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Predictors of Language Outcome for Children in the Ontario Infant Hearing Program

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Abstract

The Ontario Infant Hearing Program (OIHP) provides early interventions (i.e., hearing aids) to children who are hard of hearing (CHH) because research consistently demonstrates their benefit to language outcomes. The impact of pre-fitting language abilities on these outcomes are not well understood.

This retrospective cohort analysis examined the performance of OIHP children on the Preschool Language Scale-4 at the time of (n=47), and after (n=19), initial hearing aid intervention. Regression analyses revealed that, before amplification, hearing loss severity predicted language abilities. However, after amplification, severity of hearing loss did not uniquely predict language achievement, but rather was driven by its relationship with language at the time of amplification.

These findings suggest that hearing aids fitted early may provide a preservation benefit to the language achievement of CHH, and that this benefit is greatest for children at highest risk (i.e., children with the weakest initial language, and most severe hearing loss).

Keywords

Hearing loss; Hard of Hearing; Hearing Aids; Children; Language; Outcome; Development

List of Abbreviations

BE-4PTA: Better-ear Four Pure Tone Average

CAA: Canadian Academy of Audiology

CHH: Children who are Hard of Hearing

CI: Cochlear Implant

CIHTF: Canadian Infant Hearing Taskforce

EHDI: Early Hearing Detection and Intervention

JCIH: Joint Committee of Infant Hearing

OIHP: Ontario Infant Hearing Program

PCHL: Permanent Childhood Hearing Loss

PLS-4: Preschool Language Scale 4^{th} Edition

rSII: Residual Speech Intelligibility Index

SAC: Speech Audiology Canada

SII: Speech Intelligibility Index

Co-Authorship Statement

This work consists of three chapters: an introductory chapter (Chapter 1), an integrated manuscript (Chapter 2), and a concluding chapter (Chapter 3). I, Olivia Daub, am the sole author of Chapters 1 and 3 and the lead author of Chapter 2. I am responsible for the conceptualization of the project, data analysis, and interpretation of results with support from my co-supervisors, Dr. Janis Cardy and Dr. Marlene Bagatto. Chapter 2 is a manuscript that has been submitted for publication as a Research Note. Co-authors Dr. Bagatto and Dr. Cardy supported the data extraction and clinical interpretation of results as well as reviewed and provided feedback on the final manuscript described in Chapter 2. Co-author Dr. Andrew Johnson provided statistical support, and guided analysis selection and consideration of the variables included for the analysis. As well, Dr. Johnson reviewed the final manuscript, contributing important comments.

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Table of Contents

List of Tables

List of Figures

Chapter 1

1 Introduction

Permanent Childhood Hearing Loss (PCHL) is implicated in poor speech, language, academic, and psycho-social outcomes (JCIH, 2013; Moeller, 2000; Nelson, Bougatsos, & Nygren, 2008; Patel & Feldman, 2011; Speech-Language and Audiology Canada [SAC], 2010). Interventions, such as hearing aids, aim to provide improved access to auditory information early in development in order to support the communication development of children who are hard of hearing (CHH). Concerns regarding a lack of evidence to motivate the provision of early interventions for CHH and to guide their implementation have led to an increased research focus on exploring eventual outcome after intervention, and intervention's role in producing these outcomes. As a result, the period of development prior to hearing aid intervention has been understudied. How infants learn from disrupted auditory information before they receive hearing aids, and the role played by periods of language development prior to intervention in eventual outcomes are not well understood.

This thesis is centered around a study that examined language outcomes of CHH, with special consideration of their pre-hearing aid fitting communication abilities, to further understand the impact of hearing aid use in this population (see Chapter 2). To provide background orientation to the study, the present chapter explores the implementation of Early Hearing Detection and Intervention (EHDI) programs internationally, with a specific focus on Canadian EHDI status. Following a brief overview of language acquisition research that motivated Universal Newborn Hearing Screening (UNHS) implementation, the evidence for EHDI program benefit is reviewed.

