audiotaped with the consent of the participants in order to facilitate detailed analysis. Due to technical problems one interview was not recorded and the interviewer recorded a detailed summary of the discussion immediately following the interview. Field notes to summarize overall impressions were written after the interviews, and included documentation of any topics leading to intense dialogue, noticeable enthusiasm or hesitation.

Baseline characteristics were collected using two questionnaires, a family information questionnaire and an intake questionnaire. This information was available as part of the longitudinal study in which children from 3 sites were enrolled and was collected separately for the additional site. A brief demographic questionnaire on screening status, age of identification/intervention and type of intervention was also completed at the time of the interview to validate the information previously collected.

Data Analysis

The focus of qualitative research is on process as well as outcome, therefore data collection and data analysis proceeded concurrently. The audiotapes were transcribed and analyzed as soon as possible after the interview such that the preliminary analysis could guide the future data collection. In this sense, the theoretical frame of the research was not predefined but rather was based on the "incoming" data. Inductive thinking was applied to move from data documentation to interpretations and conclusions.

The interviewer listened to and transcribed the tapes shortly after the interview and all transcripts were entered into qualitative research software, ATLAS.*ti*. All data were reread several times for coding over several weeks to allow categories to emerge. The coding process included a constant comparative method which involves open, axial and selective coding (Strauss & Corbin, 1998). Open coding involves breaking down the data into discrete segments in order to closely study the data and label the concepts or categories. During axial coding, these categories are examined for links and connections and during the selective coding process, categories are integrated to summarize the research findings. In addition field notes and the interviewer's reflections post-interview were used in finalizing concepts and categories. Data collection continued until data saturation was reached, that is no new themes emerged during the analysis. As a quality check, one other reviewer read the coded transcripts and verified the codes/categories. Any disagreements were discussed and resolved through consensus.

Results

Description of Participants

At their discretion, parents participated in the interviews individually or with a significant other person. All were hearing parents and the 17 interviews involved a total of 21 individuals, with participation as follows: 10 mothers alone, 1 grandmother (the child's main caregiver), 2 fathers, 4 mothers and fathers together. Two of the families also had a second child with a hearing loss. Fifteen of the 17 families lived in an urban center. The majority of parents (11 of 17) had experienced the diagnostic process for a period of less than 3 years (range 11 months to 44 months) prior to participating in the interviews.

Seven babies had undergone a systematic hearing screening procedure (5 universal screening, 2 high risk screening) and ten had been referred for hearing assessments through traditional referral routes, typically through the family doctor, because of parent suspicion of a problem. The average age of diagnosis of all children in the sample was 16.1 months (range 0.8 to 41.9 months). Nine children were diagnosed before 12 months of age; six of whom had undergone systematic hearing screening. The sample was selected to include children with varying degrees of hearing loss and different hearing devices. The sample included more children with severe and profound degrees of hearing loss consistent with participation in the larger study from which the majority of families were recruited. Severity of hearing loss was distributed as follows: 1 mild, 5 moderate, 6 severe and 5 profound hearing losses. Ten children used hearing aids and seven had received cochlear implants at the time of the interview. All 17 parents reported post-secondary training, including 12 who

had completed university level studies and 5 with college or trade level education (average of 16.8 years of education, range 13-21 years). Fourteen of these 17 families, who had selected an oral communication rehabilitation approach, were enrolled in auditory-verbal therapy, reflecting the primary rehabilitation choice for families in the province of Ontario.

Benefits

The aim of this dialogue with parents was to understand the meaning and important outcomes of early identification of childhood hearing loss from the perspective of families who had experienced the process within the past few years. For the parents in this sample, the dominant theme was that early diagnosis of hearing loss is valuable and that universal hearing screening should be included as part of the Canadian health care system. Table II provides a summary of the key issues associated with early and late identification of hearing loss from the perspectives of the families interviewed in this study.

The advantages of universal screening were probably expressed most emphatically by those parents who had not been aware of their child's hearing loss until well after their first birthday. The primary negative issues of late identification of hearing loss for these families included delayed diagnosis, missed opportunity for hearing, delay in language development and frustrations associated with accessing services.

... before he was 1 year old, I was questioning whether he was hearing or not and yes some days I would think that he was hearing and other days I would think that he wasn't..., and so

of course the process of getting him into (the hospital) was a lengthy one and then he wasn't very cooperative in the booth...by the time he was fully, like he was diagnosed, he was 21 months old.... He was born about 6 months before the early screening came into place. Yeah, so that (screening) would have been ideal because when I finally did get the diagnosis, I was thrilled to know so I could move on from there. (Peter's Mum, child not screened)

... she could have had a better start to language development and to input from her Mum and Dad. So because we started behind the 8th ball, we always felt like we were trying to catch up, yeah, so I would say that was our biggest issue for sure, aside from getting over the hearing loss in the first place, I would say, for sure, that just, we were always feeling behind. (Shannon's Mum, child not screened)

<u>Bonding</u>: Despite recognizing the value of universal screening, a few parents of children diagnosed through the typical referral process, commented that not knowing about the hearing loss until a little later, was positive in that it gave them time to bond with their child as "just a baby" and to adapt to just being a mother first. Based on their experience with their children's communication development, these parents seemed to be suggesting that the current goal of identifying hearing loss in the first 3 months may not be as critical as UNHS programs uphold it to be. This potential disadvantage of screening was, in fact, articulated by parents in both screened and unscreened groups.

.....so when she was finally diagnosed, she was 25 days old so I think it was hard. ...I mean, it's beneficial to know as early as possible but it's just hard on feelings, I didn't really enjoy (Baby) because I was so sorry and frustrated,... she failed the test the second day, the nurse said, you know, maybe it's wax, maybe it's some fluid, but still I worried, we worried, of course. I think I didn't enjoy the new baby like other mothers do 100%.... Yeah, it was hard... Now...we understand, oh, she's just our baby, yeah, well she's hearing-impaired but she's still, she's our baby.... So at first, when I learned (about) her hearing loss, it's just you know one day after, I think I didn't enjoy her as much as other mothers. (Ellen's Mum, child screened)

...And to be completely honest, I had a lovely year at home with him, in that quiet land of happy baby, no idea,... because, I imagine if somebody had told me at day one or day 5 ... that something was wrong, you would have just gone off the deep end, I imagine, because you pretty much do that anyway, but later. But at least then, you have, you know, the child is your child and ... If somebody said at day 5, and you've barely sort of recovered from having the baby, never mind someone telling you that there's something wrong with the baby. (Adam's Mum, child not screened)

Nevertheless, regardless of their experience around the identification of their child's hearing loss, whether their child was screened or not screened, whether diagnosis of hearing loss was perceived as early or late, all parents concurred that the efforts to detect hearing loss at the very earliest age should be continued. Parents described important health outcomes that

centered on both the child's abilities and family functioning. The benefits or important outcomes of early identification of hearing loss experienced or hoped for by these families could be summarized into four broad themes 1) benefits related to the child's development, 2) benefits related to the family, 3) impact on the process of identification of hearing loss, 4) impact on the process of care or management after identification. Parents' views are presented through their words in the sections below; only the child's names have been changed for confidentiality reasons.

Effects on the Child

Communication development: The majority of parents, in both the screened and unscreened groups expressed that they believed early diagnosis translated into a better long-term prognosis or enhanced opportunities for their child's speech and language development. The only exceptions were parents of children with profound hearing loss who received cochlear implants and who were identified before one year of age; these parents commented that the child would not have been implanted until one year of age regardless of the age of identification. As well, some of these parents described the hearing aid experience as so frustrating and apparently not beneficial for the child that they did not feel that awareness of the hearing loss prior to accessing sound through an implant was an advantage for speech and hearing development.

But I think that if (Child) had been hearing with a cochlear implant at a year instead of at almost 2 years, the difference in his life would have been tremendous, he would have, he

would have learned everything he's learning now a year before. So, right now, where he's about 6 months behind his peer group or like the bottom level of his peer group, he would have been probably within his own range. Because, you know, what I mean, that's the process of catching up. So if he would have access to sound a year earlier, plus just all the sort of peripheral effects that having no hearing input for that long, having no sound input for that long, that it has on your nervous system, and your brain development and all of that, we're very much aware of that and how it's going to impact him for the rest of his life. So, I think, that, the sooner, he would have had, you know, the sensory deprivation basically, if he'd been spared that a year earlier, then, yeah, it would have made a huge difference, I believe that, I see that in kids who are getting their implants younger than him. And the professionals around us report the difference as well, I think it would have made a huge difference, yeah. (Logan's Mum, child not screened)

Hearing access: In addition to the longer term benefit of enhanced speech and language abilities, parents talked about the child's access to hearing and environmental awareness as an important outcome of early identification. Parents frequently referred to this period of not knowing about the hearing loss as a missed opportunity for both parents and child. One mother described her disappointment that the child had missed out on so many sounds in her young world. Although several families felt that they had closed the gap, they reflected with sadness on the early experiences of the child.

...we feel like we've been doing tons of catch up, which you know, has been successful and I honestly believe we've caught up. She's, you know, been testing at her regular level... And you know, when we look back on her first birthday, and we see her and we're singing happy birthday, and realize she was never hearing any of that. I think wow, you know, you look at those videos, I can't watch them really, I have a hard time. (Anna's Mum, child not screened)

I think it did (late identification had an impact) because, I mean, she's our perfect little girl as we look at her and we love her to death, and looking at some of the additional things we could have been doing with her if we knew that she had a profound hearing loss. I think our activities of daily living might have been a little different, in the sense of stimulation, we thought that she could hear us, so we probably would have looked at maybe some of the opportunities we're giving her with toys, and providing her with maybe some additional sounds, some vibration, clarifying the sounds in the environment, responding differently. (Helen's Mum, child not screened)

Some families felt that it was better for the child's self-concept to have been identified in infancy both for the child herself and for the family with respect to accepting the child as an individual with a hearing loss. Two parents articulated this advantage as follows:

Ok, well, I think (screening is) definitely better, ...and I guess mostly in terms of he's grown up with them (hearing aids) so that's part of who he is. You know, if he would have gotten

hearing aids at 3 years, it would have taken, it took adjustment back then but I think it would have taken more adjustment because I would have known him as a child who I would consider you know, to be normally developing including the hearing, but that was basically part of who he was almost from the beginning. ...No I mean it was traumatic for us but no I don't think, you know, looking back, I would say it was negative. I'm glad we found out right away. (Eric's Mum, child screened)

Well, I just think he'll be more accepting of it (hearing loss), first of all because he doesn't know any different, you know... It's now to educate the rest of the family, the rest of the world, that if he acts a little odd or screams at something, don't think there's something wrong with him, it's just his reaction to it. I do believe firmly that the earlier the better... (Calvin's Mum, child screened)

Effects on the Family

A range of feelings were expressed by parents of children whose hearing difficulties were identified later in the absence of systematic screening. These included guilt about the unawareness of a hearing loss, and the "emptiness" of not knowing about a problem.

Regret: Both parents of screened and unscreened children referred to the notion of guilt associated with not being aware of the presence of a hearing loss. As described by a parent below, guilt was also present when parents felt they had not been persistent enough about having the child's hearing loss identified.

Well, I guess we felt guilty doubting ourselves, for not making someone listen to us, for all that stuff. So that was the negative impact, positive was, we didn't treat him any differently for 10 months. (Sam's Mum, child not screened)

Well, I guess it was lucky that he was screened because I don't think that we would have known for quite a long time if he wasn't screened because right now even, you can have a conversation with him without his hearing aids. ...I mean, now I would have picked it up because if he doesn't have his hearing aids and we say something, then, you know, he'll ask what, or say it again... But I don't think we would have picked it up. ...I think I would have felt guilty if it had gone on for a long time...for not noticing just because we're his parents, so... (Eric's Mum, child screened)

Other families described a strong need to know, and that a void was created by not knowing whether the hearing loss was present at birth. As described by one family, there was a nagging need to just know if the hearing problem was present at birth or if something had happened during early childhood. In other words, this family thought screening could contribute to a better understanding of the etiology of the child's deafness

Stress around language gap: In talking about the language delay associated with later identification, parents sometimes moved beyond discussing the direct effects on the child's development and shared how this generated stress and anxiety for the family. In referring to

the gap in their child's development, parents talked about a sense of urgency, and a feeling of lost time. In some cases, they felt they had little time to investigate the options for their child's communication development. It is this notion of gap that differs for families of screened and referred children particularly those identified at 12-18 months. Many families expressed feelings of frustration that time was running out and they referred to the pressure they faced to act and improve the situation. One parent summarized the overall effect of the perceived gap on the child's and family's life as follows:

Well, I think that it's (late identification) created a lot more work for all of us, it's created a lot more stress in his life. ...because...he's constantly playing catch-up and he has a hard time engaging in the kinds of games that other kids are engaging in because he lacks the language skills, ...but despite all the effort that goes into, with all the therapists and everything, he's still behind, ...so, I think the impact, has been you know, a bit deflating to his childhood. And of course, the stress to us, it's changed everything about our decisions about...how he's going to be educated, about whether I'm going back to work or not, everything has been impacted. (Logan's Mum, child not screened)

Not surprisingly, the shock and loss of learning of the presence of a hearing problem was sometimes described as greater by families of children screened for hearing loss, particularly well-babies, i.e., children who had not spent time in the neonatal intensive care unit (NICU). In contrast to families who had observed their children's behavior and suspected a hearing loss for some time, parents of healthy newborns had no reason to expect a hearing problem.

There appeared to be a trend towards two distinct groups of children who had undergone screening, those whose children exhibited no neonatal problems at birth and were simply having a hearing screening test and those whose children had been admitted to a NICU. This latter group potentially differs in that they were aware that their child was potentially at risk for many child development problems and were involved in a series of assessments. In some cases, they spoke of learning of a hearing loss early as comforting in that it was easier to cope with a hearing disorder than other developmental and cognitive disabilities.

It (screening) may have offered us a certain comfort factor. Because if I remember right, the screening they make little sounds and check that the brain is getting signaled and therefore it would tell us that some of the basic pathways are good, and in our case, because there were questions about her about all sorts of things with her back then, because she was multiple issues, ...but any positive results showing that some other part of her is working...but her hearing passed through the brain and still was signaling something, that was a good sign, so there was some comfort to it, I guess. (Olga's father, child screened)

I guess because he had so many health issues at first that it's really a miracle he's alive to begin with. So, it (hearing loss) was just another thing, kind of just sort of rolled off our backs, another problem, but we'll deal with it, you know what I mean. Whereas for some parents, it's a complete shock, their normal child or they've other issues and it's just, oh dear, what do we do next. (Calvin's Mum, child screened)

Effects on the Process of Identification of Hearing Loss

Parents explained how the process of learning about hearing loss occurs in several different steps. Each step adds a dimension of anxiety and concern and it is in these steps that major differences emerged between learning of the disorder through screening and learning through the typical referral process. The way in which the process of the diagnosis of hearing loss unfolded was different for these two groups of parents. Parents of referred children frequently described the process as frustrating and confusing. Parents of screened children expressed a high degree of anxiety in waiting for the audiological assessment after receiving a "refer" from the screening test.

Diagnostic process: Initially the frustrations for parents undergoing the typical referral process emanated from having to convince the physician to refer to audiology based on parental concern, and having to wait up to 2 to 3 months for an audiology appointment. These parents noted that in fact, frequently, they themselves had identified a hearing problem and were essentially waiting for confirmation and subsequent treatment.

As exemplified by one parent's experience, it was frequently not a question of "does my child have a hearing loss?" but "how much hearing loss does my child have?" and "what can be done?"

...and every day that went by, we'd call the pediatrician's office and he wouldn't have made the appointment. ...finally, we couldn't wait anymore, we weren't sleeping, so I called the

hospital directly...and as we walked into the hospital, we were perhaps a little different than other parents who maybe go in there and are surprised at what they hear. And we were going in there pretty much knowing what the news would be. (Yvonne's Mum, child not screened)

Therefore, being on a waiting list for an audiology test was described as being on a waiting list for treatment for a condition that is already known to exist. Although, the traditional referral route appeared to proceed smoothly for some parents, others described the process as frustrating and told how they learned to "work through the system."

I just called the pediatrician and asked for a referral...then there was like a month waiting list or something like that so I started calling every day, eventually, there was a cancellation. ...they sort of left us with, he probably has some fluid on his ears, and he should come back and be tested again. So, I guess they let us leave with that in our minds, I talked to my pediatrician, he said, oh yeah, fluid, I guess that's possible,.. He didn't actually see the results himself...I guess he was happy to reassure me... And then, so, I guess we went back, it was going to be another month so I started calling again, and eventually, we got another appointment about a week later, I guess if you're persistent, you get in sooner. (Adam's Mum, child not screened)

From these parents' experiences, screening appeared to facilitate access to qualified audiology services, in that for parents of screened children, there was a clear structure in

place from screening to audiologic assessment. In these interviews, no parents of screened children identified a problem with access to audiology. However, several described the period of waiting for the audiology appointment after a "refer" result from the screening test as highly anxiety producing but felt this was inevitable and not something that could likely be improved. One parent suggested that having the community screening test located in the same facility as pediatric audiology with immediate access to an audiologic assessment would have facilitated this very beginning stage of the process.

Not surprisingly, the shock and loss of learning of the presence of a hearing problem was sometimes described as greater by families of children screened for hearing loss. However, what appears to be different in some cases, particularly where the diagnosis was late, is the attitude of parents towards health care providers, their frustrations during the diagnostic process and their understanding of the next steps. Some families of children who were referred through traditional referral methods, described a frustrating, time-consuming experience. Parents of referred children talked about the frustration not only in terms of the initial access to an audiology service but the dissatisfaction of dealing with professionals who, in their view, lacked pediatric expertise. This concern was most apparent where pediatric audiology testing was available in various clinics but where the expertise was perceived to vary depending on the number of children with hearing loss serviced.

So, then she (pediatrician) referred me to an audiologist, like...just here in (town). I always talk about, like, it was a bunch of seniors, like they'd never seen a one year old... Anyway,

so they tried testing her, had no luck and they suggested that I go back to my pediatrician, and ask her for a referral to (pediatric hospital). (Anna's Mum, child not screened)

Another parent talked about the frustration in learning about the hearing loss, as it was very unclear who was to report the details of the hearing loss to the family. She acknowledged that this particular situation may have manifested itself because the child was identified just prior to the implementation of the new provincial UNHS program which may have caused some confusion with respect to roles on the part of some clinics.

And then, I guess the audiologist tried to present it (the diagnosis) in some way, you know, you should talk to IHP, but they didn't really want to give me the final results but somebody needed to give them to you, I couldn't figure out who that person was. They didn't want to or something. And then the audiologist said well, he could maybe have hearing aids if he wants to. And I thought hearing aids, well, of course, let's do that, like let's do that today, and I remember, she put a note on his audiogram, Mother has requested hearing aids. I thought, if it's an option, why is it such a strange thing? (Adam's Mum, child not screened).

The parent continued to describe how the next steps in the process of accessing services were also difficult:

So we talked to (family support worker), she basically gave us the list you know, it can be auditory-verbal therapy, it can be total communication, here are your choices, ...so you

know, she gave us the basics of what we needed to know ...So, I was trying to track all these people down, and they're all hard to reach...and I'm leaving messages like, the kid is getting hearing aids next week and I don't know what to do, somebody needs to help me with, you know what do we do when we turn them on or what do you suggest or whatever. (Adam's Mum, child not screened)

This early identification process in audiology seemed to present less frustration and confusion for parents of screened children than for those of referred children. However, a concern was expressed that the role of the family support worker (typically a social worker) in the new Ontario Infant Hearing Program (IHP) appeared to be unclear and in some cases an entirely separate service from the clinic that identified the hearing loss. These parents generally felt that more technical information should have been available at that early time and that these details were missing in the initial contacts with social support services. However, all parents felt that the information provided on communication options was useful.

I wish the social worker knows more about hearing loss from the technical point of view The lady who worked with us, she didn't quite understand the...difference, because there are different kinds of hearing loss...not only the different approaches, yeah, in the very beginning, I like to know more about the type of her hearing loss and ...what are the signs or problems that I should watch for. Yeah, we didn't get that information at first. For example, the social worker from the IHP, it seems that the team at (hospital), they don't

work with the IHP, they don't work closely. So, it seems that there was a barrier between them...they could...communicate more with each other. It was, it was a bit confusing... (Ellen's Mum, child screened)

However, for other parents, whether their children were screened or not, there appeared to be a more seamless process between the IHP social support service and the diagnostic service at the hospital clinic. It is important to note that some children in this sample were identified during the first year of the IHP program and full services may not have been implemented in all regions. Nonetheless, these parents' comments reflect the importance of receiving essential details about the hearing loss and the path to rehabilitation services.

Effects on the Process of Care

Another potential benefit of a systematic screening program for families may be easy access to a system of rehabilitative care following the identification. Access to services after learning about the diagnosis appeared to be extremely variable, by place, by region, by family and patterns were difficult to establish. The transition to rehabilitation seemed to be related more to the professional with whom the parent happened to be in contact than any characteristic of the health care services. While a smooth transition to care would be expected to be an ancillary benefit of an infant screening program this did not necessarily prove to be the case in these parents' experiences.

Intervention services: The gaps are many and varied, but principally seemed to be due to the fact that providers were not adequately prepared to help families access comprehensive coordinated services. Other parents expressed concerns that they were not provided with information on the range of available services, both public and private. One parent described her experiences calling services to find out where to find help. Some parents found out how to access rehabilitation through the clinic where the hearing loss was identified, some through parents group, and others through the internet (world wide web). For other parents, there was a desire to receive more information about the child's prognosis for speech and language development. No clear pattern of difference emerged amongst the parents of screened or unscreened children. There was no clear indication that these structural aspects of the care system had improved for children who were screened compared to children who were referred. In fact, they were expressed in remarkably similar ways by parents from both experiences.

But the whole process after that was crazy...it was really frustrating, to get funding (for hearing aids) was really frustrating, you know. Yeah, it was hard to get and we had to get, you know, our doctor to approve that, what he was eligible for, ...it was a lot of paper work. ...We needed to know what was out there, ...we had never dealt with a hearing loss before. (Ryan's Dad, child screened)

I think, um, the two things that we were missing was, were first of all a global perspective on this whole process where someone would have said this is how things typically work out

that you have a process where a child is diagnosed, they get some kind of equipment to help them, to get sound, therapy is recommended because we see better results in kids who have therapy than those who don't, the child goes through therapy and then the goal is for mainstream schooling situation and we, you know, we deal with the teachers and train them and so on. I mean, just some kind of idea of how our life would play out. (Logan's Mum, child not screened)

However, a clear benefit that emerged for the screened group was that there was not the same sense of urgency with regards to accessing hearing aids and therapy. For the parents of screened children, there was more time to learn about services, as they were not already dealing with a gap or lost time. Consequently, most parents of children who were diagnosed early appeared to express less frustration about this lack of coordination in terms of access to service. Typically, once consistent care was established with audiology and a therapy program in place, parents were generally very satisfied with the services and expertise provided. The problem was in accessing coordinated care, and simply knowing how and where to find the required services.

Discussion

Childhood hearing loss is a disorder that changes the sense of equilibrium in the lives of families. The findings of this study indicate that childhood hearing loss has a major impact on families, whether it is diagnosed in the context of systematic infant hearing screening programs or not, whether it is identified early or late. We found many of the concerns, the

frustrations and the coping aspects for families confronted with the responsibility of caring for a child with hearing loss to be the same regardless of the age of diagnosis or route to identification.

However, this study enabled us to define from parents' perspectives the important benefits of UNHS programs. Consistent with previous reports, there was a clear consensus that screening should be available as an important component of the health system. (Watkin et al, 1995; Magnuson & Hergils, 1999; Clemens et al, 2000). While the prognosis for improved communication development was unequivocally a critical reason why families preferred UNHS over the traditional referral system, we found the stated benefits of early diagnosis and consequences associated with late diagnosis to extend beyond communication outcomes. Although some families felt that their children had been able to catch up in language acquisition despite a later start, they mourned the fact that the child did not have full access to her environment in the early period of life. The positive aspects associated with early diagnosis (or negative aspects related to late diagnosis) shared by parents were related to quality of family life as well as to measurable outcomes in language tests.

Relatively little attention has been paid to socio-emotional aspects for the child and family. In particular, families whose children were diagnosed well beyond the first year of life, following a period of suspicion of hearing loss, expressed that the waiting periods created anxiety, stress, and in some cases frustration and worry about the increased chances of delayed development. These families were often the greatest advocates for the early

identification of hearing loss. In addition to these benefits, the availability of universal hearing screening seems to have an effect on the process of learning about a child's hearing loss. Several families highlighted easy and seamless access to audiological testing as an important potential outcome of identifying children early.

It was interesting to note that, while all parents felt that UNHS should be retained as part of the health system, some families did not feel the urgency to have the child's loss identified as early as targeted by most universal screening initiatives. This appears to be in contradiction to traditional findings and assumptions that hearing loss should be diagnosed and managed by 6 months of life. This concept emerged both from parents of screened and unscreened children.

Qualitative research takes place in a purposeful sampling paradigm in order to elicit specific information on a particular issue. In this sense, the findings are not intended to be generalizable as in quantitative research frameworks. However, detailed information on this sample from four Ontario settings has been provided so that the reader can decide whether such findings are transferable to another context. A small self-selected population such as this diverse group of parents with respect to route to diagnosis, location, degree of loss, and education, provided rich and varied experiences for this analysis. Furthermore, the validity or credibility of this study was enhanced by audiotaping and transcribing the interviews verbatim shortly after data collection. Threats to interpretation were minimized by: 1) debriefing sessions with an interdisciplinary team throughout the study, 2) an audit trail

detailing procedures for data collection and analysis, and 3) a verification of transcripts, codes, and interpretations by a second reviewer.

The findings present perspectives through consumers' experiences which highlight less conventional outcomes and which might be important to consider in evaluating the contribution of infant hearing screening programs. It is important to note that screening of children from this sample took place in the first few years of a province-wide screening program or the screening was conducted through local initiatives prior to the implementation of province-wide services. Therefore, the issues raised around the diagnostic process may be related to early stages of the implementation of the universal screening program. Nevertheless, they do alert us to the importance of this aspect of a screening and management program and serve as a reminder that an effective screening initiative cannot be separated from the subsequent management of the child with hearing loss.

The study builds on previous initiatives to understand how parents experience the process of identification of hearing loss (Tattersall & Young, 2005; Luterman & Kurtzer-White, 1999; Kovarsky et al, 2004; Russ et al, 2004). This research contributes added information about the effects of early or late diagnosis of hearing loss from the perspectives of families which may have important implications for program planners and decision-makers. Program design and evaluation are important components of newly implemented UNHS services. Assessment of the effectiveness of real world interventions is complex and requires a multi-faceted approach. Our study helps to identify important outcomes of universal screening

initiatives in two ways: 1) through parents' views on the benefits related to their children's communication abilities and 2) the ancillary benefits that emerged through the parents' descriptions of learning about hearing loss and their introduction to the system of care for children with hearing loss. Parents' descriptions highlight the important role and interaction of many contextual factors in the health and well-being of children with hearing loss and their families. There has been relatively little research on the impact of the larger health care system on child and family outcomes. Such knowledge can have an impact on resource utilization and guide the distribution of limited resources.

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Table 1. Examples of questions and probes guiding the semi-structured interviews

1. Tell me how you found out about your child's hearing loss.
   Probes:
   How has learning of the hearing loss through screening (or regular referral routes) been beneficial or negative for you and your child?
   How do you think it might be different if your child's hearing had not been screened (or had been screened)?

2. What impact do you think screening (or not) will have on you and your child?
   Probes:
   How are things better/worse for your child because he/she was diagnosed early?
   How are things better/worse for you and your family because of early diagnosis?

Table 2. Families' perspectives of key issues associated with early and late diagnosis

Issue (N)	Families' typical views or experiences	
	Early diagnosis	Late diagnosis
Bonding with child (7)	No time to enjoy child before diagnosis	Time to adjust to hearing loss and bond with child
Communication development (16)	Better prognosis for speech-language development	Concerns about negative effects on speech-language development
Hearing access (8)	Early access to hearing and participation	Missed opportunities to hear and participate
Regret (7)	No regret about parent behavior	Guilt about missing the problem
Stress around language gap (10)	No sense of urgency - time to explore options	Urgent need to catch up and stress of lost time
Diagnostic process (7)	Seamless transition to audiology	Frustration with delay in accessing audiology
Intervention services (6)	Sometimes lack of smooth transition to intervention services	Sometimes lack of smooth transition to intervention services

(N) represents the number of families (of the total 17 families) identifying this issue during the interviews.

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CHAPTER 4

PARENTS' NEEDS FOLLOWING IDENTIFICATION OF CHILDHOOD HEARING LOSS[1]

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Running head: Childhood Hearing Loss

Parents' Needs Following Identification of Childhood Hearing Loss

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Abstract

Purpose: Appropriate support for families of children diagnosed with hearing impairment may have a direct impact on the success of Early Hearing Detection and Intervention programs in reducing the negative effects of permanent hearing loss. We conducted a qualitative study to explore parents' needs after learning of their child's hearing loss to better understand the important components of service delivery from families' perspectives.

Method: Semi-structured interviews were conducted with 17 families (21 parents) of preschool children in four centers in Ontario, Canada. Both parents of children exposed to neonatal screening and those identified through traditional referral routes participated. We asked parents to share their perceptions of the strengths and gaps in the care system.

Results: Although the majority of parents were satisfied with the range and quality of audiology and therapy services available, they identified gaps in the areas of service coordination, availability of information, and the integration of social service and parent support into the system. Access to audiology services appear to have been facilitated for children who were systematically screened.

Conclusions: The findings provide insights into the services most valued by families. These findings highlight the importance of eliciting parents' perspectives in designing optimal care models for children and families.

Key words: family friendly, hearing services, family-centred, early intervention, early detection

Universal newborn hearing screening (UNHS) has attracted worldwide attention as a public policy intervention aimed at improving outcomes for children with hearing loss and their families. Population-based newborn hearing screening gained momentum as an intervention in the 1990s due to new and efficient screening technologies and the realization that high-risk screening initiatives effectively identified only 40-50% of infants with hearing loss. (Durieux-Smith & Whittingham, 2000; Davis et al., 1997). Studies supporting the benefits of UNHS in improving communication development garnered interest in establishing UNHS as part of comprehensive Early Hearing Detection and Intervention (EHDI) programs (Yoshinaga-Itano, Sedley, Coulter, & Mehl, 1998).

The concept of screening of disease is endorsed by the World Health Organization when an effective treatment program is available (Strong, Wald, Miller, & Alwan, 2005). It is well recognized that UNHS constitutes the first step in a comprehensive system of care aimed at preventing or reducing the negative consequences of childhood hearing loss. Early detection must be followed by intervention including technology and rehabilitation to enable the child to participate in society. In view of the prevalence of UNHS initiatives and a recognition of the lack of documented best practices for infant hearing services, a consensus panel was convened in 2001 to develop a position statement regarding management principles and to discuss state-of-the-art practices for infants with hearing loss (Jerger, Roeser, & Tobey, 2001). The panel recommended that newborn hearing screening be embedded in a system of comprehensive services for infants which include the identification of hearing loss, family counseling, selection and fitting of technology, management and counseling. Recently, the Canadian Working Group on Childhood Hearing also

recommended that management of childhood hearing loss be addressed in a comprehensive, family-oriented, service delivery model (Canadian Working Group on Childhood Hearing, 2005).

Despite these efforts and the recognition that broad care models are desirable, several researchers have raised concerns about follow-up services to UNHS. Recent reports have indicated that although UNHS can be effective as a path to intervention, the capacity of population-based screening to improve longer-term development outcomes is uncertain (Vohr, Moore, & Tucker, 2002; Centers for Disease Control and Prevention (CDC), 2003; White, 2003). Historically, services for the management of pediatric hearing loss evolved in a piecemeal fashion with responsibilities assumed by various agencies such as health, education, and social services. Many existing treatment or intervention services have evolved prior to the current focus on UNHS, the availability of current hearing technology and the emphasis on evidence-based practice. Research undertaken in the United Kingdom in preparation for a new UNHS program, revealed several barriers to the implementation of UNHS both from professional and parents' perspectives (Robinshaw & Evans, 2003; Bamford et al., 2001). In the United States, the referral of families for early intervention has been described as the weakest link in the EHDI system (Sass-Lehrer, 2004).

Family-centred care is advocated as an important characteristic of best practices for infant hearing health services from the identification through the intervention process (Harrison & Roush, 2003; Robinshaw & Evans, 2003; Gravel & McCaughey, 2004). It has been proposed that an intervention approach that values partnerships with families and promotes self-efficacy in parents may result in higher rates of follow-through, greater

participation in early intervention and improved outcomes for children with hearing loss (Sass-Lehrer, 2004). The importance of family involvement was found in one study to be an important predictor of communication outcome in children by 5 years of age (Moeller, 2000). Process variables such as quality of relationships between professional and parents and parents' sense of efficacy have been identified as potential key factors in achieving positive outcomes (Calderon, 2000; Eriks-Brophy et al., 2006).

Likewise in Canada, as the country embarks on population-based newborn hearing screening the notion of family-centred care also prevails (Hyde, 2005). In the province of Ontario, where a universal infant hearing and communication development program was implemented in 2002, a major goal is to: "provide parents of deaf or hard of hearing infants with the services their children need in order to develop communication and language skills, and to give them the best start in life" (Government of Ontario, 2006). Aligning services with parents' expectations and preferences may be an important determinant of outcome in early intervention services. Despite the recent attention accorded to UNHS, relatively little emphasis has been placed on parents' views of their needs after the detection of hearing loss and the service models needed to address them. Attributes of service models such as parental involvement in intervention and quality of therapy, may be critical factors in determining the effectiveness of an intervention program (Yoshinaga-Itano, 2004).

With a newly implemented UNHS program in the province of Ontario, Canada, this study was undertaken to better understand, from the perspective of parents of young children with hearing loss, the supports required to give their children the best start in life. The data reported here are from a larger study designed to learn about parents' perspectives on the

benefits of early hearing detection as well as their needs after learning of a hearing disorder. The results of the first objective have been recently reported (Fitzpatrick, Graham, Durieux-Smith, Angus, & Coyle, in press) and the latter objective forms the basis of this paper. Specifically, the study sought to address the following objectives: 1) identify parents' needs during their experience in caring for a young child with hearing loss through the spectrum of care from identification to fitting of amplification to rehabilitation, 2) identify the strengths and gaps in the existing system of care, and 3) outline the important attributes of childhood hearing services from the perspective of families.

Participants and Method

This study applied qualitative research techniques in the form of semi-structured interviews to examine parents' needs following diagnosis of a child's hearing loss. In adopting a qualitative research approach, we sought an identification of the elements of care that parents considered most important rather than setting a priori expectations.

Parents from four intervention programs in three cities in the province of Ontario, Canada, were invited to participate. Audiology services were provided through hospital and university clinics and therapy was provided in a variety of settings including hospital clinics, community and home-based settings. The majority of participants were selected from a sample of 65 parents who were enrolled in a multi-center longitudinal study investigating the benefits of systematic screening for children with permanent hearing loss (Fitzpatrick et al, manuscript submitted). Families of children under age 5, with early onset permanent hearing loss (before 6 months of age), enrolled in oral communication development

programs met the study inclusion criteria. The project was approved by all four institutional review boards and written informed consent was obtained from all participants.

For the qualitative research study, we purposefully constructed a maximum variation sample of 17 families to reflect a range of child and family characteristics that could influence parents' views on services. In selecting the sample, variability was sought in route to identification (systematic screening or traditional referral practices) severity of child's hearing loss, type of hearing devices, distance to therapy program, and parental education. Consistent with qualitative research, the emerging analysis guided the number of participants interviewed and recruitment was discontinued when data saturation was reached, that is, no new themes were apparent in the interview data.

Procedure

We conducted individual in-person interviews using a semi-structured interview guide. The questions related to parents' perceptions of their needs after identification and the primary components of a service model for children with hearing loss and their families. Participants were also asked for their vision of a service model if they were redesigning the health care package for families and children with hearing loss. The two main questions were: 1) What were your needs following diagnosis of your child's hearing loss? 2) If you could redesign the system, what would be the important components of a model? The participants were encouraged to think about the strengths and gaps in the existing services to assist them in formulating their ideas. As the interviews progressed and data were analyzed,

questions were added to elucidate parents' views on topics from previous interviews with other parents, particularly with respect to service coordination and a team approach to care.

Data collection was conducted over a period of 4 months, with all interviews conducted by the same interviewer. The interviewer was a researcher with a background in audiology/therapy and several years experience as a clinician/program manager in audiology services. All but one interview took place in the families' home environment. The parents were encouraged to share their perspectives according to their level of comfort and to focus on issues of importance to them based on their experiences in caring for their child. They were encouraged to support their responses with examples from their personal experiences. All interviews were audio-taped and transcribed verbatim shortly following each meeting with one exception due to a technical equipment problem; the content of this interview was immediately summarized in an audio-recording by the interviewer.

Seventeen interview transcripts supplemented by interviewer notes and memos provided the dataset for analysis. Data collection and analyses proceeded in an iterative manner with new data guiding and confirming the developing analysis and guiding future interviews. The data were analyzed by the interviewer using the software package Atlas-*ti* and adopting techniques described by Strauss & Corbin (1998). Each line of the interview was carefully read and scrutinized to identify key concepts which were compared across interview scripts. Initial coding was broad and similarly coded data were then grouped into categories. Memos were written throughout the coding process to summarize the reflections and interpretations of the data as the analysis proceeded. Data collection was discontinued when incoming data did not appear to generate new insights. A second reviewer with

expertise in pediatric hearing loss independently reviewed the data to verify the categories/concepts identified. These categories were collapsed into themes to synthesize parents' views.

Results

Participants

Demographics information was collected from the participants to better understand family characteristics and potential factors which might influence their views. The characteristics of the children and families are detailed below.

At their discretion, parents participated in the interviews individually or with a significant other person. All were hearing parents and the 17 interviews involved a total of 21 individuals, with participation as follows: 10 mothers alone, 1 grandmother (the child's main caregiver), 2 fathers, 4 mothers and fathers together. Two of the families also had a second child with a hearing loss. Fifteen of the 17 families lived in an urban center. The majority of parents (11 of 17) had experienced the diagnostic process for a period of less than 3 years (range 11 months to 44 months) prior to participating in the interviews.

Seven babies had undergone a systematic hearing screening procedure (5 universal screening, 2 high risk screening) and ten had been referred for hearing assessments through traditional referral routes, typically through the family doctor, because of parent suspicion of a problem. The average age of diagnosis of all children in the sample was 16.1 months (range 0.8 to 41.9 months). Nine children were diagnosed before 12 months of age, six of whom had undergone systematic hearing screening. The sample was selected to include

children with varying degrees of hearing loss and different hearing devices. The sample included more children with severe and profound degrees of hearing loss consistent with participation in the larger study from which the majority of families were recruited. Severity of hearing loss was distributed as follows: 1 mild, 5 moderate, 6 severe and 5 profound hearing losses. Ten children used hearing aids and seven had received cochlear implants at the time of the interview. All 17 parents reported post-secondary education, including 12 who had completed university level studies and 5 with college or trade level education (average of 16.8 years of education, range 13-21 years). Fourteen of these 17 families, who had selected an oral communication rehabilitation approach, were enrolled in auditory-verbal therapy, reflecting the primary rehabilitation choice for families in the province of Ontario.

Parents' Views on Needs and Service Provision

The profound effect of a childhood hearing loss on the family was captured in the experiences and decisions made by parents in the early stages. As illustrated in two mothers' words below, parents described the disorder as a phenomenon that impacts not only the child but changes their lives and affects such family decisions as career, finances, and place of residence.

So, I think the impact has been a bit deflating to his childhood. And, of course, the stress to us, it's changed everything about, our decisions, about how he's going to be educated, about whether I'm going back to work or not, everything has been impacted. (Interview 7)

Once you make the decision, you want to know where your future lies. Like we quit our jobs and changed our lives and came to Canada for him. (Interview 10)

Since permanent childhood hearing loss is a lifelong condition for the child, it brings with it long-term requirements for family support in a number of areas. The views related to needs and service provision that emerged from the data were summarized into four key discussion themes: 1) components of service model, 2) coordinated care, 3) parent contact, and 4) information needs. Although these themes were frequently overlapping and inter-related they have been separated to facilitate the synthesis of the data and are elaborated in the following sections.

Components of Service

Screening: As this aspect of the interview addressed parents' service needs following the identification of hearing loss, the discussion of screening as a part of the service model was not a key focus. However, several families of children who did not benefit from early diagnosis expressed strong opinions about the importance of including this component in the overall package of services funded for children with hearing loss. All parents expressed the view that access to screening was an important part of the overall service delivery model for children with hearing loss.

Intervention: Audiology and therapy services were unanimously identified as vital components of the service model for children with hearing disorders regardless of the severity of hearing loss. A few parents expressed some dissatisfaction regarding timely access to pediatric audiology services during the initial identification stage; this appeared to apply to children diagnosed prior to the implementation of the universal screening program and has been elaborated in a previous report (Fitzpatrick et al, in press) as a potential benefit of infant hearing screening initiatives. For some parents, consistent with recent reports, there was dissatisfaction around communication of the diagnosis which appeared to emanate from the manner in which the news was delivered (Young & Tattersall, 2005).

Once parents progressed beyond the initial difficult stages, and the intervention program including amplification and therapy was established, the overwhelming majority described a high degree of satisfaction with the audiology and therapy services provided. Parents in this study were enrolled in individual family-oriented services focused on oral communication development, (14 auditory-verbal and 3 oral) at community, hospital or home-based clinics. Distance to service was a concern for some parents particularly those from rural or large metropolitan areas who were required to travel to a clinic for some or all services. It is noteworthy that a few parents of children who used hearing aids expressed the view that the focus on cochlear implants in hospital clinics led to a perception that their child's less severe hearing loss was somehow less important.

Notwithstanding these concerns, overall, parents spoke of being fortunate to live in a city or country with high quality services which effectively guided them in developing their child's oral communication skills. Throughout the interviews, parents talked frequently and

positively about the therapy, likely because of the continuing contact with the therapist who typically provided weekly sessions. The relationship and guidance from the therapist and the weekly therapy sessions were described as one of the most important factors in developing their child's language.

I'd have to say ... the services that we have had, have been remarkable, audiology and audio-verbal therapy, so really (there is) very little to change. And so three and a half years into this therapy, she looks forward to going...and...I attribute that very much to the quality of the service there and how they keep these sessions entertaining. (Interview 4)

Other service components: In addition to audiology and therapy, parents talked most frequently about social work services, however, this was usually described as ancillary to the intervention services of audiology and rehabilitation. The majority of parents appeared to have had contact with a social support worker at or shortly after learning of the hearing loss. Few parents spoke of having had exposure to psychology services except those considered for cochlear implantation and it appeared to be viewed as a discrete one-time developmental assessment for cochlear implantation with no expectation or apparent need for follow-up. For the parents, whose children had been screened, all appeared to have encountered social support services as part of the provincial infant hearing program. For the others, it seemed to vary depending on the clinic at which the hearing loss had been identified. There was great variability in parents' experiences and views on the need and value of social service supports, generally provided by social workers or family support workers. For some

parents, the initial encounters with social support appeared to be confusing as the service was separate from the audiology and/or therapy program. Parents did not present a consistent view of the role of the family or social support worker and how the individual was integrated into the process of care. A few parents felt that the social worker did not have sufficient technical or medical information about hearing loss and its consequences to provide needed counseling in the early stage. Others spoke very highly of the need for such a service in helping to navigate and access the myriad of services and financial support available. The reflections of two parents who were provided with social support shortly after the diagnosis illustrate the wide variability in parents' experiences and perceptions on the need and value for this service.