1.1 Auditory Experience and Language Development in Infancy

First language acquisition is an enormously complex task wherein infants are required to learn the phonology, prosody, lexicon, morphology, syntax, and pragmatics of their native language while also learning about basic properties of sound (i.e., which sounds constitute human speech versus environmental sounds and which variations within human speech constitute meaningful differences). Early in development, typically-developing infants possess cognitive skills that enable them to use the auditory information contained within their environment to extract language-specific rules and information. Work investigating the cognitive processing of language acquisition in infants has identified two main themes in regards to the role of auditory exposure: 1) experience is cumulative, and 2) speech input matters.

In the last trimester of pregnancy, the fetus has access to the basic prosody of the language to which she is exposed in the womb. This early exposure facilitates vowel perception within two days of birth (Moon, Lagercrantz & Kuhl, 2012). This time period also provides an anchor for segmenting phonetic properties, or words, of the infant's native language, a process often referred to as *prosodic bootstrapping* (Thiessen & Saffran, 2007; Nazzi & Ramus, 2003; Johnson & Jusczyk, 2001). Phonetic discrimination, in turn, is predictive of later language abilities (Jansson-Verkasalo et al., 2010; Tsao, Liu & Kuhl, 2004) and is additionally driven by the infant's native language. The classic theory of perceptual tuning for speech indicates that young infants are capable of discriminating speech contrasts that are present across languages (Werker & Tees, 1984). With experience, perception of nonnative contrasts diminishes and perception of native contrasts strengthens (Kuhl et al., 2008; Kuhl, Williams, Lacerda, Stevens, & Lindblod, 1992, Werker & Tees, 1984). Recent research has generated more nuanced theories of perceptual tuning such that maintenance of nonnative speech contrasts is not purely input driven but may be facilitated by social interaction (Kuhl, 2007). However, the basic premise holds: infants are able to use the speech information contained in their environment to learn their native language.

The importance of early speech information in providing a foundation for later language acquisition is highlighted in circumstances in which an infant's access to auditory information is compromised. Of particular interest to the present work is PCHL. PCHL is estimated to affect between 1 to 3 infants per 1000 live births (Mehl & Thomson, 1997) and affects both the quantity and quality of auditory information infants are able to access. Limited access to auditory information has serious implications for language learning, particularly early in infancy. As described above, infants' language learning, which is shaped by their auditory environment, follows a developmental sequence that sets a foundation for later learning. Access to poor quality or quantity speech input may interrupt early learning, or perhaps be insufficient for learning to occur, in infants with PCHL and may consequently influence later outcomes (Moeller & Tomblin, 2015). Recent work by Kuhl et al. (2008) explored the relation of phonetic discrimination and later language skills, and revealed that infants who were better at discriminating native phonetic contrasts at 7.5 months produced more words at 24 months than those who were worse at discriminating the same contrasts. Discrimination ability of nonnative contrasts produced the opposite results: 7.5 month olds who were better at discriminating nonnative contrasts produced fewer words at 24 months than infants who were worse at discriminating nonnative contrasts. The authors posited that native contrast discrimination represents *neural commitment* to a native language that facilitates later language learning whereas nonnative contrast discrimination represents neural openness and, critically, that early learning may impact the ability to learn in the future. Infants with PCHL may, therefore, experience difficulty in learning the necessary phonetic information in a timely manner resulting in impairments to their later language learning abilities. Indeed, it is well documented that some infants with PCHL experience varying degrees of difficulty with language in childhood (JCIH, 2013; Moeller, 2000; Nelson et al., 2008; Patel & Feldman, 2011; SAC, 2010). Hearing aid intervention is one factor that aims to increase an infant's access to better quality auditory input in order to facilitate this critical early learning. Put simply, hearing aids amplify the acoustic signal to reach levels sufficient to stimulate the infant's cochlea, and for the infant to detect speech sounds easily and comfortably (Seewald, Moodie, Scollie, & Bagatto, 2005). While a hearing aid does not restore normal hearing, it *does* provide higher quality access to speech information from which the infant may learn, compared to no hearing aid.