I think everyone needs that support worker, she was phenomenal, without her, I wouldn't know what to do...She was the one who set us up with speech-language pathology and everybody. Once we got the support worker, everything happened quickly, yeah. Yeah, we were trying to get everything, so I definitely think families should be hooked up with a support worker. (Interview 17)

We saw her (parent support worker) and she gave us a huge binder with many dividers. Ultimately, I had a list of providers for auditory-verbal therapy, a list of providers for sign language, a list of providers for total communication options and other than that it was empty and not very useful. ... And I guess the referral system worked but then I was finding

that either they weren't getting the information from her fast enough, or fast enough in my mind...it wasn't feeling like a lot of support was coming from there anymore. (Interview 14)

Other services that were not typically viewed as part of hearing health services were described as difficult to access, for example, developmental pediatrician, and some parents pointed out that the overall developmental needs of the child with hearing loss should be considered. Other gaps in care related primarily to funding to support equipment purchases and therapy travel costs. Parents felt that this practical aspect of care presented a barrier to optimal service and had not received sufficient attention at a policy level.

It's the typical middle class syndrome where we are still paying for services, that I think should be free, only because this is something she needs to function on a daily basis. (Interview 3)

Coordinated Service

Access to coordinated care was an important theme that emerged early in data collection and warranted further inquiry during subsequent interviews. The above quote exemplifies the frustration of parents when they perceive a system comprised of individual components where structural and operational issues present barriers. When services were viewed as fragmented, this appeared to affect, in particular, the process from diagnosis to intervention. Although, not frequent, this breakdown was perceived by families who experienced the diagnosis with and without systematic screening. Their experiences

suggested that a clear picture of the care pathway was missing and they were left without support until they accessed the therapy service. Parents who had less desirable experiences during the early stages, appeared to be more mistrusting of providers. During the interviews, these parents reflected back to their initial experience which appeared to act as a catalyst for their views on current care practices.

I would really like to deal with one organization... I wish I could deal with one organization, rather than two, or at least having these two on a good path. I wish, you know, whatever organizations are out there, they need to be working together. I wish there was a consensus of what people with hearing losses need because we want as many avenues for our child as possible. (Interview 17)

What I would like to see is that you get to have a meeting with the ENT, the audiologist, and an auditory-verbal therapist and a social worker...with everybody together in one room saying, here's where our goals are right now for your child and here are the kinds of tests we're going to run in order to determine what her future needs might be and this is what the process looks like – a flow chart if you will. (Interview 7)

The notion of coordinated team services did not have the same meaning for all parents as illustrated by the quotes below. For some, it appeared to be related to the co-location of the various health providers but to others, coordinated care depended on the communication and the shared vision of the providers most involved in the child's care.

Parents' endorsement of different models of service provision appeared to be based on their positive experiences with a model. In some cases, it likely reflected the reality of living in an urban or large metropolitan area at considerable distance from the child's audiology center. Some parents described audiology and therapy services that worked together effectively whether or not they were co-located, while in other cases, audiology and therapy were described as two distinct programs. For some families, there was a perception that one team member, usually a therapist who adopted a coordinator role created a successful "virtual" team. The therapist, with whom parents typically interacted on a weekly basis, was an important link with other team professionals.

We can see our ENT, our auditory-verbal therapist, our audiologist and the cochlear implant specialist all in one spot. We don't need to go whipping around all over the place and go to different spots. Where in other places, ..., it's very fragmented, where here, we're very fortunate, it's all in one spot and they all share information. (Interview 3)

It (the model) works very perfectly because everything just seems to gel together because ... you go to the audiologist, the speech therapist, the family doctor, the school, itinerant teacher, they have a different approach but the content and the objectives are the same. ...The therapist is very involved with him, she would normally call around to find out what's happening....everybody is kept on pace as to what is happening. She's (therapist) probably the unofficial case manager. (Interview 9)

... because we get to see the therapist most often so they know us, they can see the progress or problems. I still think the therapist is a window to see what's going on for other people in the team. We can't get to see the audiologist without an appointment, but I just thought if the therapist...relays the problems or parents' concerns to the audiologist more, and doctors more, that would be helpful. (Interview 1)

Support from Other Parents

Parent contact was acknowledged by all families as a useful adjunct to the services provided through typical health care channels. Families who strongly embraced parent support suggested that it should be an integral part of the health care system. They felt that current practice models offered this support in an ad hoc manner leaving it as the sole responsibility of a volunteer parents' group. Parent support groups or access to parental input seemed to fulfill several needs including knowledge sharing, practical information about hearing devices and community resources, prognostic information, and hope. For some parents, it also filled an emotional support need beyond what could be offered by psycho-social providers in health care. A few parents commented that it supported healthy development of their child as children saw peers with the same type of communication disorder.

And sometimes parents trust other families better than they trust professionals or understand them better, and I think, yes, because in the long-term it impacts a parent's

decision-making for their child's services and that will impact the child's outcomes as well. (Interview 7)

I guess one of the things that we wanted that you really want right from the beginning is you want to speak to parents that have had children implanted and get their feedback on what they think it's done for their child. And at the same time, we wanted to speak to adult users because children can't tell you what the hearing is. (Interview 4)

Information Needs

The requirement for information was a subject that filtered through several aspects of the discussion. From the interview data, parents' information requests could be classified into several domains: 1) hearing loss specific-information (e.g., etiology, severity of loss, hearing aid and cochlear implant technology), 2) therapy related information including therapy options and other resources in the community and 3) prognosis information.

Hearing-specific information: The majority of parents reported satisfaction with the medical and technical aspects of hearing loss received through their clinics. Parents referred to a strong need for information at the beginning of the diagnosis and a need for ongoing up-to-date information throughout the continuum of care particularly in the technology domain, e.g. bilateral cochlear implantation. There were no obvious differences in parents' views across the spectrum of hearing loss or route to identification of hearing loss. Families of children with mild to moderate hearing losses talked about the importance of information as

much as parents of children with profound hearing impairments. There were also no perceptible differences in the information needs identified by parents of children who were diagnosed with hearing impairment in the presence or absence of systematic screening programs.

I will say it's overwhelming in the beginning, you want the information, however, at the same time you're trying to take in so much information, that it is a lot to take in. But they did provide us with all sorts of literature, videos, support through our auditory-verbal therapist, through the audiologist, regular appointments to keep us in contact. (Interview 3)

Therapy and resource options: The large majority of parents expressed that information on various therapy options had been adequately presented either by the clinic audiologist/therapist or a social support worker. However, several families felt they were left without adequate support in locating various resources. Information on options for resources in the community such as home tutoring, additional support through the educational system were however, less positively viewed.

The (Program) was very diplomatic in giving us information. I sort of figured out what was going on after the fact but I appreciated how they did it. They never said this is what you should do...they sort of kept always making sure, we were aware of things. (Interview 13)

Like I said, first there was a lot of figuring things out so it would be nice to have somebody so that you know everything that's available, that way you can start figuring out what you need for your child. (Interview 6)

For parents to share information and tips...we've got lots of tips on equipment, even for things like income tax purposes. We found out about a tutor service through the parent group, we didn't know about it. So I just think some of these steps maybe need to be in place. (Interview 3)

I asked the clinic (about other services) because I did want something for the other week because I found that therapy kind of re-energized me, sort of focused me... So that's when my a-v therapist brought up (other program) and home visits so that was the only way I found out about that and I was a little miffed ... because it was never brought to my attention. (Interview 11)

Information on child's prognosis for oral communication development: In the early stages of learning about the hearing loss, information about the child's prognosis was often lacking. The importance of this was described by one parent as follows:

I guess we wanted to know what his loss would translate into, so we knew he had a loss, and we knew the severity of the loss...we had all the details, but what we really wanted to know is what does this really mean...Does it mean that he'll grow up having to learn sign or will

he be able to just use, language, spoken language, regular classroom placement? (Interview 6)

The following extracts from interviews reveal some of the many ways parents expressed this desire for additional information on their child's potential to develop spoken communication. Essentially, parents wanted the best available evidence supporting intervention outcomes.

You have to see the success, maybe that's what I'll highlight to you.... So my husband was reading about how high the unemployment rate was for people who are deaf and some of the other negatives and the stigma attached to that, so we needed to kind of see the other side to know that there was hope. (Interview 4)

Not that we want to be, like comparing, but you want to see where he <u>should</u> be at,...even though we know there are so many things involved. ...We were searching the internet to find stories of kids being implanted because we were trying to get him implanted as early as possible. (Interview 10)

One of the other things we found just kind of somewhat lacking is like, some of the research results, for example,...most kids in this range tend to learn these sounds of these types of words by this age ...they have that for normal kids. (Interview 10)

Parents' views on access to information: Information sources included health care/education providers, printed materials such as brochures, books, and journals, media such as videotapes of children, the internet, and personal communication primarily with other families of children with hearing loss. While parents generally felt that their health and education providers were well informed and willing to share information, they offered concrete suggestions on how the health service could ensure that information both on programs and technical-medical is available in an ongoing, up-to-date way.

I'd almost have a kit, maybe literature, up-to-date literature with books and videos, not too too much, but to have some things there for the beginning so that families don't need to find it...it's right there for them when they need it and they want it. So that's a huge thing, I think that I would ask, only because we felt we were kind of running for everything we needed and maybe knowledge is power. (Interview 3)

Parents described a certain comfort in being able to consult multiple sources for information including, professionals, books and journals, videos, and the internet. The internet emerged as a prominent source of information for parents, both for locating resources (e.g., clinic programs, parent support groups) and up to date technical information. A parent with a child identified prior to the implementation of universal screening, described how she used the internet to locate a rehabilitation service. Others described using the internet as a resource for lesson plans and a link to various professionals to obtain different perspectives on therapy options. Parents of children with cochlear implants described

manufacturer's websites and online discussion groups as a valuable source of information. Some parents judged that the internet was not sufficiently exploited as a resource by the health system.

...We got back up to the cottage (after the diagnosis), and so we logged onto the internet and just started reading. And we went to the (Program) website, I think that's where we got the most amount of information, read the testimonials from the parents....we phoned them and talked to them about it and that was our first link. Now, I know of all of these other services and programs but that was my link. (Interview 13)

Discussion

Population-based newborn hearing screening presents new opportunities for children with hearing loss and their families. However, there is a realization that the success of newborn screening is largely dependent on the implementation of adequate support programs for children and families (Yoshinaga-Itano, 2004; Hyde, 2005; White, 2003). Using a qualitative research approach, this study identified parents' views on service needs for childhood hearing loss in the context of a newly implemented universal screening program. Several broad conclusions about parents' needs following diagnosis of a child's hearing loss were extracted from the 17 family interviews. In general, parents value quality audiology and therapy services as core support services.

While the amount and location of therapy programs were problematic for some families, the majority were very satisfied with the guidance and professional caliber of these

services. In addition to these core services, families identified a variety of other desirable supports from the publicly funded care system including social service support, financial support for devices and travel, and contact with other parents who had experienced the process of hearing loss. Two systemic problems appeared to affect the process of receiving appropriate support for some families in the early stages of diagnosis: 1) knowledge of and access to the various services/resources that were available and 2) the lack of coordination within clinics and between various clinic providers and agencies. With time, parents seemed to learn how to navigate the system and were generally very satisfied with the ongoing services that were provided.

The finding that parents seek hearing-specific medical and technology details as well as intervention-related information is consistent with previous research (DesGeorges, 2003; Robinshaw & Evans, 2003; Luterman & Kurtzer-White, 1999). An additional important finding of our research which has received cursory attention in the literature was the desire of many parents to have some notion of the prognosis for a child with a specific hearing loss and the sense that this information was lacking. While this need has been mentioned in previous studies (Kurtzer-White & Luterman, 2003; Neuss, in press), it was a particularly noticeable theme in this study and may reflect the perceptions of educated and informed consumers. It is unclear from the literature whether this information is not provided because clinicians are unaware that parents desire such information, are uncomfortable discussing prognostic information, or simply do not have evidence-based prognostic indicators.

While parents generally felt that the providers they encountered were highly knowledgeable about hearing loss, they also commented that they would like to receive

professional guidance in seeking information beyond that offered by clinic providers. The internet emerged as an importance access point for parents, however several families expressed that this source was not sufficiently supported and maximized by providers in health services. The finding that parents rely heavily on the internet for information emphasizes the shift in how patients seek information as described by other health researchers (Hardyman, Hardy, Brodie, & Stephens, 2006). The benefits of receiving timely and ongoing information were described as increased involvement in and more-informed decision-making (e.g., regarding cochlear implantation), and greater satisfaction with services provided. In other areas of health such as cancer care, information seeking has been demonstrated to play a critical role in individuals' efforts to cope with the negative impacts on quality of life associated with a disorder (Rutten, Arora, Bakos, Aziz, & Rowland, 2005).

Although this sample drawn from four Ontario centers included a diverse group of parents, their needs were remarkably similar. No clear trends emerged based on severity of hearing loss, type of sensory device or route to identification. At present, there are clearly major differences among centers on how they approach and coordinate care, however, these interviews suggested that no one model is necessarily ideal for all families. As research in other fields has demonstrated, patients' views on appropriate health care appear to be largely impacted by that which is familiar to them (Porter & Macintyre, 1984). However, there are clearly several important attributes of services from the perspectives of families across the system, namely, high quality therapy and audiology services, well coordinated care with communication between involved professionals, and ongoing easy access to up-to-date information. In summary, this study demonstrated that although parents are typically

satisfied with the quality of intervention services, certain gaps in how the diagnostic information is delivered, how parents locate services and resources, and access to ongoing information need to be addressed in implementing pediatric hearing care programs that meet consumer preferences. Services that meet parents' preferences may better engage families in the care process and therefore lead to better developmental outcomes (Moeller, 2000; Calderon, 2000).

The findings complement and expand on previous work by illuminating more specifically the needs and preferences of parents of very recently diagnosed children. Our findings related to information needs are consistent with previous research that has addressed the question of families' wishes after identification of hearing loss (Minchom, Shepherd, White, Hill, & Lund, 2003; Rousch, 2000; Luterman & Kurtzer-White, 1999). While parents in our study welcomed information about various communication options, (e.g., oral and signing programs), in contrast to previous reports, this did not emerge as an important theme in the interviews (DesGeorges, 2003; Rousch, 2000). This may be due to the fact that the information was readily available or, as stated by some parents, that the decision about choosing an oral communication option was clear from the time of diagnosis. In our research, parents were most concerned with receiving guidance on how to access the various oral programs that were available. Our findings also differed from studies emanating from the United States, where the physician or medical home plays a role in managing the child with hearing loss (Gravel & McCaughey, 2004). There was little discussion of physician involvement for screened children beyond the initial referral phase for families of children identified through traditional referral routes. This may simply

reflect a difference in how services are organized in the two countries with audiology and therapy responsible for management of the child with hearing loss separately from primary health care.

In our research, parents' views on the initial diagnostic period are consistent with recent qualitative research findings in the United Kingdom (UK) where professional manner and communication emerged as a significant predictor of experience during the diagnostic process (Tattersall & Young, 2005). In our study, although there was a high level of satisfaction with audiologic services, the initial delivery of information also emerged as a weakness in the system for some families. A broad overview of parents' service needs over time has received limited attention in pediatric hearing despite the current emphasis on patient perceptions in health care and on involving patients in the evaluation of health care interventions. A comprehensive study on parents' views of service provision conducted in 1999 in the UK in preparation for newborn hearing screening services found that parents valued coordinated services, access to parent support groups and family-centred care (Robinshaw & Evans, 2003). To our knowledge, our study is the first in the Canadian context to examine parents' perceptions of service provision and specifically the perceptions of parents who have selected oral communication development.

A strength of our study is that 17 families from four different clinical programs in the province of Ontario participated, therefore the themes reflect a collective experience rather than that of any one service model. All but two participants were drawn from primarily urban areas so the perspectives of rural families have received less attention. However, the study findings are based on a varied sample of parents of children who differed in severity

of hearing loss, types of hearing devices, route to diagnosis and time since diagnosis. Considerable details and description have been provided so that the reader can decide whether these findings are transferable to other settings. The study results are based on rigorous qualitative techniques whereby one interviewer ensured consistency in data collection. Threats to validity were minimized through regular meetings with an interdisciplinary committee, rigorous data collection, audiotaping and verification of the concepts by a second reviewer.

This study is a starting point for several other important questions related to service models for children and families affected by hearing loss. Through the discussions with families, we endeavored to capture the important characteristics of service models but did not attempt to quantify the relative importance of the various attributes. While factors such as age of identification and severity of hearing loss have been shown to impact communication outcomes, the extent to which health services acts as a determinant of outcome requires further investigation. In future research, we plan to use the data from this study to develop a conjoint analysis questionnaire to quantify parent preferences for the various attributes of care. This can shed light on how to weight these various needs in care models. Second, the needs of families who reside outside major regional centers require further examination. Third, this study was conducted within three to four years after the implementation of a province-wide universal neonatal screening program. As the provincial newborn hearing screening program matures, further research should evaluate whether some of the initial problems and lack of coordinated services have been resolved. Given the proliferation of early detection and intervention programs, continuing research using a

variety of methodological approaches to better understand parents' needs in order to tailor and evaluate programs is of utmost importance. Further approaches should focus on how to implement care models with the active involvement of patients.

Early detection of hearing loss accompanied by optimal intervention services may reduce or even prevent delayed development. The results of this study highlight the value of eliciting parents' perspectives on support services that affect their child and family. Parents' views of appropriate services may have a strong influence on their willingness and ability to be involved in their child's care as well as in decision-making at various times during the care process. The findings from our research can be used in optimizing models of care for children and families, ensuring that parents' perspectives are taken into account throughout the continuum of care. Because effective communication is known to be associated with optimal health outcomes, understanding what, when and how information should be delivered to patients becomes vital to ensuring the delivery of appropriate care. Such knowledge can help tailor programs and services to achieving more effective communication and patient-decision making and perhaps ultimately improved outcomes for children with hearing loss and their families.

This study provides an awareness of parents' needs from their perspectives and their description of current strengths and gaps in the system. From a policy perspective, an understanding of how parents value the components of service delivery can provide insights into the most valued services for families. There has been little attention in the literature on models of service delivery and their impact on child outcomes and parents' motivation and ability to follow treatment programs. Although screening methods are highly prescriptive

and protocol oriented, best practices for intervention services following identification of hearing loss have not been well-defined. One goal of a well-implemented hearing screening program is to offer a coordinated system of care (Hyde, 2005). This study could be used as a starting point in informing the development of quality indicators for program evaluation.

In summary, this study contributes to the growing evidence-base in pediatric hearing health care privileging the consumers' voice – families of children with hearing loss. Our study adds to the understanding of parents' perceptions of their needs in parenting and developing oral communication skills in their young children who have permanent hearing loss. By virtue of its intensive and long-term nature, service provision for young children with hearing loss involves a social contract with families which must be recognized and given priority. Parents want to be recognized as active partners in providing the optimal system of care for their child. These finding support the need to include parents in decision-making about designing programs. Early detection through population screening alone may not be sufficient to improve outcomes unless practices are in place to support and guide parents in facilitating their child's language development. Further understanding of parents' needs may improve the delivery of childhood hearing services and maximize the investment in newborn hearing screening.

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CHAPTER 5

PARENTS' PREFERENCES FOR SERVICES FOR CHILDREN WITH HEARING LOSS: A CONJOINT ANALYSIS STUDY[1]

[1] Formatted for Ear and Hearing

Parents' Preferences For Services For Children With Hearing Loss: A Conjoint Analysis Study

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Abstract

Objective: Early identification of permanent childhood hearing loss through universal newborn hearing screening is rapidly becoming a standard of care. However, it is well recognized that hearing screening must be embedded within a comprehensive system of rehabilitation and parent support services. This study was undertaken with parents of young children with permanent hearing loss to examine their preferences for characteristics associated with intervention services. A secondary goal was to explore whether preferences may differ according to patient subgroups.

Design: Conjoint analysis, a preference-based economic technique, was used to investigate parents' strength of preferences. A cross-sectional survey which consisted of hypothetical clinic scenarios was developed based on information from qualitative interviews with parents. The questionnaire was administered to parents receiving intervention services in the province of Ontario, Canada, shortly after the implementation of a universal hearing screening program. The sample was recruited from three different clinical programs.

Results: A total of 48 respondents completed the questionnaire. The participants varied by screening status of the child (25 screened, 23 not screened), by type of device (23 hearing aids, 25 cochlear implants) and by region. All five characteristics of care that were selected for inclusion in the survey were found to be statistically significant attributes of services: coordinated services, access to parent support, access to information, frequency and location

of services. In this study, parents showed a preference for clinic rather than home-based services. The results also suggest a preference towards once a week therapy services rather than services two to three times weekly. In particular, parents valued service models that consisted of well-coordinated care with access to support from other parents. Differences in respondents according to hearing screening status (screened or unscreened), type of hearing device (hearing aid or cochlear implant) or region (Ottawa or Toronto) did not appear to affect parents' preferences for attributes of care.

Conclusions: Conjoint analysis is a useful technique for quantifying parents' preferences for care. The values expressed by parents provide insights into the aspects of a service model that should receive consideration in the development of programs for young children with hearing loss and their families.

Key words: hearing loss, screening, conjoint analysis, patient preferences, hearing services

Introduction

Early detection of permanent childhood hearing loss has received considerable attention in recent years due to the widespread implementation of population-based newborn screening programs. It is postulated that early detection represents an opportunity for improved and perhaps even age appropriate communication development outcomes for children with hearing loss when screening is embedded in a comprehensive system of care (Jerger, Roeser, & Tobey, 2001; Hyde, 2005). Children and families typically require long-term services, many of which have evolved with a long and complicated history in terms of appropriate therapy methods and practices. The advent of new technologies coupled with newborn hearing screening, has moved care from institutional practices to a greater focus on inclusion with hearing children providing opportunities for children that were not previously available.

Although considerable emphasis has been placed on developing appropriate screening services, far less attention has been accorded to the intervention process and defining the system of care for families of children following diagnosis of hearing loss. Advocates agree that early intervention is crucial to a successful early hearing detection program and that detection without intervention is likely of limited value (Jerger et al., 2001; Canadian Working Group on Childhood Hearing, 2005). The care structure has developed in a patchwork fashion over time prior to the advent of Universal Newborn Hearing Screening (UNHS) programs and consists of numerous arrangements including medical-based services, community health services and other support services. Contrary to previous generations, the implementation of newborn hearing screening programs means that the

majority of families will now enter hearing management programs well before the child's first year. It is therefore imperative to reconsider what constitutes an effective intervention program for families of young children.

Traditionally, patients have had minimal involvement in decision-making related to the organization of health care services. However, in recent years, a greater consideration of patient views (preferences) and consumer involvement in health care planning has been advocated (Ryan & Farrar, 2000). Such involvement is however, only of benefit to planning if the input from those affected by the services is informative and scientifically sound. However, measuring patient preferences for the various components of health care service models is undoubtedly complex. One technique for quantifying patient preferences which has gained momentum in investigating other health care questions is that of discrete-choice experimentation or conjoint analysis (CA). The CA approach which was originally developed for consumer studies, particularly marketing, is increasingly used in medicine to assess patients' preferences (Albus, Schmeisser, Salzberger, et al., 2005). Conjoint analysis is a survey method encompassing data collection and analysis which holds a theoretical basis in mathematical psychology (Ryan et al., 2000). Early applications of CA in health occurred in the 1990s when the technique was applied to elicit patient preferences in health research areas such as doctor-patient relationships, outcomes and health care services (Vick & Scott, 1998; Ryan, 1999; Ryan & Farrar, 2000).

Conjoint analysis has emerged as an assessment procedure which allows individual opinions and preferences to be considered in a quantifiable manner. Underlying CA is the theory that any product, for example, a given health care service, can be described by its

characteristics, frequently referred to as attributes in conjoint analysis research. The extent to which an individual values a service depends on the levels of these characteristics. In particular, CA can provide information to estimate the relative importance people attribute to certain aspects of care, measure how individuals are willing to trade between these characteristics (attributes) and estimate the overall satisfaction or utility that individuals gain from various forms of health services provision. This information can assist decision-makers in quantifying the individual impact of various characteristics on overall patient satisfaction, in setting priorities for various health care services, and in making decisions about the most efficient way to provide a service.

In hearing health care, qualitative and survey research has been conducted to better understand parents' experiences and needs at the time of diagnosis and early intervention (Rousch, 2000; Tattersall & Young, 2005; Robinshaw & Evans, 2003). However, previous studies have not investigated the effect of various characteristics on how parents value the services. Little is known about the relative value parents place on the characteristics of services, for example, home-based versus clinic-based service, psycho-social services as part of the care package, or co-location of services. Typically, health care providers, partially influenced by availability of resources, decide how to deliver services to families of children with hearing loss.

This research was undertaken in the context of a broader study aimed at understanding parents' views on the benefits of early detection of childhood hearing loss and their needs in managing the hearing loss. Specifically, we applied conjoint analysis to examine parent preferences for services available through a publicly funded health care

system for newly identified children with hearing loss. The objectives were to determine the relative importance of attributes of services from families' perspectives and whether parents had fixed values about appropriate services or whether they were willing to trade off attributes of programs. We also hoped to examine in an exploratory way whether there were obvious differences in values placed on certain attributes between individuals with different characteristics, for example, parents of children with early or late hearing loss identification, hearing aids or cochlear implants and different degrees of severity of hearing loss. In the context of a newly implemented population based hearing screening in Ontario, Canada, our overall interest was in contributing to an understanding of what constitutes an effective system of care following universal hearing screening from the perspective of the consumer.

Materials and Methods

This study consisted of a cross-sectional questionnaire-based conjoint analysis with families of pre-school children diagnosed with a hearing loss. Conjoint analysis involves a well-defined methodology which has been applied and described by several investigators in health services research (Ryan et Farrar, 2000; Ratcliffe, Buxton, McGarry, et al., 2004; Bishop et al., 2004). The five stages of a conjoint analysis and their application in this study are described below.

Establishing the Attributes

Stage one of the study involved identifying the principal attributes or characteristics of the services for young children with hearing loss and their families. Individual semi-

structured interviews with 17 families (21 caregivers) of preschool age children with hearing loss provided the main source of data for the conjoint analysis attributes. Parents of children who were either diagnosed through systematic screening or traditional referral routes were asked to convey their needs and views of an ideal service delivery model. They were asked to reflect on their perceptions of the strengths and gaps in the current system of care. The interview information was supplemented by a comprehensive literature review. Five key attributes were selected as important to families: location of therapy services (access), amount (frequency) of therapy services, coordination of services, availability of parent support programs, and availability of information. Although many other desirable characteristics were identified, narrowing down the key attributes to five or six is required in order to design a manageable conjoint analysis questionnaire. In this study, we prioritized the attributes from the key themes that emerged from these interviews. A detailed description of the qualitative interviews which informed this study is reported in previous papers (Fitzpatrick, Graham, Durieux-Smith, Angus, & Coyle, in press; Fitzpatrick, Angus, Durieux-Smith, et al., Reference Note 1).

Level of Attributes

The second stage in a conjoint analysis approach involves defining levels for each of the attributes. Typically, two or three levels per attribute provide an appropriate number of choices for the eventual questionnaire. One of the greatest challenges in defining levels is identifying the appropriate spectrum of possibilities for each attribute. Of particular difficulty in this study was defining qualitative levels (e.g. good information access, poor

information access). To conduct a meaningful conjoint analysis, it is necessary that the levels of the attributes represent realistic choices. Secondly, a key premise underlying CA is that individuals will trade off levels of attributes. Therefore, the choices for each attribute must be such that individuals are willing to trade between them. Consequently, in our study, availability and quality of therapy/audiology services were not included as attributes of service provision because previous studies as well as the research which informed this study suggested that parents would not be willing to trade these components of a care model. Similarly, it became apparent during the interviews that all parents highly favored the availability of universal hearing screening as part of the health care system and therefore this service was not included as an attribute (Fitzpatrick et al., in press). The levels selected for each of the five attributes of service models were considered to be realistic and representative of the spectrum of services available as described by parents during the qualitative interviews. The five attributes with their two to three levels and definitions are summarized in Table 1.

The Questionnaire Scenarios

Since all possible attributes and levels cannot be presented in a manageable self-completion questionnaire, these combinations are typically reduced to a reasonable number of choices (hypothetical scenarios) in conjoint analysis studies. The attributes and levels for this study yielded a possibility of 72 scenarios. Using the computer software package Speed version 2.1, a fractional factorial design was created to reduce the original possible combinations to 16 scenarios. The software also applies an orthogonal random effects

experimental design to control for multicollinearity between the independent variables. A random number generator was used to divide the 16 scenarios into 8 pairwise choices. Previous CA research has suggested that individuals can comfortably complete surveys containing up to 16 pairwise choices (Ryan, 1999). Within each pair-wise choice, scenarios were described as Clinic A and Clinic B with their individual characteristics. Table 2 provides an example of one discrete-choice question included in the final questionnaire. Participants were instructed to choose the clinic they would prefer to attend with their child. An additional ninth scenario (a pairwise comparison in which one clinic was clearly the superior service model) was included in the final questionnaire as an internal consistency check to verify that respondents understood the task.

Establishing Preferences and Selection of the Sample

The final questionnaire included a section on instructions, the 9 questions (pairwise scenarios) and detailed definitions of each attribute and level. Prior to distribution to participants, the questions were piloted with a group of 7 clinicians in hearing health care to verify the clarity of the questions and ease of completion. Minor changes were made to the introduction and to the layout of the definitions. Participants were asked to choose clinic A or clinic B based on the profile of the clinic's services.

Participants. The study sample was drawn primarily from 61 parents from three cities in Ontario, Canada who were enrolled in a longitudinal multi-center study investigating the impact of hearing screening on the development of children. To increase the sample size,

additional families who were receiving therapy through the participating clinics, were invited to participate. Consistent with the original study, all children were enrolled in intervention programs in English with a focus on oral communication development.

A questionnaire was sent by electronic or paper mail to the 57 families who agreed to participate in this phase of the study; these parents received one telephone or email reminder to return the questionnaire. Additional questionnaires were distributed to parents receiving services in two clinics from which the original sample was drawn. The study received ethics approval from the four participating institutions and written informed consent was obtained from each participant.

Baseline information including age of diagnosis/intervention, education levels, and severity of hearing loss had previously been collected through a demographics questionnaire with the families enrolled in the original study; these data were also collected from the additional clinic participants.

Data Analysis

Consistent with conjoint analysis techniques described in previous reports, (Bishop et al., 2004; Ryan et al., 2000) data were analyzed using a random effects probit model to account for the multiple responses from each individual. All data were analyzed using the statistical software STATA (*Version 7).* The overall utility (benefit) function to be estimated was represented by:

$$V = {}_1location + {}_2frequency + {}_3services + {}_4parent + {}_5information + e + u$$

where V is the utility or benefit in choosing one clinic A compared to another clinic B and 1 to 5 are the independent variables to be estimated as detailed in Table 1. Due to their categorical nature, the variables, *location* and *services*, were transformed into two dummy variables each to allow regression modeling. The unobservable error terms are represented by e and u, where e is the error term due to difference amongst observations (for a given respondent and u is the error term due to differences amongst respondents (Ratcliffe et al., 2002). The coefficients with their statistical significance show whether the attributes impact respondent choices and the relative importance of each attribute. The ratio of the coefficients show whether respondents are willing to trade an improvement in one attribute for a reduction in another attribute. For example, the coefficient associated with access to parent support can be divided by the coefficient attached to access to information to estimate the extent to which parents are willing to accept a reduction in information access to have an improvement in parent support.

We also explored differences between respondents who differed by route to referral (screened, not screened), type of device worn by the child (hearing aid or cochlear implant) and by geographical region (Ottawa or Toronto).

Results

A total of 48 conjoint analysis questionnaires were returned and constituted the data for this study. All nine scenarios were completed for each questionnaire and all questionnaires met the internal consistency check (i.e., all respondents selected the expected clinic for this question). Therefore, no questionnaires were rejected from the data analysis.

Description of Participants

The 48 respondents were parents or primary caregivers of a total of 51 children with hearing loss who were born between 1998 and 2005. The study was conducted within four years after the introduction of universal newborn hearing screening in the province of Ontario, Canada. Approximately half of the families had come to the intervention process through a systematic screening program (targeted or universal screening) and half had learned of the child's hearing loss through traditional referral routes. All of the children were enrolled in auditory-oral or auditory-verbal intervention services. More than half of the children had a profound degree of hearing loss (52.1%) and were using cochlear implants at the time of the survey. Family education data were compiled according to the highest education level for one parent. All but 2 parents had completed some degree of post-secondary education with the majority having completed a college or university-level education (77.1%). Further details of the characteristics of the respondents are summarized in Table 3. For the three families with more than one child with hearing loss, demographic data for the oldest child were used in the summary statistics.

Survey Results

As shown in Table 4, the coefficients for all attributes in the regression model are significant at the $P < .01$ level, indicating that all five attributes were considered to be important characteristics of a service model by these respondents. Specifically, the results for each attribute show that these parents preferred: 1) clinic-based services to home-based services, 2) coordinated services through one agency compared to either services that were

not coordinated or a model without psycho-social services, 3) access to parent support independently rather than through health services and 4) access to information through their clinical programs. The weight of each coefficient indicates the relative importance of each attribute in determining overall utility or benefit. The relative size of the coefficient (-8.07) shows that service coordination dominated all other attributes for this group of parents in choosing a preferred clinic. Overall, the findings suggest that parents most highly favor a coordinated service model with intervention provided in a clinical setting with good access to information and with parent support organized separately from the health system. While access to information was valued in the model, it received less weight relative to other factors in the choice of a preferred service model.

The ratio of the coefficients indicates the relative strength of preference of respondents between attributes. For example, these respondents would be 2.7 times more willing to accept a reduction in their access to information to ensure that they have parent support available as part of the service model (i.e., the ratio of the coefficient for parent support of 4.63 to the coefficient for information of 1.70 is 2.72). Similarly, parents would be 4.8 times more willing to give up access to information to have well-coordinated services with access to psycho-social support (ratio of coordinated services to information = 4.75).

We also explored, using separate regression models, the differences in responses for participants according to three additional variables: screening status (screened, not screened), hearing device (hearing aid or cochlear implant) and region (Ottawa or Toronto). When entered as covariates in the regression model, none of these variables was significant and did not add any additional information to the model. Therefore, in this study, screening

status, type of hearing device or region did not explain any significant variance in preferences between respondents. We also examined whether preferences varied across these subgroups by adding screening status and region as interaction terms in the model. The results of segmentation of respondents by screening status and region showed no differences in the preferences attached to attributes. The small sample size precluded statistical analysis according to type of hearing device.

Discussion

The technique of conjoint analysis has gained broad support as an approach to inform decision-making across a range of health care issues. This study represents the first time, to our knowledge, that the method was applied to elicit preferences for health services from parents of children with permanent hearing loss. Previous efforts at defining pediatric hearing care models have used qualitative research and surveys to highlight aspects of care that are desirable to families but have not measured or prioritized specific elements of service provision in a quantitative manner.

Our results paired with the qualitative data which informed this survey support parents' preferences for a multi-component service model that offers coordinated access to services in a clinical-type setting with parent support and good access to information. Above all other components, coordinated services in audiology, therapy and other support services dominate all other preferences. The frequency of therapy, once a week versus to two to three times per week although statistically significant, was accorded less importance than several other attributes by these families. This may reflect the fact that when parents

perceive that they are receiving quality guidance as suggested by the interviews, they do not feel that additional therapy visits enhance their child's potential to develop communication. During the interviews, some parents expressed that while having a tutor come to the home was beneficial to supplement the work of the therapist and parent, they felt that additional visits to a therapist were not necessary.

One surprising finding was that parents favored a clinic-type setting over home-based services or even services that alternated between home and clinic. This may reflect a certain bias in the sample in that the majority of families were involved in clinical intervention programs. It may also represent the unwillingness of families to trade the clinical services they have for home-based service, that is if there is a perception that the same caliber of services are not offered in a home setting. In other words, the choice may reflect an unwillingness to trade the quality of care for the convenience of a home-based service. In summary, these results suggest a preference for a focus on a well-coordinated clinical service model with parent support and good information access as part of the care model.

Previous reports suggest that defining the levels of the attributes can be the most complicated part of constructing a conjoint analysis questionnaire. In this study, the attributes and levels were informed by the qualitative interviews with parents who were in the process of experiencing the system of care. These parent interviews provided the contextual information for the study, helped define the attributes and ensure that the levels selected were plausible and reasonable for parents who use the service. However, with conjoint analysis, the researcher is limited to a relatively small number of attributes,

therefore not all characteristics identified in the parent interviews could be included. For this study, the most common components of the model were selected for inclusion based on thematic analysis (Fitzpatrick et al., Reference Note 1). A further limitation of this study, described in other conjoint analysis studies,(Ryan, McIntosh, & Shackley, 1998; Viney, Lancsar, & Louviere, 2002) was that patients could only rank components of models that they have experienced or that they could foresee. This may, in part explain parents' relatively strong preference for services offered in a clinical setting. Patients receiving treatment in other health systems could have different preferences for care. In our study, no significant differences in strength of preferences could be accounted for by screening status of the child, type of device used by the child or the city in which the family lived. However, due to the relatively small sample size, subgroup analyses were not possible based on factors which have been shown to impact preference outcomes such as education, income and geographical location (rural or urban). These subgroups could place different values on certain services, for example, home-based interventions or the availability of parent support through their clinical programs.

This study quantifies the values that patients place on various components of a service delivery model for children with hearing loss and their families. Parents are concerned with more than just the rehabilitation service and are interested in a "package" of care.

Furthermore, parents are willing to trade changes in the probability of one service being available to have another service more easily accessible. This study provides information on parent preferences for service model characteristics beyond the mere

inclusion of quality therapy and audiology services. Information on the utility (benefits) patients associate with aspects of the service delivery is of particular relevance for program planning and development. This information can be applied by clinical programs when designing comprehensive intervention services for children with hearing loss and their families. The findings of this study also have implications for decision makers as the results can influence how resources will be allocated in designing and improving clinical services.

There is considerable consensus in health care that, while the effectiveness of an intervention is important to users of health care, other aspects of the care process are also important, that is, the make-up of the required treatment and the process of delivery. Conjoint analysis shows promise as a technique for addressing these difficult questions. Further studies with diverse samples of families coping with childhood hearing loss, for example, rural populations and families of lower socio-economic status, can contribute additional information to inform the design of effective intervention programs in pediatric hearing care. This research would also be strengthened through future studies examining the preferences of clinicians working in infant hearing services since many of the decisions about how programming is delivered are made by individual clinics based on their views and beliefs of what parents need.

In summary, evidence has been provided for the importance of various attributes of a service model for children with hearing loss. Conjoint analysis is an established technique that invites user input into health care decision making. To our knowledge, no previous study has applied established conjoint analysis techniques to investigate parent preferences for services related to childhood hearing loss. Meeting parents' preferences for services

may have an effect on their ability and willingness to provide the recommended intervention program for their children. There is some research suggesting that parental involvement in the intervention program may be an important variable in determining eventual outcomes in children with permanent hearing loss (Moeller, 2000; Calderon, 2000). It is plausible that parental involvement and therefore the effectiveness of universal newborn hearing screening initiatives can be improved when intervention services meet parents' preferences for care.

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Table 1. Attributes and levels in the conjoint analysis study

Attribute	Level
Location of therapy	• Clinic • Home or community • Alternate between clinic and home or community
Frequency of therapy	• One time/week • 2 to 3 times/week
Coordinated services (e.g., therapy, audiology, psycho-social)	• Coordinated through same agency as therapy • Services available – not coordinated through one agency • No psycho-social services available
Access to parent support	• Organized as part of agency/health services • Independently accessed by parent (e.g., parent groups)
Access to information in early stages	• Provided through clinic audiologists/ therapists (e.g. verbal, literature, videos) • Not easily available through clinic

CA Design: $2^3 \times 3^2$ *Note: The final questionnaire included detailed definitions of each attribute and level.*

Table 2. Example of discrete-choice question

	Clinic A	**Clinic B**
Location of therapy	Home or community	Home or community
Frequency of therapy	1 time/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Coordinated through same agency as therapy
Access to parent support	Independently accessed by parent (e.g. parent groups)	Organized as part of agency/health services
Access to information in early stages	Provided through clinic audiologists/therapists (e.g., verbal, literature, videos)	Not easily available through clinic

Which Clinic would you prefer? A ☐ B ☐

Table 3. Characteristics of respondents (n=48)

	Number	Percent (or range)
Screening status		
Screened	25	52.1%
Not screened	23	47.9%
Age diagnosis		
before 9 months	24	50.0%
after 9 months	24	50.0%
Region		
Ottawa	27	56.3%
Toronto	21	43.7%
Parental education (mean yrs)[1]	17.9	range 13 to 23 years
Degree of hearing loss		
Mild	1	2.1%
Moderate	14	29.2%
Severe	11	22.9%
Profound	22	45.8%
Device type		
Hearing aids	23	47.9%
Cochlear implants	25	52.1%

[1] *Defined as the highest number of years of education for one parent*

Table 4. Results of random effects probit model (n=48)

Attribute	Coefficient	(95% CI)	P-Value
Locdum1	3.50	(1.61, 5.39)	<0.001
Locdum2	3.36	(1.83, 4.89)	<0.001
Frequency	-2.38	(-4.02, -0.75)	0.004
Serdum1	-8.07	(-10.72,-5.42)	<0.001
Serdum2	-3.74	(-5.42, -2.06)	<0.001
Parent support	-4.63	(-7.03, -2.24	<0.001
Information	1.70	(0.98, 2.43)	<0.001

CI: confidence interval

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CHAPTER 6

INTEGRATED DISCUSSION AND CONCLUSIONS

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This chapter summarizes and integrates the findings of the research in terms of implications for theory, population health, practice and policy. The results are interpreted in relation to the conceptual framework which guided this project. This concluding chapter finishes with suggestions for further research.

Summary of the Research

Objectives of the Research

Against a backdrop of new population-based newborn hearing screening initiatives in Canada, this study was undertaken to examine the benefits of early identification of childhood hearing loss and the needs of families of affected children. Adopting mixed methods, the objectives of the study were addressed through three interrelated inquiries: 1) an examination of traditional communication outcomes (chapter 2), 2) an exploration of families' experiences with the screening, assessment and intervention process as well as their needs (chapters 3 and 4), and 3) a survey of families' preferences for the various characteristics of service models (chapter 5).

Context

Although there is a considerable body of literature substantiating the effectiveness of population screening for the early detection of childhood hearing loss, the evidence for a

clear association between early identification and future communication skills remains inconclusive.[1] While studies in the 1990s focused on communication outcomes to measure the benefits of early identification, more recently, investigators have questioned whether speech and language measures should be the ultimate outcome for evaluating the effectiveness of newborn hearing screening.[2,3] Previous research has served to point out that many child, family and contextual factors may mitigate the impact of later identification and influence outcomes.[4] Consequently, there is a growing recognition that screening is the first step in a comprehensive care process to minimize the impact of childhood hearing loss on individuals and society.

Theoretical Basis of the Research

The framework adopted for this study (Figure 1, Chapter 1) attempted to capture this notion of multiple outcomes and multiple influencing factors. In this conceptual framework, outcomes were conceptualized as consisting of: communication outcomes (child), process outcomes and family outcomes, all of which were examined during the course of this study.

The influencing and interrelated factors for infant hearing development were categorized as follows:

Environmental Factors: hearing technology, geographic location, type of intervention, access to intervention and other services, and service delivery models

Child Factors: age of diagnosis, severity of hearing loss, developmental level, and temperament.

Family Factors: family functioning, socio-economic status, family resources, and previous experience.

The framework served to categorize the extant literature and provided a reference for the research objectives and questions addressed in this study. Contextual outcomes (environmental factors) have received little attention by way of scientific examination in comparison to outputs of newborn screening programs and communication outcomes. This doctoral research represents, in essence, a paradigm shift in newborn screening research in that it examined broader outcomes and explored determinants of outcome beyond proximal factors. Furthermore, the research privileged the perspective of those intricately involved in the care process – the parents.