It is additionally important to consider that PCHL is *not* a language disorder per se: infants with PCHL who are exposed to fluent sign language input undergo early speech perception processes, such as perceptual tuning to signed phonemes (Palmer, Fais, Golinkoff, & Werker, 2012), and develop their language systems normally. Language impairments in infants with PCHL are secondary to their sensory impairments in situations where the infant is exposed to environments in which spoken languages are the primary mode of language input. Although estimates have been difficult to obtain, it is commonly accepted that less than 10% of infants with PCHL are born to parents where one, or both, are hard of hearing, and even fewer (<5%) to parents who are Deaf (Mitchell & Karchmer, 2004). It is therefore the case that an overwhelming majority of infants with hearing loss are born into families where oral language is used (either in isolation, or as a compliment to sign/signed language) and must learn it from disrupted auditory input (Crowe, Fordham, McLeod, & Ching, 2014; Crowe, McLeod, McKinnon, & Ching, 2014; Li, Bain, & Steinberg, 2003).

1.2 Hearing Aids Increase Access to Auditory Information

Hearing aid fitting is not an all-or-nothing event, but rather a process. Hearing aid prescription is based on audiological assessments that identify the minimum volume (i.e., threshold) across relevant speech frequencies where the individual child detects the sound. Ear- and frequency-specific hearing thresholds and a measurement of the acoustics of the child's ear canal serve as the basis for the child's hearing aid prescription. The prescriptive targets provide specific levels where the hearing aid should amplify speech so that the infant can hear it easily, and provide an upper limit so that loud sounds coming into the hearing aids do not cause uncomfortable listening. In this regard, hearing aid quality refers to an *individual* hearing aid's ability to amplify frequencies to meet an *individual* child's prescriptive targets. The process of assessing the child's hearing, developing the prescriptive targets, and then verifying that the hearing aid meets the recommended levels is conducted by a pediatric audiologist who executes an evidence-based hearing aid fitting protocol (Bagatto, Scollie, Hyde, & Seewald, 2010; Bagatto et al., 2016). By completing this process accurately, the affected infant or child can access the greatest amount of speech spectrum possible for his unique level of hearing loss when wearing the hearing aids. The amount of speech to which an individual child has access can be operationalized as the proportion of the speech spectrum provided by the hearing aid, known as the *speech intelligibility index* (SII). This metric is available in clinical hearing aid fitting software and is considered in concert with a child's hearing thresholds within EHDI programs in order to evaluate the quality of the hearing aid fittings. SII has been used in many recent EHDI outcomes studies, such as the Outcomes for Children with Hearing Loss study (OCHL; Ambrose, Walker, Unflat-Berry, Oleson, & Moeller, 2015; Tomblin, Oleson, Ambrose, Walker, & Moeller, 2014; Tomblin et al., 2015).

Although the SII facilitates inferences about the quality of a hearing aid fitting, it is not an informative value in isolation. SII values must be interpreted in relation to the individual child's unique hearing levels. Due to technological limitations, hearing aids are not able to grant perfect audibility to every child. Data from Bagatto and colleagues (2011) indicate that as severity of hearing loss increases, the average proportion of the speech spectrum accessible with high quality hearing aids drops from 100% (mild hearing loss) to 40% (severe hearing loss) due to SII's relation with an individual child's hearing levels. The amount of *aided* SII that a child achieves is constrained by their *unaided* SII. Therefore, a child with a moderate-severe hearing loss may only have an aided SII of 0.4 (access to 40% of the speech spectrum) while wearing a high quality hearing aid, whereas a child with a mild hearing loss may have an aided SII of 0.9 (access to 90% of the speech spectrum). In each case, the hearing aid might be well fitted. Therefore, evaluating the quality of the hearing aid fitting on the basis of SII is best considered in relation with a child's individual hearing levels. Comparing an individual child's aided SII and severity of loss using Bagatto and colleagues' (2011) norms is an easily interpreted method of determining whether a child's aided SII values are appropriate. Other work has developed a method for capturing hearing aid quality in a single value called *residual* SII (rSII; first introduced in Tomblin et al., 2014; see Tomblin et al., 2015 for additional applications). The rSII value is a standardized residual of aided SII whereby variance in aided SII due to unaided SII is removed using a piece-wise regression with 2 linear functions (one for unaided SII values above 0.16 and one for SIIs below 0.16; Tomblin et al., 2014). rSII values, therefore, capture hearing aid quality in a way that statistically controls for the relationship between aided and unaided SII and provides a value for the unique SII provided by the hearing aid. The rSII is particularly useful in its ability to be used as a predictor in data analyses (see 1.3 for examples). The normative values used by Bagatto and colleagues (2011) are used in the present work to evaluate the quality of hearing aids worn by children in our sample to define our population although the rSII is an important consideration for future iterations of this work.

1.3 Supporting Infants with Hearing Loss: EHDI Programming

Given the importance of early auditory information in language acquisition, the lasting effects of early auditory deprivation, and the availability of technologies capable of increasing access to auditory information, EHDI programs have been developed in order to support the early detection, identification, and intervention of infants with PCHL. In advocating for EHDI provision, taskforces have emerged in Canada (Canadian Infant Hearing Task Force, CIHTF) and the United States (Joint Committee in Infant Hearing, JCIH) that “strongly support the establishment and maintenance of an integrated, consistent and culturally-sensitive UNHS program” (SAC, 2010) in all regions of their respective countries.

Starting in 1994, the JCIH developed comprehensive guidelines for appropriate EHDI implementation. While the JCIH has refined and updated these guidelines to reflect current best practices (JCIH 2000, 2007, 2013), they have provided a series of benchmarks for EHDI programs to meet in order to maintain quality. The goal of UNHS, the first step in an EHDI program, is to screen 95% of all infants for hearing loss born under the program’s purview within one month of birth. If the infant fails screening, confirmation of hearing loss should be completed by 3 months of age, and if permanent hearing loss exists, intervention should begin by 6 months of age (JCIH, 2007). Interventions may include speech-language therapy, personal hearing aids when appropriate, connection to fluent American Sign Language resources, and additional family supports based on an individual family’s preferences and the child’s needs. The benchmarks developed by the JCIH have impacted EHDI programming beyond U.S. borders and have been included in Canadian recommendations (SAC, 2010) and Ontario protocols (Bagatto et al., 2010).

Gaining government support and resources for the development of quality EHDI programming has been a considerable challenge, particularly in establishing the importance of UNHS for infants who are born without risk factors for hearing loss. Previous models of hearing screening required that it only be completed for children born with risk factors such as a family history of permanent hearing loss, syndromes, or additional medical conditions thought to compromise hearing ability, or consequences of prematurity (e.g., anoxia; Nelson et al., 2008). By contrast, infants without risk factors for childhood hearing loss, who comprise approximately 50% of the PCHL population (Nelson et al., 2008), received audiological testing as concerns emerged under a wait-and-see approach. This resulted in the average age of hearing loss identification occurring at approximately 2½ years (Mehl & Thomson, 1998), after substantial fundamental language learning has taken place in children without hearing loss (Jansson-Verkasalo et al., 2010; Kuhl et al., 2008; Tsao et al., 2004). Proponents of UNHS argued that PCHL is far more common than other congenital disorders for which infants are screened at birth, and causes considerable difficulty in speech, language, academic, and psychosocial outcomes (JCIH, 2013; Moeller, 2000; Nelson et al., 2008; Patel & Feldman, 2011; SAC, 2010). For instance, phenylketonuria affects 7 infants per 100,000 live births and costs $40,500 USD per confirmed diagnosis whereas PCHL affects 260 infants per 100,000 live births and costs between $9,600 to $12,300 USD per confirmed diagnosis (Mehl & Thomson, 1998)**.** For these reasons, PCHL has been described as a "neurological emergency" (SAC, 2010, p.2), which precisely highlights the importance of the timing and severity of consequences of the disorder. The theoretical demonstrations of the importance of early auditory information, consequences associated with PCHL, and prevalence of the disorder provided the foundation for the provision of newborn hearing screening for *all* infants, not just those considered to be at high risk.