Method

The mixed methodology approach adopted for this study was largely influenced by the framework in that a broader examination of outcomes and factors, some of which were not amenable to traditional measures, required both quantitative and qualitative methodologies. Following an examination of traditional communication outcomes using quantitative methods (inquiry 1), a qualitative methodology was applied in inquiry 2 to understand parents' viewpoints on outcomes. This inquiry, helped interpret and expand the findings from the quantitative analysis. This same qualitative research also served to explore families' needs; these findings were then used sequentially to inform the conjoint analysis to

quantify parents' preferences for the attributes of care models. The conjoint analysis corroborated and reinforced the findings of the qualitative analysis.

Summary and Interpretation of Findings

Inquiry 1

The first inquiry of the thesis involved an analysis of existing data that were collected as part of a comprehensive research project investigating multiple developmental outcomes in screened and unscreened children. This analysis was undertaken specifically to examine the association between age of diagnosis and communication outcomes. Our interest lay in examining whether typical communication outcome measures could inform the discourse on the effectiveness of population hearing screening. Group comparisons showed that screened children in this sample did not perform at higher levels than unscreened children and children diagnosed prior to 12 months of age did not demonstrate stronger speech and language skills than those diagnosed after 12 months.

A strength of this outcome study, which served as the basis for the first analysis (inquiry 1), was the child and family characteristics of the study group. All children had early onset deafness and absence of identifiable additional disabilities. All were in oral language rehabilitation services and most were from families of similar socio-economic status. The multi-center dataset consisted of a comprehensive battery of standardized speech and language measurement tools. The data were rigorously collected by trained examiners who

were separate from the child's clinical program and blinded to the extent possible to the severity of hearing loss of the children.

The failure to show differences in communication development in this inquiry may have been partly due to the fact that there was only an average of 13 months difference between screened and unscreened children. In addition, the study timelines (three year time period for the data in inquiry 1 may have been too short to demonstrate differences in children according to age of diagnosis. First, the effect may not be discernible in speech and language measures for young children and may become more apparent with more sensitive and sophisticated language and literacy measurement tools. Secondly, if population screening affects multiple changes at different levels of the system, these may not have permeated the system, given that the data were collected within 2 to 3 years of a newly implemented program.

Although a province-wide recruitment strategy would have strengthened this study, such an approach was not feasible given the absence of a provincial database for children with hearing loss. All eligible patients followed in the clinical programs were invited to participate in the quantitative study, thereby avoiding the problem of convenience samples discussed by Thompson and colleagues.[1] However, a major limitation for the quantitative inquiry is the sample size despite three years of recruitment in this multi-center initiative. The multitude of variables that interact with age of diagnosis/intervention coupled with the difficulty in recruiting large population-based samples continue to present challenges in

elucidating the question of the impact of population hearing screening on communication development measures.

Notwithstanding these limitations, the prospective nature of the quantitative research sets it apart from several earlier studies conducted in the 1990s that addressed the effectiveness of newborn hearing screening in improving communication development. While quantitative studies investigating developmental outcomes in the late 1990s categorically found UNHS to be conducive to better outcomes[5,6] and for many years formed the science-base for UNHS, these studies were rated as poor to fair quality in a subsequent systematic review. The rating was based on the use of convenience samples and weak reporting of subject selection and attrition rates.[7] The inability in this thesis research to show differences in communication development between early and later identified children is consistent with a recent population-based study in Australia, however, the results contrast with those of a second controlled study in the UK, where superior outcomes in language but not speech were documented in early identified children. The study undertaken for this thesis coupled with the Australian study suggests that the clinical context, that is, type and quality of intervention, as well as parental involvement may be important factors that interact with age of identification and numerous other contributors in determining eventual communication skills.

These findings coupled with previous literature[1,8] reinforced the importance of examining outcomes beyond typical communication measures and motivated the next stage of the thesis.

Inquiry 2

In addition to helping interpret the findings on communication outcomes, the qualitative interviews revealed that parents perceived several other benefits for the child and family related to early detection of a child's hearing disorder. These positive outcomes have not been captured in traditional outcome measurements of the effectiveness of early identification on children's development. In addition to improved speech and language development, families perceived child benefits related to access to hearing and self-esteem. Benefits for the family centred on reduced frustration in accessing diagnostic services as well as feelings of less urgency and less guilt related to late diagnosis of hearing loss. Families also envisioned that infant screening programs would help overcome some of the barriers in transitioning to therapy and other resources following diagnosis.

A second objective of the qualitative interviews was to understand families' needs in caring for a child with special requirements. While quality therapy and audiology services were of primary importance to all families, other characteristics of service provision that emerged from the data included a service model that is well coordinated and facilitates access to ongoing medical, technical, prognostic and resource information. In addition, access to

support from other parents of children with hearing loss was identified as having positive benefits for the family.

A potential weakness of the qualitative research (inquiry 2) lies in the fact that there was limited representation from families living in rural areas, therefore experiences and needs could be different in smaller and more isolated regions of the province and country.

Inquiry 3

The purpose of the conjoint analysis survey was to ascertain the strength of families' preferences for the various attributes of services. The findings complement many of the interpretations from the qualitative analysis. In particular, the results of the conjoint analysis questionnaire demonstrated that, above all other characteristics of service models, parents valued coordinated services that were available through one agency or at least managed through one agency. Parents also showed preferences for parent support services and good access to information through their clinical programs. Overall, the survey indicated that the preferred model was coordinated, clinic-based services with a range of audiology, speech and psycho-social services, access to parent support and information available through the health service program. Although coordinated services and parent supports have emerged as important issues in other studies, this inquiry represents the first time that they have been quantified by parents through a preference-based forced-choice exercise.

The inclusion of parents of children who were newly and recently identified with hearing loss strengthened this cross-sectional survey in that respondents were selecting hypothetical services at the time that they were experiencing the care process with their child. Although the sample size for the conjoint analysis survey was sufficiently large to assess statistical inference, the sample size did not permit sub-group analyses according to some of the factors that may affect responses such as various degrees of hearing loss, socio-economic status, and rural versus urban regions.

Integration of Findings

The use of mixed methodologies applied sequentially throughout this research complimented and strengthened the findings. The qualitative analysis (inquiry 2) added depth to the quantitative data collected through traditional speech and language measures (inquiry 1) and facilitated interpretation of the findings. The notion of becoming "caught up" which emerged in the family interviews, paired with parents' high satisfaction with the quality of the therapy services, suggested that despite a later start for some of the children, communication skills comparable to earlier identified children could be achieved.

The qualitative research is unique in that it addresses the gains from early identification from the perspective of those affected. Previous research has investigated parents' views on screening[9] including both parents of screened and unscreened children and more recently parents' experiences with screening and diagnosis.[10] However, to our knowledge, no previous research has used qualitative methods to specifically explore outcomes beyond

traditional speech and language measures from the perspective of parents. This study makes a unique contribution in addressing the value of screening using a broader definition of outcome than previous research. Other studies have also examined parents' needs[11,12] for services but none has specifically examined parents' views on the components of service models.

The information on parents' needs and the identification of several desirable characteristics of services collected in inquiry 2 formed the basis of the conjoint analysis survey (inquiry 3). While the needs data from the qualitative interviews informed the content of the conjoint analysis survey, the results of the survey complimented and confirmed the themes that emerged in the qualitative interviews. Given that this study adopted a unique methodological approach in applying conjoint analysis to explore parents' preferences for attributes of service models, the qualitative interviews were an important prerequisite to defining meaningful attributes of care.

Revised Conceptual Framework

The new knowledge acquired in this study furthers the understanding of infant hearing research in regard to the current focus on early identification. Figure 2 presents a conceptual view of how the new findings align with and extend the initial framework which guided the study. Overall, this study has demonstrated that families of young children with hearing loss, regardless of whether they entered the process through newborn screening or not, value early identification initiatives as a core component of a system of infant hearing services.

Although the effectiveness of screening in improving specific communication outcomes could not be quantified through objective measurement tools, this research provides compelling evidence that parents either experienced or envisioned several positive child and family benefits from early identification. These benefits from the perspectives of families combined with the evidence in the literature for improved access to hearing, which emerged as an important benefit from families' perspectives, provide support for universal hearing screening.

The conceptual framework which guided the study has been reconfigured to integrate and advance this new knowledge. The outcomes are arranged and defined as three interrelated outcomes: communication outcomes, process outcomes, and impact on family outcomes. In the modified framework, each outcome category has been further defined to incorporate the findings of the study. The outcomes have been rearranged to reflect that they are not discrete and separate phenomena but rather have the potential to interact with each other. For example, in this study, families who perceived that their children had access to hearing (communication outcome) described the positive impact of early identification of hearing loss on the family whereas some families who were concerned about their child's poor communication skills attributed it to late diagnosis and referred to the increased guilt and anxiety at the family level. Families who experienced difficulty with the referral process to audiology (process outcome) also associated this experience with increased stress, frustration, and anxiety for the family and with reduced opportunities for the child to learn through hearing. The definition of outcomes provided by this study moves the field beyond

the narrow and more traditional boundaries common to previous investigations of the benefits of population screening.

The objective evidence for superior language outcomes from early intervention remains inconclusive. This is due to the difficulty in controlling for many other variables such as severity of hearing loss, technological advances in hearing devices, family involvement, type of intervention and type of service models. The findings of this thesis permit an elaboration of these factors in the framework. First, the boxes for environmental factors, family factors and child factors have been repositioned (reordered from left to right) as environmental factors appeared to most closely interact with family factors to facilitate or create barriers to meeting families' needs such as technology and intervention services. For example, families who described financial hardship which in turn interfered with timely access to hearing technology described a barrier that is directly influenced by family circumstances. This factor has the potential to impact the child's access to hearing, and eventual outcomes in multiple domains. Second, in particular, the environmental factors box has been rearranged to include and reflect the findings of the study. In this reconfigured framework, service providers, geography and service models are seen as dynamic and interrelated factors that can influence and determine the availability of other components, e.g., hearing technology, access to service. In addition, coordination of care, parent support, and information access which emerged as strong themes in the qualitative interviews and received high preference values in the conjoint analysis have been added as characteristics

Conceptual Framework for Infant Hearing Loss

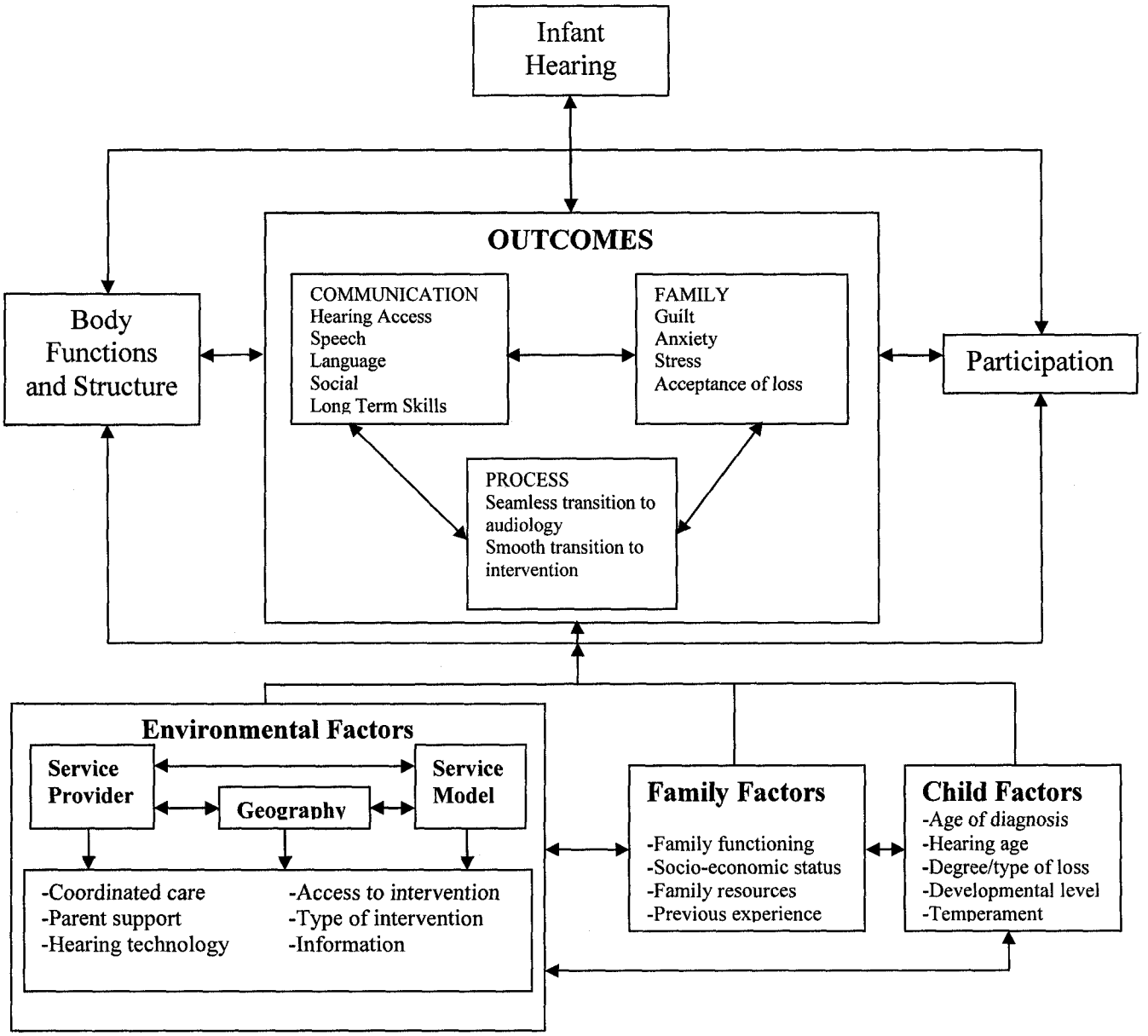

Figure 1. Revised conceptual framework of outcomes and factors

of the environment. Finally, in the category of child factors, *hearing age* (age at which child begins to hear sound) has been added in addition to age of diagnosis to reflect this concept, which so strongly emerged in this research, both as a positive consequence of population screening (i.e., access to hearing) and as a contributor to outcome from the perspective of families.

Relevance to Population Health

Infant hearing screening continues to occupy an important place on national and international health agendas as a population health intervention aimed at improving healthy child development for children affected with permanent hearing disorders. An important rationale for any population screening program is to identify a problem such that it can be managed earlier, thereby improving health results and reducing the disparities in outcomes across members of a population. Accordingly population infant hearing screening has received widespread support on the basis of 1) the burden of the disorder to society and 2) the assumption that early identification of hearing loss can prevent or reduce delays in child development.

This doctoral thesis endeavored to move beyond the traditional biomedical boundaries of audiology research and apply a population health perspective in examining the benefits of universal hearing screening.[13] A population health perspective informed the conceptualization of this research project in a number of ways. Consistent with population health principles, this project focused on increasing the science-base for neonatal hearing

screening by examining outcomes in a domain where clear evidence for improved communication outcomes has been difficult to establish. However, the project moved beyond typical speech and language measurements to include a broader perspective of family and contextual outcomes.

Population health encourages not only an evidence-based approach but is largely concerned with the determinants of health outcomes. This study included an exploration of factors beyond the proximal determinants of health to better understand what influences outcomes in children with hearing loss. In line with a population health view, this study adopted a mixed methodology approach to uncover multiple intervening factors. In particular, this study assembled evidence which points to the importance of context in modifying the association between functioning and specific individual determinants such as age of diagnosis of a hearing disorder.

Hearing screening is a population intervention that may reduce, minimize or eliminate the negative consequences of childhood hearing impairment. An important focus in this study was on understanding parents' needs in order to maximize the potential benefits of population screening. Most importantly, parents' views on what makes a preventive measure like hearing screening beneficial and what makes it work were at the core of this study. As such, this study is aligned with a population health approach which shifts thinking from clinical treatment to determining how population health interventions can have the greatest impact on outcomes.[14] Increasingly, there is a realization that infant

hearing screening cannot be isolated from the subsequent management of hearing loss and family supports. The findings of this research contribute to the growing interest in designing appropriate post-screening intervention services.

Population health is concerned not only with seeking to better understand the many influences on health but also using the knowledge to change health practices and policies to improve health. The findings of this research suggest that several environmental or contextual factors can change outcomes for children and their families. In particular, health services, one of the determinants of population health models,[14] emerged from this research as a core determinant requiring attention in the future development of population screening programs. The findings of this thesis can be used to design intervention services to maximize the new opportunities presented by universal screening.

Assembling the findings in the revised framework brings to light the fact that population infant hearing screening is not a single intervention but rather a catalyst for multiple interventions which potentially affect different levels of the system, including the individual (child), family, service provision and societal levels.[15] This further explains why it may be extremely difficult to isolate the effects of screening (age of diagnosis) on longer-term communication outcomes as the implementation of universal screening programs have typically translated into multiple interventions at multiple levels of infant hearing care.[3,16,17] Furthermore, as reflected in this framework and described by Edwards and colleagues,[15] many of these factors (e.g. hearing technology, type of intervention) can be viewed as

layered or "nested" within other determinants (e.g., service model of care). Interventions such as universal screening can effectively create synergies which can optimize the impact of screening and ultimately affect the ability of children and families to more fully participate in society.[15]

Finally, the scholarly inquiry undertaken in this research resulted in a review and modification of the original guiding framework. The modified framework developed to communicate the findings is grounded in a population health perspective. A hallmark of a population health perspective is the acknowledgement of the complex and overlapping interactions among the various determinants of health. The synthesis of the findings into a new framework which encompasses multiple outcomes and a diversity of determinants of outcomes provides a common language for audiologists, language and other specialists, program administrators and policy makers interested in population infant hearing practice and research.

Overall Strengths and Limitations of the Research

A strong point of this research is that families from three to four different regions were involved in the three inquiries thus representing children from diverse clinical service models. Thus, the findings cannot be attributed to any particular service provider or program. Drawing from this sample for the subsequent inquiries meant that considerable demographic and clinical information was already available. This permitted a purposeful sampling approach for the qualitative interviews that took account of diverse family

characteristics which potentially influence experiences and views on benefits and needs. Common inclusion and exclusion criteria were applied to the quantitative and qualitative inquiries. The findings should be transferable to other centers particularly in urban areas with practice models focused on oral communication development. The research findings are based on a strong multi-method study design as well as rigorous data collection and data analysis methods. The project also benefited from regular debriefing sessions with an interdisciplinary research team. The combined use of quantitative and qualitative research helped to confirm or better understand the results and thus served to triangulate the data, contributing to the overall validity of the research findings.

A limitation of the research lies in the fact that, for practical constraints of time and funding, the sample was drawn primarily from two large cities in one Canadian province. The absence of a central database for infant hearing also restricted recruitment of participants primarily to two large clinical diagnostic centers. Given the potential for subject selection bias using these recruitment methods, the sample was potentially lacking in terms of rural representation and socio-economic diversity and was possibly biased towards children with more severe hearing loss. It is possible that families who described themselves as resourceful and who were able to access care in certain clinical contexts were able to overcome the consequences of late confirmation of hearing loss. The relationship between the age of detection of hearing loss and later communication skills may depend on the specific child, family and environmental context in which differences in age of detection are

present. Likewise, family-perceived views on outcomes and needs may have been influenced by these characteristics.

A second limitation is related to the timing of the study in that it was undertaken within two to three years following the implementation of the provincial infant hearing program (including universal screening). This timeline was partly motivated by the practical need to enroll a comparison group of unscreened children in the study who were diagnosed in approximately the same time period. However, starting the research in the early stage of the new program likely affected 1) the characteristics of the sample, for example the project included a large number of children from the "at-risk" category (primarily NICU graduates) and 2) certain characteristics of services, for example access to audiology and other services.

Implications of the Research

Although Canada has made considerable progress in recent years toward implementing population newborn hearing screening as a standard of care, systematic screening services are not available for approximately half of Canadian children. In the current era of evidence-based care, in light of the uncertainty in the literature, providers and policy makers continue to question the benefits of universal screening. The communication, process and family outcomes defined in this study can inform clinical priorities and policy decisions in Canada and other countries. This research showed that although there remains uncertainty about the specific contribution of early identification to later communication skills, parents

embrace the opportunity for better hearing and the potential for improved speech and language.

From a clinical audiology perspective, this thesis was largely motivated by questions and concerns about the implications at the clinical practice level of providing services for young infants. The exploration of parents' needs supplemented by the conjoint analysis survey provide important information on the characteristics of a service model that are most important to parents in promoting their children's development, these are attributes which clinics can strive to offer families when they implement programs. On a practice level, attention should be given to providing parents with the best available information about their child's prognosis at diagnosis and throughout the continuum of care, ensuring that parents have an understanding of the next steps in the care path. In addition, clinicians should make efforts to link parents of newly identified children with other parents of children with hearing loss. Since communication between professionals emerged as an important characteristic of coordinated care, service providers should give priority to ongoing communication with other services involved with the family. Given the lack of clarity of the role of social worker or family support worker for some families in this study, clinicians should endeavor to clarify this role for families at the outset so that all providers are viewed by the family to be working as a team in the best interest of the child and family.

Access to information emerged as an important attribute and expectation of care from the viewpoint of families. Information about technology and resources was frequently cited as

the number one need at the time of diagnosis. In busy clinics, parents perceived that this service was sometimes overlooked. Clinics should review their information dissemination practices keeping in mind that in addition to print and video materials, families use the internet as an important resource.

From a health services perspective, the findings have implications for regions that are implementing new population screening programs or improving existing services. The needs identified by parents in the qualitative interviews and the preferences quantified through the conjoint analysis can inform the design of future programs. Consideration should be given to establishing programs which offer seamless coordinated services where parents have easy access to a variety of information sources, quality professional care, as well as other experienced parents who can support them throughout the care process.

Future Research

This research lays the groundwork for a future research program in population screening and infant hearing. First, efforts should be undertaken to continue collecting meaningful results in children from screening and non-screening programs to determine whether the findings observed in this study are typical or whether they are restricted to particular clinical settings. Second, longer-term outcome should be collected on children in this study and similar studies in an effort to determine whether the effects of early identification are revealed at a later age through more sophisticated and discerning language and literacy measures. A systematic evaluation of the impact of age of diagnosis on children with cochlear implants

needs to be conducted. Given that children are typically not implanted until 12 months of age (according to current age of implantation criteria), early identification may not have the presumed effect on outcomes in this population of children. That is, the inclusion of children with cochlear implants in future studies may actually obscure the effects of early identification on communication development.

Although parents welcome population screening, the current goal and sense of urgency to fit affected children with hearing technology by 6 months of age should be reviewed for two reasons: 1) the inability of the present study to quantify measurable differences in communication skills and 2) the qualitative findings that some parents may need time to adjust to the diagnosis before proceeding with amplification. As children and families enter the health care system earlier through population screening, they consume health services for a longer period of time which can be expected to place a greater economic burden on hearing health services. Current management models should be examined in light of these new demands and the new opportunities for hearing provided by early identification of childhood hearing loss. Furthermore, given the number of parents who identified the need for better prognostic information at diagnosis and beyond, research should be directed to developing developmental markers to measure progress in children educated in oral language programs.

This project identified coordinated services as an important characteristic of parent-perceived family-centred care. Accordingly, there is a continued need to identify the

components of coordinated services from the perspective of families as family-centred service is likely to be an important determinant of the effectiveness of early intervention.

The importance of service models has received little scientific investigation and may have been overlooked as a strong determinant of outcome in children with hearing loss and their families. Given the strong influence of service providers on the care model, the technique of conjoint analysis can also be applied to elicit the preferences of service providers for attributes of service provision. Furthermore, the implementation of universal screening may impact multiple levels of the system including the education/training of clinicians and the provision of rehabilitation services. Given that newborn screening has been practiced for a few years in some regions, future research with a focus on lessons learned at a grassroots level can provide important insights to assist with the implementation of new initiatives.

In view of the fact that population screening is a new initiative, program evaluation of its impact on existing services and on child and family outcomes is warranted. The framework generated from this research defines pathways and inputs that could be targeted in intervention and evaluation design. Specifically, the framework highlights several outputs that can be used in program evaluation. In addition, the influencing factors detailed from the perspective of families, provides a list of inputs that should be given attention in designing evaluations.

Summary

Currently, an estimated 50% of Canadian children with hearing impairment are being identified through population screening programs. The articles in this thesis (Chapters 2-5) summarize a comprehensive study of children with hearing loss and their families. It is comprehensive in the breadth of outcomes examined and in the investigation of determinants of outcome. The findings are relevant to the implementation of future population screening initiatives and to clinical practice and policy making for infant hearing services. This thesis represents the first research in the Canadian population health context to undertake a comprehensive investigation of the impact of newborn hearing screening on child and family outcomes. Parents' views support the value of newborn hearing screening as a health service. However, despite some evidence that hearing screening improves outcomes, the full benefits will not be realized unless appropriate services are in place to support families. By understanding parents' perceptions of outcomes and needs, practitioners and decision-makers may be able to develop services to maximize the benefits of population screening. In summary, as the only Canadian multi-method study to address infant hearing screening benefits and intervention needs, this research is an important step in enhancing the knowledge-base and laying a foundation for future research in this population health intervention for Canada's youngest citizens.

Statement of Contributions

E. Fitzpatrick (EF), the doctoral candidate assumed responsibility for this research project. She was closely supported by a thesis committee from the University of Ottawa that included Professors Andrée Durieux-Smith, Doug Angus, Ian Graham and Doug Coyle. The interdisciplinary thesis committee provided expertise in infant hearing science, health sociology, health policy, and health economics. All four professors were involved in the conception of the research project. Regular meetings were held to monitor progress, assist with interpretation of findings and provide ongoing guidance as recruitment and methodological questions arose.

With the exception of the first inquiry, EF led the project through the data collection, data analysis and synthesis stages. For inquiry 1, EF took responsibility for the analysis of communication outcome data collected as part of a multi-center investigation examining multiple developmental outcomes in early and late diagnosed children. Prior to starting doctoral studies, EF was involved as a co-investigator in this multi-center study led by principal investigator, Dr. Andrée Durieux-Smith (thesis supervisor). The data analysis undertaken as part of the doctoral work provided the foundation for the remainder of the doctoral thesis. EF took responsibility for Research Ethics Board approvals, recruitment, data collection, data analysis, interpretation and synthesis for inquiries 2 and 3.

EF assumed responsibility for writing the four manuscripts with guidance from thesis committee members. For manuscript 1, input was also obtained from the 3 co-authors. Dr.

Alice Eriks-Brophy, Dr. Janet Olds and Dr. Robin Gaines who were co-investigators in the larger infant hearing study. EF wrote all drafts of the manuscripts. Final manuscripts were reviewed and approved by all co-authors prior to submission.

Other contributors

In consultation with the thesis committee, other individuals were involved in this study as required. For the first inquiry, R. Denondan provided initial statistical planning support and I. Gaboury provided statistical consultation. J. Whittingham from the Audiology Research Laboratory at the Children's Hospital of Eastern Ontario Research Institute extracted data from the study database, provided descriptive statistics for the study sample, and conducted preliminary statistical analyses. For the second inquiry, D. Neuss verified the data coding. I. Gaboury provided statistical support for the conjoint analysis in the third inquiry.

Knowledge Translation

Throughout this project, several knowledge translation activities took place. In addition to the acceptance of one peer-reviewed article, presentations have been made at clinical rounds, scientific conferences and meetings. The findings from inquiries one and two have been shared with clinicians at two of the participating clinics. In addition, the findings have motivated and informed the preparation of a grant application to establish benchmarks in children who use cochlear implants.

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- Fitzpatrick, E. Schramm, D. & Durieux-Smith, A. Intervention after cochlear implantation: Preliminary findings. In M. Tong, V. Chan, & van Hasselt, A. (Eds,). *Proceedings of the 5th Asia Pacific Symposium on Cochlear Implant and Related Sciences*, Bologna, Medimond 2006;159-163.

Presentations:

- Ottawa-Carleton District School Board
- Rehabiliation Patient Services Unit, CHEO
- CHEO Research Institute
- The impact of infant hearing screening on communication development (paper submitted Nov, 2006). *11th International Conference on Cochlear Implants in Children*
- Baby Benchmarks (grant in preparation, Neuss, Fitzpatrick, Olds, et al.)

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Appendix 1

Search Strategy

Search Strategy

1	exp Hearing Disorders/
2	infant/ or infant, newborn/
3	1 and 2
4	limit 3 to humans
5	limit 4 to english language
6	4 not 5
7	limit 6 to abstract
8	5 or 7
9	exp Mass Screening/
10	screen$.tw.
11	exp Hearing Tests/
12	9 or10 or 11
13	8 and 12
14	cochlear Implants/
15	exp Hearing Aids/
16	Exp Manual Communications/
17	Exp "Rehabilitation of Hearing Impaired"/
18	esp haring disorders/dt, rh, su, th
19	14 or 15 or 16 or 17 or 18
20	8 and 12
21	13 or 20
22	exp Hearing Disorders/
23	limit 22 to humans
24	limit 23 to english language
25	limit 24 to ("preschool child (2 to 5 years)" or "child (6 to 12 years)" or adolescent (13 to18 years)")
26	19 and 25
27	exp Evaluations Studies/
28	Follow-Up Studies/
29	meta analysis/
30	exp Clinical Trials/
31	27 or 28 or 29 or 30
32	26 and 31
33	limit 32 to (controlled clinical trial or guideline or meta analysis or multicenter study or practice guideline or randomized controlled trial or review, multicase)
34	32 or 33
35	21 or 34

Reference: http://www.ncbi.nlm.nih.gov/books/bv.fcgi?rid=hstat3.section.1866 (Retrieved April 15, 2006)

Appendix 2

Research Ethics Board Certificate – University of Ottawa

Research Ethics Board Certificate - Children's Hospital of Eastern Ontario

Research Ethics Board Certificate - Hospital for Sick .Children

Research Ethics Board Certificate - Learning to Listen Foundation

Research Ethics Board Certificate – University of Western Ontario

Université d'Ottawa University of Ottawa
Service de subventions de recherche et déontologie Research Grants and Ethics Services

HEALTH SCIENCES AND SCIENCE RESEARCH ETHICS BOARD

CERTIFICATION OF ETHICS APPROVAL

This is to certify that the University of Ottawa Health Sciences and Science Research Ethics Board (REB) examined the application for extension of ethics approval for the research project **Population Infant Hearing Screening to Intervention: Determinants of Outcome from the Parents' Perspectives (file H 09-05-07)** submitted by Elizabeth Fitzpatrick and supervised by Andrée Durieux-Smith of the Faculty of Health Sciences. This project received initial ethics approval on November 1, 2005 by the REB as meeting appropriate ethical standards set out in the Tri-Council Policy Statement and in the Procedures of the University of Ottawa Research Ethics Boards. The University of Ottawa REB members accordingly gave it a one-year extension of ethics approval. This ethics renewal certification is retroactive to November 1, 2006 and valid until November 1, 2007.

Rita D'Alessandro
Protocol Officer for Ethics in Research
For Dr. Daniel Lagarec, Chair of the
Health Sciences and Science REB

November 15, 2006
Date

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Université d'Ottawa University of Ottawa
Service de subventions de recherche et déontologie Research Grants and Ethics Services

November 15, 2006

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Object: **Population Infant Hearing Screening to Intervention: Determinants of Outcome from the Parents' Perspectives (file H 09-05-07)**

Dear Professor Durieux-Smith and Ms. Fitzpatrick,

You will find enclosed the Health Sciences and Science Research Ethics Board renewal certification for your research project above-mentioned.

During the course of the study, any modifications to the protocol or forms may not be initiated without prior written approval from the REB. You must also promptly report to the REB all adverse events or experiences encountered by participants.

The renewal certification is retroactive to November 1, 2006 and valid until November 1, 2007. Please submit an annual status report to the Protocol Officer in November, 2007 to either close the file or request a renewal of ethics approval. This document can be found at: http://www.rges.uottawa.ca/ethics/application_dwn.asp

A copy of this renewal approval will be sent to Research Services, if necessary.

Please do not hesitate to contact me at extension 5387 if you should have any questions.

Sincerely,

Rita D'Alessandro
Protocol Officer for Ethics in Research
For Dr. Daniel Lagarec, Chair of the Health Sciences and Science REB

01/53E

October 26, 2005

RECEIVED OCT 27 2005

Dr. Carole Gentile
Chair
Research Ethics Board
Children's Hospital of Eastern Ontario

Dear Dr. Gentile:

Re: Proposal 01/53E – The impact of screening and case finding on the functional status of children with a hearing impairment (Durieux-Smith et al.)

The CHEO Research Ethics Board recently approved an amendment (August 29, 2005) for the above research study, which involves expanding the study as part of my doctoral thesis work at the University of Ottawa. As a doctoral candidate at the University of Ottawa, the university REB also reviews the institutions' approved forms. The university has now completed its review and requires minor revisions to the Information Letter/Consent Form.

I am therefore attaching the revised forms for your review and re-approval. There are two consent forms, one for parents of children with hearing loss and one for parents of children with normal hearing. To facilitate your review, please find attached one copy with the revisions highlighted as well as one copy without highlighting. Thank you for your assistance with this project. I anxiously await your approval of the revised forms as the University of Ottawa REB will not issue an ethics clearance certificate until your approval is received.

Elizabeth Fitzpatrick, M.Sc. AUD(C)
Doctoral Candidate / Research Associate
University of Ottawa / CHEO Research Institute

CHEO Research Ethics Board
APPROVAL

Chair's Signature: _____

Date: November 8, 2005

The Hospital for Sick Children
Research Ethics Board

RECEIVED
DEC 12 2005

Amendment Request Form

1- REB File Number: 1000004514

2- Study Title: *The impact of screening and case finding on the functional status of children with a hearing impairment*

3- Describe the proposed study amendment or modification with rationale. For each item, please specify whether it is **Minor** eg, administrative changes such as deleting the name of a co-investigator, or **Major** eg, change in sponsorship that causes the investigator to have a conflict of interest, adding an intervention such as additional blood tests, or any substantive change that will be made to the consent form.
Please note; commercial sponsors will be charged a $500 REB review fee for amendments that require full Board review. An amendment to expand this project as part of E. Fitzpatrick's doctoral studies was recently approved (approval issued Aug 17, 2005), however, the University of Ottawa requires minor revisions to the Information Letter Consent form (attached).

4- Science Review: Science review may be needed for **major** amendments. If in doubt, please take advice with the REB Office. Please attach a copy of the completed science review form.

5- Will this amendment alter the study monitoring requirements?

● No

○ Yes **(please describe)** _____

6- What follow up action do you recommend for HSC study subjects who are already enrolled in the study?

○ Inform study subjects ASAP

○ Revise the consent/assent forms **(Please attach a copy with the changes highlighted)**

○ Other **(please describe)** _____

● No action Required – *No subjects have started this part of the study.*

7- Does this amendment alter the level of monitoring required for this study? If uncertain, please discuss with the Clinical Research Office staff; Julie Gibson or Velma Marzinotto.

○ Yes ● No ○ Perhaps

8- If Health Canada approved the original protocol (effective September 2001), their approval may also be required for this proposed amendment.

9- If the study sponsor requires a formal letter of approval, please attach a draft letter and forward an electronic copy as well.

10- Signature of Primary Investigator _____ Date 26/10/2005

11- Signature(s) of Co-Investigator(s)* _____ Date _____
***for Major amendments only**

12- Signature of Clinical Chief or Supervisor _____ Date 27/10/5

13- Approved & Signature of REB Chair _____ Date DEC 12 2005

August 1, 2005

Amendment Request - Hospital for Sick Children Research Ethics Board

Re: ***Proposal 1000004514 – The impact of screening and case finding on the functional status of children with a hearing impairment (Internal collaborator, Dr. Robert Harrison)***

The following amendments are requested for the above multi-centre study which has been ongoing at the Children's Hospital of Eastern Ontario (CHEO) since 2001 and since 2004 at two Toronto sites, the Hospital for Sick Children (HSC) and the Learning to Listen Foundation. To date, 66 children with hearing loss and their parents have been enrolled in the study from the three sites. An additional 51 children with normal hearing have been enrolled at the CHEO site.

Amendment 1 (minor):
Elizabeth Fitzpatrick, a doctoral candidate at the University of Ottawa and Research Associate at the CHEO Research Institute has been involved as a co-investigator at the Children's Hospital of Eastern Ontario since the beginning of the study. Elizabeth will be added to the HSC submission as a co-investigator.

Amendment 2 (major):
As a co-investigator on this research project, Elizabeth Fitzpatrick would like to expand the study protocol as part of a doctoral dissertation. Dr. Andrée Durieux-Smith, the principal investigator for the study, is co-supervising the doctoral thesis. Dr. Robert Harrison is the internal collaborator at the Hospital for Sick Children. The thesis proposal was defended and approved at the University of Ottawa on June 24, 2005 (Appendix 1). In addition to the current sites, children will be recruited for the doctoral study at the National Audiology Center, University of Western Ontario.

Specifically, the objectives of the thesis are: 1) to better understand the benefits of population hearing screening both through an examination of traditional clinical outcomes and parents' perspectives, 2) to identify the essential components of family-centred care and the relative value that parents place on the various attributes of service.

An overview of the four interrelated inquiries for the doctoral thesis including the objectives, research questions, and research methods are attached in Appendix 2. Inquiry 1 involves a systematic review of the literature. Inquiry 2 is part of the original project. *Inquiries 3 and 4* are the amendments to the original project. A brief summary including ethical considerations is provided for the three inquiries which involve patient data:

1) Inquiry 2: This inquiry involves a preliminary analysis of the communication outcome data already collected as part of the original project. The purpose of this preliminary analysis is to examine trends in age of identification and communication development outcomes. Data collection for this project which has already been approved by the HSC Ethics Board is ongoing (Proposal 1000004514).

2) Inquiry 3: This phase involves qualitative interviews with approximately 6-8 parents in the Toronto centers who are enrolled in the current study (total of 18-20 across all sites). The objective of these semi-structured individual interviews is to explore the benefits of early identification of hearing loss and to identify parents' needs for health services following diagnosis. Interviews will take place in the parents' home unless a parent prefers a meeting in another appropriate milieu such as the Audiology Clinic. A copy of the interview guide is attached (Appendix 3).

3) Inquiry 4: The final inquiry involves a conjoint analysis questionnaire to be mailed to all parents of children with hearing loss and parents of hearing children who are enrolled in the existing study and who consent to participate. The purpose of this survey is to elicit and quantify parents' preferences for service delivery. The conjoint analysis survey will be constructed based on the data collected during the qualitative interviews.

No additional patient recruitment is required for these additional components of the study. Elizabeth Fitzpatrick will contact parents of children with hearing loss who are currently enrolled in the study from the HSC site using the attached invitation letter (Appendix 4). A consent form is attached in Appendix 5.

The collaboration from HSC over the past year is greatly appreciated. Further details on any aspects of the study will be provided as required.

Research Team:

Elizabeth Fitzpatrick, M.Sc.
PhD Candidate, University of Ottawa
Research Associate
CHEO Research Institute

Andrée Durieux-Smith, PhD
Principal Investigator
Faculty of Health Sciences
University of Ottawa

Dr. Robert Harrison, PhD
Internal Collaborator
Hospital for Sick Children

University of Ottawa Doctoral Thesis Committee:
Doug Angus, MA, Institute of Population Health
Andrée Durieux-Smith, PhD, Faculty of Health Sciences
Ian Graham, PhD, Faculty of Nursing
Doug Coyle, PhD, Dept. of Epidemiology and Community Medicine

Dear Elizabeth:With apologies for the delay, I am writing to respond to your letter requesting formal written approval of LTLF to your continuing with the Project and continuing to involve LTLF families in your study. As you know, LTLF has never established a Research Ethics Board {"REB"}, primarily because we do not get involved in research of the kind you are undertaking. As a result, we put your original proposal to the REB of North York General Hospital, only to discover that the process was extremely lengthy and unlikely to result in a decision without an enormous amount of due diligence and effort.After discussion with you, we decided to put the proposal to our Board of Directors who agreed that, assuming written consents from all participating families were obtained and adequate privacy protections were in place, there was no objection to the Project proceeding in the manner outlined.You have now indicated that you wish to have the decision of our Board, in place of an REB, renewed. I am writing to confirm that the the original Board determination to approve remains in effect and, while there is an extension to the Project through an expanded study programme, there seems to be no material change in the purpose and objectives of the Project and, therefore, no reason to change the decision of the Board previously given. I trust this note will be sufficient to allow you to proceed with the Project as described in your recent correspondence.Kind regards,

John CraigPresidentLearning to Listen FoundationJohn CraigPartnerMcMillan Binch Mendelsohn LLPBCE Place, Suite 4400Bay Wellington Tower181 Bay StreetToronto, Ontario M5J 2T3Direct Tel: 416.865.7128Toll Free: 1.888.622.4624Fax: 416.865.7048john.craig@mcmbm.comwww.mcmbm.com

Office of Research Ethics
The University of Western Ontario
Room 00045 Dental Sciences Building, London, ON, Canada N6A 5C1
Telephone: (519) 661-3036 Fax: (519) 850-2466 Email: ethics@uwo.ca
Website: www.uwo.ca/research/ethics

Use of Human Subjects - Ethics Approval Notice

Principal Investigator: Dr. R. Seewald
Review Number: 11709E **Revision Number:**
Protocol Title: Population infant hearing screening to intervention: Determinants of outcomes from the parents' perspectives
Department and Institution: Audiology, University of Western Ontario
Sponsor:
Ethics Approval Date: September 16, 2005 **Expiry Date:** August 31, 2007
Documents Reviewed and Approved: UWO Protocol, Letter of Information & Consent
Documents Received for Information:

This is to notify you that The University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines; and the applicable laws and regulations of Ontario has reviewed and granted expedited approval to the above named research study on the approval date noted above. The membership of this REB also complies with the membership requirements for REB's as defined in Division 5 of the Food and Drug Regulations.

This approval shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB's periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

During the course of the research, no deviations from, or changes to, the protocol or consent form may be initiated without prior written approval from the HSREB except when necessary to eliminate immediate hazards to the subject or when the change(s) involve only logistical or administrative aspects of the study (e.g. change of monitor, telephone number). Expedited review of minor change(s) in ongoing studies will be considered. Subjects must receive a copy of the signed information/consent documentation.

Investigators must promptly also report to the HSREB:
a) changes increasing the risk to the participant(s) and/or affecting significantly the conduct of the study;
b) all adverse and unexpected experiences or events that are both serious and unexpected;
c) new information that may adversely affect the safety of the subjects or the conduct of the study.

If these changes/adverse events require a change to the information/consent documentation, and/or recruitment advertisement, the newly revised information/consent documentation, and/or advertisement, must be submitted to this office for approval.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

Chair of HSREB: Dr. John W. McDonald
Deputy Chair: Susan Hoddinott

Ethics Officer to Contact for Further Information			
☒ Karen Kueneman	☐ Janice Sutherland	☐ Susan Underhill	☐ Jennifer McEwen

This is an official document. Please retain the original in your files.

cc: ORE File
Faxed: Y / N

Appendix 3

Informed Consent – Children's Hospital of Eastern Ontario

Informed Consent – Hospital for Sick Children

Informed Consent - Learning to Listen Foundation

Informed Consent – University of Western Ontario

Information Letter

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

Dear Parent:

We are inviting you to participate in a research project aimed at learning more about the process of early identification of hearing loss and the services needed after the diagnosis. This research is being conducted as part of Elizabeth Fitzpatrick's doctoral studies in the Population Health Program at the University of Ottawa under the supervision of Professors Doug Angus and Andrée Durieux-Smith. The investigator, Elizabeth Fitzpatrick is also a Research Associate at the Children's Hospital of Eastern Ontario (CHEO) Research Institute.

You are being invited to participate as a parent who is already enrolled in a research project being carried out by researchers at CHEO and the University of Ottawa. In this ongoing project, researchers are examining whether the age at which a hearing loss is diagnosed has an effect on the child's communication and learning abilities

We are inviting you to participate in this new research project which will expand on the information being collected in the project in which you are currently enrolled. In this new project, we would like to learn about: your experiences with the diagnosis of your child's hearing loss, your needs after learning of the hearing loss, and your views on the important aspects of services for you and your child. In 2002, the Ontario Ministry of Health and Long Term Care began an Infant Hearing Program (IHP) for all babies born in Ontario. Since that time, programs have been announced in other Canadian provinces. The finding of this study will provide information which will help programs in designing appropriate services for young children with hearing impairment.