Bess and Paradise (1994) notably criticized early support for UNHS based on a lack of evidence that UNHS without intervention follow-up benefits infants with PCHL, and on a lack of evidence to guide follow-up intervention selection and protocol. As such, government policies, funding, and development of UNHS programs without access to audiological assessment and intervention services were presently unjustified. The validity of Bess and Paradise's initial criticisms of UNHS recommendations are reflected in the variability of research attempting to identify benefits associated *only* with early identification of hearing loss. Simply *identifying* a hearing loss is not sufficient in isolation to benefit infants and children with hearing loss. Early identification is beneficial in its relationship with early intervention such that if a child is identified early, she can receive appropriate intervention early. To this end, early research examining only the role of age of identification has produced inconsistent results: some work demonstrates improved outcomes for early identification (Wake et al., 2016; Yoshinaga-Itano, 2003a; Yoshinaga-Itano, Coulter, & Thomson, 2001) while other studies failed to find differences (Wake et al., 2016; Korver et al., 2010; Fitzpatrick, Durieux-Smith, Eriks-Brophy, Olds, & Gaines, 2007). Inconsistencies in operationalizing early versus late identification have further muddied the literature. Benefits of early identification have been documented when *early* is operationalized as before 6 months (Ching et al., 2010; Yoshinaga-Itano, 2003a; Thompson et al., 2001) or before 3 months (Vohr et al., 2008; Yoshinaga-Itano, 2003b).

Given that the benefits of early identification depend on its relation with early and suitable intervention, and given both the criticisms of implementing UNHS in the absence of follow-up and the related difficulty in demonstrating benefits of UNHS in isolation, recent research has turned away from exploring UNHS as a single predictor. Instead, as outlined by JCIH benchmarks, UNHS is considered as an important component of an EHDI program and the most recent work has focused on identifying specific factors that predict improved outcomes for infants and children with hearing loss within EHDI programming.

The OCHL study is currently the most extensive exploration of additional factors predicting various outcomes following EHDI intervention. The OCHL study used an accelerated longitudinal design to track over 300 CHH after they were fitted with hearing aids and explored their speech and language outcomes using composites of a number of speech and language measures, as well as language sampling, to understand their speech and language development. Notable factors predicting speech and language performance include the quality of hearing fittings, hours of daily hearing aid use, and features of caregiver’s speech to the child (Ambrose et al., 2015; Tomblin et al., 2015). The quality of the hearing aid fitting, as indexed by rSII in particular, impacted the eventual speech and language outcomes of CHH as well as their developmental trajectories. In addition to rSII, speech and language outcome and development was predicted in the OCHL study by the child's unique amount of hearing aid experience (in both hours per day and months of use; Tomblin et al., 2015).

The shift in research attention away from the effects of age of identification and towards exploring EHDI programs and individual differences has provided important insights into EHDI best practices and components of a child's intervention that may maximize speech and language outcomes. As research continues to move in this direction, it is important to continue to consider the spirit of what early studies on the effects of age of identification and amplification aimed to assess: how do periods of unamplified development impact eventual outcome? Despite goals to fit infants with hearing aids as early as possible in development (with targets set to be 6 months; Bagatto et al., 2010, 2016; JCIH, 2007; SAC, 2010), infants still experience periods of development where language learning takes place without amplification (see section 1.1 for a discussion), and then must reconcile the amplified input with their developing linguistic system. Furthermore, hearing aids do not restore normal hearing. As described above, even with access to high quality hearing aids, CHH may not have access to the full speech spectrum depending on their unique level of hearing loss. The task of reconciling improved, but still not normal, auditory information with a linguistic system that has been developing under degraded auditory input is considerable and is currently not well understood.

Understanding how infants with PCHL acquire language prior to amplification, how they utilize amplified input, and how their language abilities prior to amplification relate to eventual language outcomes, are foundational in understanding the benefits provided by hearing aids specifically, and EHDI programs broadly. Any intervention must operate on the child's pre-existing development, and it is therefore crucial to understand how pre-intervention abilities respond to intervention. Studying pre-amplification language abilities clarifies not only the nature of benefit provided by amplification, but also furthers an understanding of the sorts of effects that hearing loss is having on infants' language abilities very early in development – perhaps providing evidence to suggest even earlier interventions, such as parent training, to maximize the eventual benefit received from amplification.