This study involves two phases:
1) Interviews with you to better understand the experiences around diagnosis, how early or late diagnosis affects you and your child, and your perceptions of your needs after diagnosis. As well, we would like to learn what is important to you in a system of care for you and your child. If you agree to take part, we will meet with you at your home (or other convenient setting) for a 45 to 60 minute interview to ask you some questions about your experiences and thoughts on these topics. The interviews will be audio-recorded. The interviews will be scheduled at your convenience.
2) Based on the interview information, a questionnaire will be developed to investigate the important characteristics of services after a child's hearing loss is diagnosed. We will ask you to complete this short questionnaire, which is expected to take 20 to 30 minutes, by email or paper mail. The questions will ask you to make choices about the services that are important in providing you with support in developing your child's communication skills.

Please note: The number of parents required for part one of the study is smaller than that required for the second part of the study. For the interviews, we need participation from as many different parents as possible, for example parents of children with different degrees of hearing loss, different hearing devices and different ages of diagnosis. Therefore, all parents who indicate an interest will be contacted but not all parents will be asked to do the interview. However, <u>all</u> parents will be contacted and asked to complete the questionnaire in part 2 of the study.

In addition, we would like to access the questionnaires and other demographic data that have been collected as part of the study, in which you are currently participating. All information will be kept strictly confidential. Your identity will not be disclosed to any person, except in the event of a medical emergency or if required by law. Your name or the name of your child will not be identified in any publication, report or presentations resulting from this study. Comments from the interviews (both verbal and written) may be quoted but you will not be identified.

The data produced from this study will be carefully stored on computer with password protection or in the Audiology Lab at the Children's Hospital of Eastern Ontario Research Institute. Only members of the research team will have access to the data. The data will be destroyed 5 years after publication of the results.

If you would like to participate in this study, please contact Elizabeth Fitzpatrick. You will be asked to sign the attached consent form. If you have any questions, you may contact Elizabeth at 613-745-9591 or or by email at: . Elizabeth will contact you shortly to ask whether you are interested in participating in the study. If you choose not to participate or decide to withdraw from the study, this will in no way affect the care received by your child at the Children's Hospital of Eastern Ontario and the data collected will not be used without your consent.

Elizabeth Fitzpatrick, M.Sc.
PhD Candidate, University of Ottawa
Research Associate, Children's Hospital of Eastern Ontario Research Institute
Tel.
Email:

Research Consent Form

I acknowledge that the research procedures described above, and of which I have a copy, have been explained to me. I have been given an opportunity to ask questions concerning the study process and the questions, which I have asked, have been adequately answered. In addition, I know that I may contact the principle investigator (whose contact information is below) if I have further questions either now or in the future. I have been assured that personal records relating to this study will be kept confidential. I understand that my identity will not be disclosed to any person, except in the event of a medical emergency or if required by law. I understand that I can withdraw my consent and stop my participation in the study at any time and for any reason. I have been reassured that this will not affect the quality of care that I or any member of my family receives at the Children's Hospital of Eastern Ontario. I understand that if any knowledge gained from this study becomes available that could influence my decision to continue in this study, I will be promptly informed.

I may contact the Chair of the Research Ethics Board for information regarding participant's rights in research studies at (613-737-7600, x3272) however, this person cannot provide any medical information with regard to this study. If I have any questions with regards to the ethical conduct of this study I may contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 159, Ottawa, ON K1N 6N5, tel.: 613-562-5841, email: ethics@uottawa.ca.

I have received a copy of the Information Letter hereto attached and I have received a copy of this consent form.

I, _____, consent to participation in the study described above.

_____Date_____Tel._____Email_____
Signature of parent or guardian)

Name of witness _____
(printed)

Signature _____ Date _____

I HAVE EXPLAINED THIS STUDY TO THE PARTICIPANT AND I AM SATISFIED THAT IT IS UNDERSTOOD.
Name and Title _____
(printed)
Signature _____ Date _____

Research Team

Elizabeth Fitzpatrick, AUD(C)
Principal Investigator
Population Health Ph.D. Program
University of Ottawa
Ema:
Tel:

Doctoral Thesis committee (University of Ottawa):
Doug Angus, M.A., Institute of Population Health
Andrée Durieux-Smith, Ph.D., Faculty of Health Sciences
Ian Graham, Ph.D, Ottawa Health Research Institute
Doug Coyle, Ph.D, Department of Epidemiology and Community Medicine

Research Consent Form

Title of Research Project:

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

Investigator(s):

Robert V. Harrison, Ph.D., Hospital for Sick Children, Toronto
(416) 813-6535

Elizabeth Fitzpatrick, Doctoral Candidate, University of Ottawa

Doctoral Thesis Committee:

Andrée Durieux-Smith, Ph.D., University of Ottawa, Ottawa, Co-Supervisor
(613) 562-5800, ext. 8017
Doug Angus, M.A., University of Ottawa, Ottawa, Co-Supervisor
(613) 562-5800, ext. 4720
Ian Graham, Ph.D., University of Ottawa, Ottawa
(613) 562-5800
Doug Coyle, Ph.D., University of Ottawa, Ottawa
(613) 562-5800

Purpose of the Research:

This research is being conducted as part of Elizabeth Fitzpatrick's doctoral studies in the Population Health Program at the University of Ottawa under the supervision of Professors Doug Angus and Andrée Durieux-Smith. The investigator, Elizabeth Fitzpatrick is also a Research Associate at the Children's Hospital of Eastern Ontario (CHEO) Research Institute and has been involved in the original research project in which you are already enrolled: *The impact of case finding on the functional status of children with a hearing impairment.* These new aspects of the project will expand on the information being collected in the project in which you are currently participating.

The new parts of this research project are aimed at learning more about the process of identification of your child's hearing loss and the services needed after the diagnosis. In this new project, we would like to learn about: your experiences with the diagnosis of your child's hearing loss, your needs after learning of the hearing loss, and your views on the important aspects of services for you and your child. In 2002, the Ontario Ministry of Health and Long Term Care began an Infant Hearing Program (IHP) for all babies born in Ontario. Since that

Learning to Listen Foundation

Tel: 416-491-4648
Fax: 416-491-7215

Information Letter

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

Dear Parents,

You are being invited to participate in a study which expands on a research project entitled *The impact of case finding on the functional status of children with a hearing impairment* in which researchers at the Children's Hospital of Eastern Ontario, the University of Ottawa and the University of Toronto are examining whether the age at which a hearing loss is diagnosed has an effect on the child's communication and learning abilities.

We are inviting you to participate in new parts of this research project aimed at learning more about the process of identification of your child's hearing loss and the services needed after the diagnosis. This research is being conducted as part of Elizabeth Fitzpatrick's doctoral studies in the Population Health Program at the University of Ottawa, under the supervision of Professors Doug Angus and Andrée Durieux-Smith. The investigator, Elizabeth Fitzpatrick is also a Research Associate at the Children's Hospital of Eastern Ontario (CHEO) Research Institute and has been involved in the original project.

In this new project, we would like to learn about: your experiences with the diagnosis of your child's hearing loss, your needs after learning of the hearing loss, and your views on the important aspects of services for you and your child. In 2002, the Ontario Ministry of Health and Long Term Care began an Infant Hearing Program (IHP) for all babies born in Ontario. Since that time, programs have been announced in other Canadian provinces. The findings of this study will provide information which may help programs in providing appropriate services for young children with hearing impairment.

This new part of the study involves two phases:
1) Interviews with you to better understand the experiences around diagnosis, how early or late diagnosis affects you and your child, and your perceptions of your needs after diagnosis. As well, we would like to learn what is important to you in a system of care for you and your child. If you agree to take part, we will meet with you at your home (or other convenient setting) for a 45 to 60 minute interview to ask you some questions about your experiences and thoughts on these topics. The interviews will be audio-recorded. The interviews will be scheduled at your convenience.

2) Based on the interview information, a questionnaire will be developed to investigate the important characteristics of services after a child's hearing loss is diagnosed. We will ask you to complete this short questionnaire, which is expected to take 20 to 30 minutes, by email or paper mail. The questions will ask you to make choices about the services that are important in providing you with support in developing your child's communication skills.

Please note: The number of parents required for part one of the study is smaller than that required for the second part of the study. For the interviews, we need participation from as many different parents as possible, for example parents of children with different degrees of hearing loss, different hearing devices and different ages of diagnosis. Therefore, all parents who indicate an interest will be contacted but not all parents will be asked to do the interview. However, <u>all</u> parents will be contacted and asked to complete the questionnaire in part 2 of the study.

Consent :

By signing this form, I agree that:

1) You have explained this study to me. You have answered all my questions.
2) You have explained the possible harms and benefits (if any) of this study.
3) I know what I could do instead of taking part in this study. I understand that I have the right not to take part in the study and the right to stop at any time. My decision about taking part in the study will not affect my health care at Sick Kids.
4) I am free now, and in the future, to ask questions about the study.
5) I have been told that my medical records will be kept private. You will give no one information about me, unless the law requires you to.
6) I understand that no information about who I am will be given to anyone or be published without first asking my permission.
7) I have read and understood pages 1 to 4 of this consent form. I agree, or consent, to take part in this study.

Printed Name of Subject & Age	Subject's signature & date
Printed Name of person who explained consent	Signature & date
Printed Witness' name (if the subject/legal guardian does not read English)	Witness' signature & date

If you have any questions about this study, please contact Elizabeth Fitzpatrick at or by email at or contact Dr. Robert V. Harrison at the Hospital for Sick Children at (416) 813-6535. If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-813-5718. If you have any questions with regards to the ethical conduct of this study you may contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 159, Ottawa, ON K1N 6N5, tel.: 613-562-5841, email: ethics@uottawa.ca.

In addition, we would like to complete a brief family information questionnaire. All information will be kept strictly confidential.

If you would like to participate in this study, please sign the consent form. If you have any questions, you may contact Elizabeth Fitzpatrick directly at by email at

Thank you for your attention,

Elizabeth Fitzpatrick M.Sc., Cert AVT®
Doctoral Candidate
University of Ottawa

Warren Estabrooks, M.Ed., Cert AVT® (Internal Collaborator)
Director
Auditory Learning Centre
Learning to Listen Foundation
(416) 491-4648 ext 8239

Mila De Melo, M. Fga., S-LP, Aud(C), Cert AVT® (Internal Collaborator)
Coordinator of Research and Development
Auditory Learning Centre
Learning to Listen Foundation
(416) 491- 4648 ext 8243

Learning to Listen Foundation

Tel: 416-491-4648
Fax: 416-491-7215

Research Consent Form

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

Investigator(s):

Elizabeth Fitzpatrick, M.Sc., Cert AVT®
Doctoral Candidate, University of Ottawa

Warren Estabrooks, M.Ed., Cert AVT® (Internal Collaborator)
Director
Auditory Learning Centre
Learning to Listen Foundation, Toronto
(416) 491-4648 ext 8239

Mila De Melo, M. Fga., S-LP, Aud(C), Cert AVT® (Internal Collaborator)
Coordinator of Research and Development
Auditory Learning Centre
Learning to Listen Foundation, Toronto
(416) 491- 4648 ext 8243

Doctoral Thesis Committee:
Andrée Durieux-Smith, Ph.D., University of Ottawa, Ottawa, Co-Supervisor
(613) 562-5800, ext. 8017
Doug Angus, M.A., University of Ottawa, Ottawa, Co-Supervisor
(613) 562-5800, ext. 4720
Ian Graham, Ph.D., University of Ottawa, Ottawa
(613) 562-5800
Doug Coyle, Ph.D., University of Ottawa, Ottawa
(613) 562-5800

Purpose of the Research:
This research is being conducted as part of Elizabeth Fitzpatrick's doctoral studies in the Population Health Program at the University of Ottawa, under the supervision of Professors Doug Angus and Andrée Durieux-Smith. The investigator, Elizabeth Fitzpatrick is also a Research Associate at the Children's Hospital of Eastern Ontario (CHEO) Research Institute and has been involved in the original research project, *The impact of case finding on the functional status of children with a hearing impairment.* These new aspects of the project will expand on the information being collected in the project.

The new parts of this research project are aimed at learning more about the process of identification of your child's hearing loss and the services needed after the diagnosis. In this new project, we would like to learn about: your experiences with the diagnosis of your child's hearing

loss, your needs after learning of the hearing loss, and your views on the important aspects of services for you and your child. In 2002, the Ontario Ministry of Health and Long Term Care

began an Infant Hearing Program (IHP) for all babies born in Ontario. Since that time, programs have been announced in other Canadian provinces. The findings of this study will provide information, which may help programs in providing appropriate services for young children with hearing impairment.

Description of the Research:
This new part of the study involves two phases:
1) Interviews with you to better understand the experiences around diagnosis, how early or late diagnosis affects you and your child, and your perceptions of your needs after diagnosis. As well, we would like to learn what is important to you in a system of care for you and your child. If you agree to take part, we will meet with you at your home (or other convenient setting) for a 45 to 60 minute interview to ask you some questions about your experiences and thoughts on these topics. The interviews will be audio-taped. The interviews will be scheduled at your convenience.

2) Based on the interview information, a questionnaire will be developed to investigate the important characteristics of services after a child's hearing loss is diagnosed. We will ask you to complete this short questionnaire, which is expected to take 20 to 30 minutes, by email or paper mail. The questions will ask you to make choices about the services that are important in providing you with support in developing your child's communication skills.

Please note: The number of parents required for part one of the study (interviews) is smaller than that required for the second part of the study. For the interviews, we need participation from as many different parents as possible, for example parents of children with different degrees of hearing loss, different hearing devices and different ages of diagnosis. Therefore, all parents who indicate an interest will be contacted but not all parents will be asked to do the interview. However, all parents will be contacted and asked to complete the questionnaire in part 2 of the study.

In addition, we would like you to complete a brief family information questionnaire. This information will be used to summarize the characteristics of children whose parents are participating in this part of the study. All information will be kept strictly confidential.

New information from this study or other studies may affect whether you want to continue to take part in the study. If this happens, we will tell you about this new information.

Potential Harms:
We know of no harm that taking part in this study could cause you.

Potential Discomforts or Inconvenience:
Both parts of this research are non-invasive and should not cause any discomfort. The interviews are about 45 minutes and will take place in your home whenever possible and at a time that is convenient for you. The questionnaire can be returned by email or paper email.

Potential Benefits:

To individual subjects:
You (your child) will not benefit directly from participating in this study. You will be provided with a summary of the group findings for this research at the end of the project.

To society:
The findings may provide decision makers with information about what services are important to parents of children with hearing loss and therefore may influence decisions about which services public health programs should provide. The information from this study may also help programs in providing appropriate services for young children with hearing impairment and their parents.

Confidentiality:
We will respect your privacy. Comments from the interviews (both verbal and written) may be quoted but you will not be identified. No information about who you are will be given to anyone or be published without your permission, unless the law makes us do this. For example, the law could make us give information about you

- If a child has been abused
- If you have an illness that could spread to others
- If you or someone else talks about suicide (killing themselves), or
- If the court orders us to give them the study papers

We will put a copy of this research consent form in your child's Progress Record at the Auditory Learning Centre, Learning to Listen Foundation.

The data produced from this study will be carefully stored on computer with password protection or in the Audiology Lab at the Children's Hospital of Eastern Ontario Research Institute. Only members of the research team will have access to the data.. The data will be destroyed 5 years after the publication of the research.

Reimbursement:
We do not anticipate that there will be any expenses related to taking part in this study as the interviews will take place in the home. The questionnaire can be emailed or sent by paper mail, in which case, a stamped envelope will be provided.

Participation:
It is your choice to take part in this study. You can stop at any time and the data collected will not be used without your consent. The care you get at the Learning to Listen Foundation will not be affected in any way by whether you take part in this study.

New information that we get while we are doing this study may affect your decision to take part in this study. If this happens, we will tell you about this new information. And we will ask you again if you still want to be in the study.

If you become ill or are harmed because you took part in this study, we will treat you for free. Your signing this consent form does not interfere with your legal rights in any way. The staff of the study, any people who gave money for the study, or the hospital are still responsible, legally and professionally, for what they do.

Sponsorship:

The funder for the original research project in which you are participating is the Canadian Language and Literacy Research Network. These new aspects of the project have not received additional funding. As a doctoral student, Elizabeth Fitzpatrick has received fellowships from the Social Sciences and Humanities Research Council and Advanced Bionics Corporation.

Consent:

By signing this form, I agree that:

1) You have explained this study to me. You have answered all my questions.
2) You have explained the possible harms and benefits (if any) of this study.
3) I know what I could do instead of taking part in this study. I understand that I have the right not to take part in the study and the right to stop at any time. My decision about taking part in the study will not affect my health care at the Learning to Listen Foundation.
4) I am free now, and in the future, to ask questions about the study.
5) I have been told that my medical records will be kept private. You will give no one information about me, unless the law requires you to.
6) I understand that no information about who I am will be given to anyone or be published without first asking my permission.

7) I have read and understood pages 1 to 4 of this consent form. I agree, or consent, to take part in this study.

Printed Name of Subject & Age	Subject's signature & date
Printed Name of person who explained consent	Signature & date
Printed Witness' name (if the subject/legal guardian does not read English)	Witness' signature & date

If you have any questions about this study, please contact either Elizabeth Fitzpatrick at [redacted] or by email at [redacted], Warren Estabrooks at (416) 491-4648 ext. 8239, or Mila de Melo at (416) 491-4648 ext.8243. If you have any questions with regards to the ethical conduct of this study you may contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 159, Ottawa, ON K1N 6N5, tel.: 613-562-5841, email: ethics@uottawa.ca.

University of Western Ontario Letter of Information/Consent

Letter of Information

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

We are inviting you to participate in a research project aimed at learning more about the early identification of hearing loss in children and the services needed after the diagnosis. This research is being conducted as part of Elizabeth Fitzpatrick's doctoral studies in the Population Health Program at the University of Ottawa under the supervision of Professors Doug Angus and Andrée Durieux-Smith. The investigator, Elizabeth Fitzpatrick is also a Research Associate at the Children's Hospital of Eastern Ontario (CHEO) Research Institute. Parents from several Ontario sites are being invited to participate in this project. The H.S. Leeper Speech and Hearing Clinic at the University of Western Ontario (UWO), through Dr. Richard Seewald and Dr. Marlene Bagatto has agreed to be involved with the study.

In 2002, the Ontario Ministry of Health and Long Term Care began an Infant Hearing Program (IHP) for all babies born in Ontario. Since that time, programs have been announced in other Canadian provinces. Researchers at the Children's Hospital of Eastern Ontario, the University of Ottawa and the University of Toronto are currently conducting a research project with children and their families to examine whether the age at which a hearing loss is diagnosed has an effect on the child's communication and learning abilities. We are now collecting new information that will expand the project described above. We would like to learn about: your experiences with the diagnosis of your child's hearing loss, your needs after learning of the hearing loss, and your views on the important aspects of services for you and your child. The finding of this study will provide information that may help programs design appropriate services for young children with hearing impairment. This study involves two phases:

1) Interviews with you to better understand the experiences around diagnosis, how early or late diagnosis affects you and your child, and your thoughts about your needs after diagnosis. As well, we would like to learn what is important to you in a system of care for you and your child. We will include parents of children whose hearing loss was identified through screening and without screening. If you agree to take part, Elizabeth Fitzpatrick will meet with you at your home (or other convenient place) for a 45 to 60 minute interview to ask you some questions about your experiences and thoughts on these topics. The interviews will be audio-recorded. The interviews will be scheduled at your convenience.

Please note: For the interviews, we need participation from parents of children with different characteristics, for example children with different degrees of hearing loss, different hearing devices and different ages of diagnosis. Therefore, all parents who indicate an interest will be contacted but not all will be asked to do the interview. However, <u>all</u> parents will be asked to complete the questionnaire in part 2 of the study.

2) Based on the interview information, a questionnaire will be developed to investigate the important characteristics of services after a child's hearing loss is diagnosed. We will ask you to complete this short questionnaire by email or paper mail. The questions will ask you to make choices about the services that are important in providing you with support in developing your child's communication skills. The questionnaire is expected to take about 20 to 30 minutes. In addition, you will be asked to complete two short questionnaires to collect information about your child's hearing loss and family characteristics. All information will be kept strictly confidential and will be carefully stored on computer with password protection or in the Audiology Lab at the Children's Hospital of Eastern Ontario Research Institute. You or your child will not be identified in any discussion, publication or presentation of the results unless you give specific consent. Comments from the interviews (both verbal and written) may be quoted but you will not be identified.

Participation in this study is voluntary. You may refuse to participate, refuse to answer any questions or withdraw from the study at any time with no effect on your child's future care. The data collected will not be used without your consent. If you have any questions about your rights as a research participant or the conduct of the study you may contact the Director of the Office of Research Ethics at The University of Western Ontario (519)661-3036 or ethics@uwo.ca. If you have any questions with regards to the ethical conduct of this study you may also contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 159, Ottawa, ON K1N 6N5, tel.: 613-562-5841, email: ethics@uottawa.ca. There are no known risks to your participation in this study. Representatives of the University of Western Ontario Health Sciences Research Ethics Board may require access to your study-related records or may follow-up with you to monitor the conduct of the research.

With your consent, your clinic will give Elizabeth Fitzpatrick your telephone number so that she can contact you regarding this study. If you have any questions, you may also contact Elizabeth Fitzpatrick at or by email at: . If you agree to participate in this study, we will ask you to sign the attached consent form.

Thank you for your attention,

Elizabeth Fitzpatrick, M.Sc.
Audiologist and Ph.D Candidate
University of Ottawa

Richard Seewald, Ph.D
National Audiology Centre
University of Western Ontario

Consent Form

Population infant hearing screening to intervention: Determinants of outcome from the parents' perspectives

I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction.

Printed Name of Participant	Signature of Participant	Date
Name of Person Obtaining Informed Consent	Signature of Person Obtaining Informed Consent	Date

Research Team
Elizabeth Fitzpatrick, AUD(C)
Ph.D Candidate, Population Health Program
University of Ottawa

Dr. Richard Seewald
Dr. Marlene Bagatto
National Centre for Audiology
University of Western Ontario

Doctoral Thesis Committee
University of Ottawa:
Doug Angus, M.A.
Andrée Durieux-Smith, Ph.D
Ian Graham, Ph.D
Doug Coyle, Ph.D

Appendix 4

Manuscript 1 Submission Letter: Journal of Medical Screening

Manuscript 2 Decision Letter: International Journal of Audiology

Manuscript 3 Submission Letter: American Journal of Audiology

Manuscript 4 Submission Letter: Ear and Hearing

You replied on 16/10/2006 6:07 PM.

Fitzpatrick, Elizabeth

From: Jan Mackie [**Sent:** Mon 16/10/2006 5:22 AM
To: Fitzpatrick, Elizabeth
Cc:
Subject: RE: Journal of Medical Screening - Manuscript 06098
Attachments:

Dear Dr Fitzpatrick

Manuscript 06098

Thank you for sending us your manuscript entitled "The impact of newborn hearing screening on communication development", which is being considered for publication. We shall contact you again when the paper has been reviewed.

Yours sincerely

Janette Mackie
Editorial Office
J Med Screen

You forwarded this message on 25/08/2006 6:23 PM.

Fitzpatrick, Elizabeth

From: [redacted] **Sent:** Fri 25/08/2006 6:20 PM
To: Fitzpatrick, Elizabeth;
Cc:
Subject: International Journal of Audiology - Decision on Manuscript ID TIJA-2006-05-0061.R1
Attachments:

25-Aug-2006

Dear Ms. Fitzpatrick:

It is a pleasure to accept your manuscript entitled "Parents' Perspectives on the Impact of the Early Diagnosis of Childhood Hearing Loss" in its current form for publication in the International Journal of Audiology. The comments of the reviewer(s) who reviewed your manuscript are included at the foot of this letter.

Thank you for your fine contribution. On behalf of the Editors of the International Journal of Audiology, we look forward to your continued contributions to the Journal.

Sincerely,
Prof. Ross Roeser
Editor in Chief, International Journal of Audiology
roeser@utdallas.edu

Reviewer(s)' Comments to Author:

You forwarded this message on 24/11/2006 2:13 PM.

Fitzpatrick, Elizabeth

From: **Sent:** Thu 23/11/2006 1:50 PM
To: Fitzpatrick, Elizabeth
Cc:
Subject: AJA-06-0035 successfully submitted
Attachments:

Dear Ms. Fitzpatrick,

Thank you for submitting your new or revised manuscript "Parents' Needs Following Identification of Childhood Hearing Loss" to the The American Journal of Audiology. The manuscript will be assigned to an Associate Editor who will oversee its review.

You can check the status of your manuscript at any time by logging on to your AJA Author Center at http://mc.manuscriptcentral.com/ajoa .

If you have any questions about your manuscript during the review process, please contact the Editorial Administrator at 301-897-5700, ext. 4600, or e-mail aja@asha.org.

Please refer to your manuscript with its manuscript number, AJA-06-0035, in all future correspondence.

Information on ASHA's submission policies can be found in the printed journal as well as online at http://aja.asha.org/.

ASHA journals publish scholarly papers ranging from data-based research reports to reviews and tutorials that present no new data. Notwithstanding the differences in these types of papers, they all must contain a structured abstract.

If you are invited to resubmit your manuscript, the Editor will ask you to reformat your abstract if it does not already comply with this requirement. To read more about structured abstracts, visit http://aja.asha.org/misc/ifora.dtl#preparing.

Once again, I thank you for the opportunity to review your work. Let me know if I can assist you at any stage of the review process.

Sincerely,
The American Journal of Audiology Editorial Office

You forwarded this message on 21/12/2006 7:55 PM.

Fitzpatrick, Elizabeth

From: Ear and Hearing **Sent:** Thu 21/12/2006 7:53 PM
To: Fitzpatrick, Elizabeth
Cc:
Subject: EANDH Submission Confirmation for Parents' Preferences for services for children with hearing loss: A conjoint analysis study
Attachments:

Dear Research Associate Fitzpatrick,

Your submission entitled "Parents' Preferences for services for children with hearing loss: A conjoint analysis study" has been received by the journal editorial office.

You will be able to check on the progress of your paper by logging on to Editorial Manager as an author.

http://eandh.edmgr.com/

Username: efitzpatrick
Password: fitzpatric235

Your manuscript will be given a reference number once an Editor has been assigned.

Thank you for submitting your work to this journal.

Kind Regards,

Ear and Hearing

Appendix 5

Intake Questionnaire

Intake Questionnaire

Date: _____

Name of person answering this questionnaire: _____

Relationship to the child: Mother ☐ Father ☐ Other ☐ _____

Child's Name Last: _____ First: _____ Initial: _____

Date of Birth ___ ___/___ ___ ___/___ ___
DAY / MONTH / YEAR

Gender: ☐ Male ☐ Female

Mother's Name Last: _____ First: _____ Initial: ____

Phone (home): _____ Phone (work): _____

e-mail: _____

Street: _____

City: _____ Prov: _____ Postal Code: _ _ _ _ _ _

Father's Name Last: _____ First: _____ Initial: ____

Was your child born earlier than expected? YES NO

If *yes*, how many weeks early was the birth? _____

What was your expected due date? ___ ___/___ ___ ___/___ ___
DAY / MONTH / YEAR

Has your child visited an ear nose and throat specialist in the past year because of problems with his/her ears? YES NO

Has your child had surgery to have tubes put in his/her ears in the past year? YES NO

Has your child had ear infections in the past 12 months? YES NO

How many ear infections has your child had in the last year? _____

Do you have any concerns about your child's development? YES NO

Is your child being followed by a professional for developmental concerns? YES NO

Are you aware of any children or adults in your family who have a hearing loss, e.g. brothers, sisters or cousins of your child or yourself? *If yes, see the next page for additional questions* YES NO

Reference: The impact of screening and case finding on the functional status of children with a hearing impairment

Family History of Hearing Loss Questionnaire

What is the relationship of the family member with hearing loss to your child?

_____

Was this hearing loss suspected to be present from birth? Yes ☐
No ☐

If no, at what age was the hearing loss suspected? _____

At what age was the hearing loss confirmed? _____

Did this hearing loss follow a difficult birth or admission to a neonatal intensive care unit? Yes☐
No ☐

Did this hearing loss follow a serious childhood illness, e.g. meningitis, cancer, measles, mumps? Yes ☐
No ☐

Did this hearing loss follow chronic ear infections? Yes☐
No ☐

Is there a suspected cause for the hearing loss? Yes☐
No ☐

Does this person wear a hearing aid(s)? Yes☐
No ☐

Does this person use a cochlear implant? Yes☐
No ☐

Do they use services such as closed captioning to watch TV? Yes☐
No ☐

Does this person have speech that is difficult to understand? Yes☐
No ☐

Does this person use sign language? Yes☐
No ☐

Reference: The impact of screening and case finding on the functional status of children with a hearing impairment

Appendix 6

Family Information Questionnaire

Family Information Questionnaire

To help us understand all of the data we are collecting we would like to ask you to provide some information about your family. Your answers will be completely confidential and your identity will not be revealed to any person. This information will only be reported as group summaries.

Who is completing this questionnaire?

☐ Mother ☐ Father ☐ Both ☐ Other (please specify)_____

What is the structure of your family?

☐ Single parent (joint custody) ☐ Two parent ☐ Foster parent(s)
☐ Single parent (sole custody) ☐ Adoptive parent(s) ☐ Other

Please list the gender and year of birth for all children living in your household. Please indicate how these children are related, (e.g. sister, brother, half brother or sister, step sister or brother, etc.)

Gender	Year of birth (e.g. 1976)	Relationship to other children in the home
☐ male ☐ female		
☐ male ☐ female		
☐ male ☐ female		
☐ male ☐ female		
☐ male ☐ female		
☐ male ☐ female		

What is the language most commonly used in your home? (Please check all that apply)

Mom	Dad		Mom	Dad		Mom	Dad	
☐	☐	English	☐	☐	Signed English	☐	☐	Portuguese
☐	☐	French	☐	☐	Greek	☐	☐	Vietnamese
☐	☐	Arabic	☐	☐	Hungarian	☐	☐	Ukrainian
☐	☐	Chinese	☐	☐	Italian	☐	☐	Punjabi
☐	☐	Cree	☐	☐	Korean	☐	☐	Spanish
☐	☐	German	☐	☐	Persian (Farsi)	☐	☐	Tagalog (Filipino)
☐	☐	ASL	☐	☐	Polish			
☐	☐	Other (please specify) _____						

Who cares for your child during the day?

		How many days per week?	How many children share this daycare?
	Mom or Dad at home		
	Another family member		
	Home-based daycare		
	Daycare centre or preschool		
	School (for example JK or SK)		
	Other _____		

What is the main language used with your child during the day?

	English		ASL		Polish		Ukrainian		
	French		Signed English		Portuguese		Vietnamese		
	Arabic		Hungarian		Punjabi				
	Chinese		Italian		Spanish				
	Cree		Korean		Tagalong (Filipino)				
	German		Persian (Farsi)		Greek				
	Other (please specify) _____								

How would you identify your ethnic origins? (please mark all that apply)

Mom	Dad		Mom	Dad	
		Canadian (English / French)			Chinese
		French			Polish
		British			Portuguese
		German			Black
		Scottish			Filipino
		Irish			Latin American
		Italian			Japanese
		Ukrainian			Korean
		Dutch (Netherlands)			Aboriginal (North American Indian, Méti Inuit, Eskimo)
		South Asian (East Indian, Pakistani, Punjabi, Sri Lankan)			Arab / West Asian (Armenian, Egyptian, Iranian, Lebanese, Moroccan)
		South East Asian (Cambodian, Indonesian, Laotian, Vietnamese)			Other (please specify) _____

How many years of elementary and high school have you completed?

	Mom	Dad
no schooling		
less than 6 years		
7 years		
8 years		
9 years		
10 years		
11 years		
12 years		
13 years		

Have you ever attended any other kind of school such as a university, community college, business school, trade or vocational school, CEGEP or other post-secondary institution?

	Mom		Dad	
Yes		If yes, for how many years? _____		If yes, for how many years? _____
No				

What certificates, diplomas or degrees have you obtained? (Please check all that apply)

Mom	Dad	
		None
		Secondary (high) school graduation certificate or equivalent
		Diploma or certificate from at a trade, technical or vocational school
		Diploma or certificate from a business school
		Diploma or certificate from a community college, a nursing school or CEGEP
		Bachelor's or undergraduate degree(s) (e.g., B.A., B.Sc., LL.B.)
		Master's degree(s) (e.g. M.A., M.Sc., M.Ed.)
		Degree in medicine, dentistry, veterinary medicine or optometry
		Doctorate (e.g., Ph.D., D.Sc., D.Ed.)
		Other _____

Are you currently working at a job or business? (Please include part-time jobs, seasonal work, contract work, self-employment, baby-sitting or any other paid work)

Mother	☐	Yes	☐	No	☐	At home with children	☐	Currently attending school	☐	Permanently unable to work
Father	☐	Yes	☐	No	☐	At home with children	☐	Currently attending school	☐	Permanently unable to work

What kind of work are you doing (for example, janitor, medical lab technician, accounting clerk, manager of engineering department, supervisor of data entry unit, secondary school teacher, fishing guide, etc)?

Mother _____

Father _____

What is your combined family income from all sources before taxes (for example, wages and salaries, income from self-employment, employment insurance, pensions, child support, alimony etc.)?

	Below $20,000
	$20,000 to less than $30,000
	$30,000 to less that $40,000
	$40,000 to less than $50,000
	$50,000 to less than $60,000
	$60,000 to less that $70,000
	$70,000 to less than $80,000
	more than $80,000

THANK YOU VERY MUCH FOR YOUR HELP

Reference: The impact of screening and case finding on the functional status of children with a hearing impairment

Appendix 7

Individual Semi-Structured Interview Guide

Individual Semi-Structured Interview Guide

Study ID:_____

Start with introduction and general conversation to establish rapport
Provide information on purpose of interview. I am meeting with parents to better understand the impact of early diagnosis of hearing loss through infant hearing screening on the family. I will be talking with several parents of young children with hearing loss about their experiences with the diagnosis of the hearing impairment.
I would like to hear about how you learned of your child's hearing loss and how you think it has made a difference for you and your child. I would also like to hear about your needs once you learned that your child has a hearing loss and about what kind of services you feel were/are the most appropriate in guiding you in developing your child's communication.
Explain procedure. I will ask you some questions to guide our conversation but feel free to talk to me about your experiences and to add any information you feel is important. Please don't hesitate to ask for clarification if any questions are unclear.

1. Tell me how you found out about your child's hearing loss.
   Probe: How has learning of the hearing loss through screening (or regular referral routes) been beneficial or negative for you and your child?
   How do you think it might be different if your child's hearing had not been screened (or had been screened)?

2. What impact do you think screening (or not) will have on you and your child?
   Probe: How are things better/worse for your child because he/she was diagnosed early?
   How are things better/worse for you and your family because of the early diagnosis?

3. I am interested in understanding your needs when your child was first diagnosed and later after the diagnosis?
   Probe: What kind of information from service providers did you find helpful in the beginning?
   What information or guidance did you need in the months following the diagnosis (for example, after the hearing aid fitting)?
   What kind of supports did you need, e.g. social worker, therapist, family?
   What else was/is needed for you to help your child develop?

4. If you could create a perfect health system for you and your child, what would it look like, that is, what types of services should it provide?
   Probe: What kinds of professionals, type of setting, and frequency of visits would be helpful?
   Tell me about the guidance you would like in helping your child develop communication skills
   What are we (clinics) doing well and what do we need to do better?
   What do you perceive as the gaps in service?

General Information:

Location of interview: Home ☐ Other
Informant: Mother ☐ Father ☐ Other ☐
Screening status: Screened ☐ UNHS ☐ Targeted ☐ Not screened ☐
Screening category: Well-baby ☐ NICU ☐ At-risk ☐ No-risk ☐
Age of child_____ Age at diagnosis _____ Age at intervention_____
Degree of hearing loss: Mild ☐ Moderate ☐ Severe ☐ Profound ☐
Hearing Technology Hearing aids ☐ Date of fitting _____ Cochlear Implant ☐ Date of Surgery_____
Diagnostic center_____ Intervention center(s) _____
Type of intervention _____ Frequency _____
Other disabilities _____ Interviewer Comments (Use reverse side)

Appendix 8

Conjoint Analysis Questionnaire

Description of Questionnaire and Instructions:

This questionnaire contains descriptions of service models for families of young children with hearing loss. The questionnaire was designed based on information collected during interviews with parents in 2005 to 2006 about their service needs.

The questionnaire consists of 9 hypothetical scenarios in which you are asked to choose between two clinics that have different characteristics and therefore provide a somewhat different service model. The questionnaire should take about **20** minutes to complete. For each choice, please check **one box only** - Clinic A **or** Clinic B, as your preferred clinic. A definition of each clinic characteristic is provided. Please refer to these when answering the questions.

Consent
Please sign and return the **consent form** with the questionnaire. If you are completing the questionnaire electronically, please send the consent form by paper mail. Please note that since all interviews in this study have now been completed, you are only consenting to participation in the questionnaire component of the study. (*Note: Parents who participated in an interview with Elizabeth have already signed the consent form and do not need to complete another form*).

****

Thank you for completing the questionnaire and participating in this study. If you have any questions, do not hesitate to contact the study investigator:

Elizabeth Fitzpatrick, Email: efitzpatrick@cheo.on.ca, Tel: 613-738-3908.
Children's Hospital of Eastern Ontario Research Institute, 401 Smyth Road, Ottawa, ON K1H 8L1.

*Please check **one** box only for each question.*

Question 1

	Clinic A	**Clinic B**
Location of therapy	Home or community	Home or community
Frequency of therapy	1 time/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Coordinated through same agency as therapy
Access to parent support	Independently accessed by parent (e.g. parent groups)	Organized as part of agency/health services
Access to information in early stages	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)	Not easily available through clinic

Which Clinic would you prefer? A ☐ B ☐

Question 2

	Clinic A	**Clinic B**
Location of therapy	Home or community	Clinic
Frequency of therapy	2 to 3 times/week	2 to 3 times/week
Coordinated services (e.g. therapy, audiology, psycho-social	Services available - not coordinated through one agency	Coordinated through same agency as therapy
Access to parent support	Organized as part of agency/health services	Organized as part of agency/health services
Access to information in early stages	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Question 3

	Clinic A	**Clinic B**
Location of therapy	Alternate between clinic and home or community	Alternate between clinic and home or community
Frequency of therapy	1 time/week	2 to 3 times/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Services available - not coordinated through one agency
Access to parent support	Organized as part of agency/health services	Independently accessed by parent (e.g. parent groups)
Access to information in early stages	Not easily available through clinic	Not easily available through clinic

Which clinic would you prefer? A ☐ B ☐

Question 4

	Clinic A	**Clinic B**
Location of therapy	Clinic	Alternate between clinic and home or community
Frequency of therapy	2 to 3 times/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Services available - not coordinated through one agency
Access to parent support	Independently accessed by parent (e.g. parent groups)	Independently accessed by parent (e.g. parent groups)
Access to information in early stages	Not easily available through clinic	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Question 5

	Clinic A	**Clinic B**
Location of therapy	Alternate between clinic and home or community	Clinic
Frequency of therapy	1 time/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	Services available - not coordinated through one agency	Services available - not coordinated through one agency
Access to parent support	Organized as part of agency/health services	Independently accessed by parent (e.g. parent groups)
Access to information in early stages	Not easily available through clinic	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Question 6

	Clinic A	**Clinic B**
Location of therapy	Clinic	Clinic
Frequency of therapy	1 time/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Coordinated through same agency as therapy
Access to parent support	Organized as part of agency/health services	Organized as part of agency/health services
Access to information in early stages	Not easily available through clinic	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Question 7

	Clinic A	**Clinic B**
Location of therapy	Home or community	Alternate between clinic and home or community
Frequency of therapy	2 to 3 times/week	2 to 3 times/week
Coordinated services (e.g. therapy, audiology, psycho-social	Services available - not coordinated through one agency	Coordinated through same agency as therapy
Access to parent support	Independently accessed by parent (e.g. parent groups)	Independently accessed by parent (e.g. parent groups)
Access to information in early stages	Not easily available through clinic	Not easily available through clinic

Which Clinic would you prefer? A ☐ B ☐

Question 8

	Clinic A	**Clinic B**
Location of therapy	Clinic	Alternate between clinic and home or community
Frequency of therapy	1 time/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	Services available - not coordinated through one agency	Services available - not coordinated through one agency
Access to parent support	Organized as part of agency/health services	Organized as part of agency/health services
Access to information in early stages	Not easily available through clinic	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Question 9

	Clinic A	**Clinic B**
Location of therapy	Alternate between clinic and home or community	Alternate between clinic and home or community
Frequency of therapy	2 to 3 times/week	1 time/week
Coordinated services (e.g. therapy, audiology, psycho-social	No psycho-social services available	Coordinated through same agency as therapy
Access to parent support	Organized as part of agency/health services	Independently accessed by parent (e.g. parent groups)
Access to information in early stages	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)	Provided through clinic audiologists/therapists (e.g. verbal, literature, videos)

Which Clinic would you prefer? A ☐ B ☐

Definition of terms in questionnaire

Location of therapy ***Refers to the location in which regular therapy services are provided.***	
Clinic	Refers to a hospital, school or other setting where you take your child for regular therapy sessions (typically, this is not in your neighborhood).
Home/Community	Refers to therapy services provided at your home or in a community location near your home, (e.g. within a few kilometers).
Alternate between clinic and home or community	Refers to a therapy program provided in more than one location, for example, one week, you take your child to a hospital clinic and the next week, a therapist comes to your home or a nearby community setting.

Frequency of therapy ***Refers to the amount of therapy you and your child receive. It is assumed that therapy sessions are provided weekly and last approximately one hour.***	
1 time/week	You have access to therapy through the health care system 1 time per week.
2 to 3 times/week	You have access to therapy 2 to 3 times per week through the health care system.

Coordinated services (e.g. therapy, audiology, psycho-social (e.g. social work, psychology) ***Refers to the notion that typical services for children with hearing loss and their families are coordinated through a single agency or clinic such that there is good communication with the parents and between the various professionals involved.***	
Coordinated through same agency as therapy	All services are coordinated through the same agency or clinic that is responsible for providing the child's therapy services. There is good communication and seamless transition between services.
Services available – not coordinated through one agency	Refers to a situation where the services required are available but are coordinated or managed through different agencies. Ex. therapy is provided in one setting while audiology and/or psycho-social services (e.g. social work, psychology) are provided by another clinic or agency.
No psycho-social services available	Refers to a situation where audiology and therapy are available but psycho-social services (social work and/or psychology) are not available as part of the services offered for children with hearing impairment.

Access to parent support ***Refers to contact with parents of children with hearing loss who can provide emotional support and/or information and resources. Parents may include those who are currently experiencing the process of helping a child with a hearing loss (i.e. children are of preschool age) or those who have older children and have already experienced many of the aspects of raising a child with a hearing loss.***	
Independently accessed by parent	Refers to a situation where you seek out parent support yourself by attending organized parent meetings or informally meeting other parents. In this situation, the clinic may provide you with the information that parents' groups exist but you take the initiative for connecting with other parents.
Organized as part of agency/health services	Refers to a situation where the clinic that is providing your child's therapy arranges, in a systematic way, parent support as part of the services it offers you. For example, the clinic may host organized meetings or may offer you the opportunity to meet with parents by arranging a meeting, having a parent contact you, etc.

Access to information in early stages ***Refers to the ease with which you can access information about technology, medical aspects of hearing loss, and rehabilitation and possible long term results for your child.***	
Provided through clinic audiologists / therapists	Refers to the situation where the clinic provides significant amount of information in various formats, e.g. verbal, written literature, internet sites, videos, etc.) on an ongoing basis. You feel that you are well informed about the child's loss, kept up to date about technologies and aware of your child's progress and likely outcomes.
Not easily available through clinic	Refers to the situation where you perceive that information is not readily or easily available through the clinic where you receive your therapy and/or audiology services and that you have to seek out information from other sources in order to feel adequately informed.