1.4 EHDI: A Canadian Perspective

Canadian healthcare services are federally governed by the 1984 *Canada Health Act,* which dictates principles of healthcare provision that provinces and territories must meet in order to continue receiving federal funds (Health Canada, 2012). Specific models of healthcare and service provision are otherwise determined by provincial and territorial governments and are becoming increasingly regionalized (Donald & Philippon, 2008). Whereas regions of a province or territory make decisions with regard to service provision, it is the province/territory's responsibility to ensure that these decisions abide by the *Canada Health Act*.

The variability of policies both within and across provinces and territories is reflected in the provision of EHDI programs at a national level. In 2014, SAC partnered with the Canadian Academy of Audiology (CAA) to form the Canadian Infant Hearing Task Force (CIHTF) to assess the state of Canadian EHDI services with the position that "all children in Canada deserve access to proper hearing screening and timely intervention to reach their full potential" (CIHTF, 2014, p.2). The CIHTF assessed programs that were administered at a provincial or territorial level rather than regional or hospital based programs. The CIHTF found that only five provinces (British Columbia, Ontario, Nova Scotia, Prince Edward Island, and New Brunswick) had province-wide programs and that these were of varying quality in terms of percentages of infants screened, program standards, and outcome tracking. The remaining eight provinces and territories did not have a province-wide EHDI program, with screening ranging from 10% (Manitoba) to 90% (Yukon and Newfoundland & Labrador) of infants (see CIHTF, 2014 for a summary of CIHTF findings).

Two years later, response to the 2014 assessment has been positive. Program development has begun in provinces where there were previously no services (Alberta, Quebec, Manitoba) and improvement in diagnostic and management protocols has occurred in others (Ontario, Nova Scotia, PEI). The adoption of OIHP protocols has also occurred in the Northwest Territories and Yukon. Despite these advancements, provincial support is absent or declining in some regions (Saskatchewan, New Brunswick) and Canadian services, overall, are still inadequate (CIHTF, 2016). The CIHTF advocates for an increase in federal, provincial, and territorial commitment to EHDI excellence in order to overcome current difficulties and, in doing so, has written to the provincial/territorial Health Ministers regarding their assessments of each province/territory, as well as to the federal Health Minister.

1.5 EHDI: Ontario as a Canadian Leader

The provinces of Ontario and British Columbia have well-developed EHDI programs. As the CIHTF 2016 progress report outlined, OIHP protocols are being adopted by the Northwest Territories and Yukon. Examining the success of the BC and Ontario EHDI programs in improving various outcomes for children born with hearing loss has the capacity to inform the refinement of protocols as well as provide evidence of the benefits of EHDI programs within a Canadian context to bolster support for the provision of EHDI services elsewhere in Canada and worldwide.

The OIHP was implemented in 2002 to provide EHDI services to infants with PCHL and CHH in the province of Ontario. The OIHP provides family centered support including audiological services and connections to other service providers until the child is 6 years old and evidence-based protocols have been developed in order to "facilitate the affected child's development of communication skills and readiness for school" (Bagatto et al., 2010, p. S71). Within the OIHP framework, parents are partners who are included in all levels of decision making, including preferred mode of communication (oral or manual), and intervention options (referral to speech-language pathologists and provision of hearing assistance technologies). One component of OIHP service is the provision of high quality amplification to "improve functional auditory capacity and participation in hearing- and communication-specific situations" (Bagatto et al., 2010). The OIHP amplification protocol supports the JCIH recommendations to provide hearing aids by 6 months of age (Bagatto et al., 2010, 2016). Additionally, the OIHP monitors their ability to adhere to EHDI program benchmarks (e.g., screening by 1 month, identification by 3 months, intervention by 6 months), as well as facilitate improved child outcomes. These assessments are facilitated by a clinical database wherein de-identified patient information is stored.

The auditory development of infants and children within the OIHP who wear hearing aids is monitored by clinicians using the UWO PedAMP, an evidence-based protocol using functional outcome measures such as the LittlEARS Auditory Questionnaire (Tsiakpini et al., 2004) and Parents' Evaluation of Aural/Oral Performance of Children (PEACH) rating scale (Ching & Hill, 2005a; Bagatto et al., 2011, 2016). Bagatto and colleagues (2011) evaluated the performance of 352 children from the Southwest and Toronto regions of the OIHP on the measures included in the UWO PedAMP. 223 children in their sample had normal hearing and 129 had permanent hearing loss. Results indicated that typically developing children with PCHL in the OIHP were meeting auditory development milestones on the LittlEARS, while their peers with comorbidities demonstrated initial typical auditory development, but delays beginning to emerge after their first birthday. Similarly, typically developing children performed within normal limits on the PEACH questionnaire, although their performance was significantly predicted by degree of hearing loss (Bagatto et al., 2016). These analyses demonstrate the utility of evidence based outcome measurement protocols in EHDI services. The analyses by Bagatto et al. demonstrate not only the appropriateness of outcomes achieved by typically developing children enrolled in the OIHP, but also provided performance ranges against which to compare future cohorts of OIHP children. Despite the quality and utility of the UWO PedAMP for evaluating audiological outcomes, the monitoring of speech, language and communication outcomes have not been well explored within the OIHP.

The language outcome assessment tool used by OIHP communication development service providers since 2009 is the *Preschool Language Scale, 4th edition* (PLS-4; Zimmerman et al., 2002). It has not yet been examined at the program level. Although the OIHP is currently in the process of refining their language assessment protocols, examining previously collected data could provide a foundation upon which to base future tool selection. The PLS-4 is a clinician administered, norm referenced omnibus language measure containing both an Auditory Comprehension and Expressive Communication subtest and has been used in the evaluation of language abilities of CHH

(Ching et al., 2010, 2013; Fitzpatrick et al., 2007; Hogan, Stokes & Weller, 2010; Jackson & Schatschneider, 2014; Moeller, 2000; Stika et al., 2014; Wake et al., 2016). Although the PLS-4 has failed to detect subtle differences amongst different interventions (Wake et al., 2016; Fitzpatrick et al., 2007; Ching et al., 2013) or between CHH who were younger than 1½ years old and typically hearing children (Stika et al., 2014), some work has reported that 3-year-old CHH perform at, or below, one standard deviation below the mean on this measure (Ching et al., 2010). As an omnibus measure, the PLS-4 is particularly useful in capturing a child's overall language ability. Specific areas of deficit, for instance limited vocabularies or grammatical impairments are not, however, isolated by the PLS-4.

1.6 The Present Work

Through the project described in Chapter 2, we aimed to address theoretical gaps in EHDI program research, as well as evaluate the language outcomes of CHH enrolled in the OIHP who wear hearing aids. The cognitive processing that infants with PCHL use in acquiring their language is not well understood. Speech perception and the cognitive mechanisms involved in language learning in hearing loss populations have largely been studied in children who receive cochlear implants and not those who use hearing aids. Furthermore, studies of language outcomes in children who use hearing aids have not considered the role of pre-amplification language development in eventual language outcome. It is also of interest to evaluate the language outcomes of children within the OIHP to document the program's success in intervention, inform the development of a language assessment protocol, as well as highlight the utility of a well-developed language assessment protocol in contributing to not only program evaluation, but also theory generation.

Chapter 2 of this thesis is a manuscript that has been submitted as a Research Note for peer review. The paper aimed to address the following three questions:

1) To what extent does hearing loss impact language performance prior to amplification? We predicted that increased levels of hearing loss would be associated with decreased levels of language ability prior to amplification.

2) Can hearing aids ameliorate these effects? We predicted that the receipt of hearing aids would improve language performance.
3) What factors predict the magnitude of change in language performance after amplification? We predicted that pre-amplification language ability and severity of hearing loss would impact the magnitude of growth that occurred after hearing aid fitting.

Chapter 2

2 Language Outcomes in Children who are Hard of Hearing: The Role of Language Ability Before Hearing Aid Intervention

2.1 Background

It is well documented that early auditory experience dramatically impacts and shapes the language development of typically hearing infants and children (Kuhl et al., 2008; Maye, Weiss, & Aslin, 2008; Maye, Werker, & Gerken, 2002; Werker & Tees, 1984). In cases where access to auditory information is compromised by permanent childhood hearing loss, the importance of early access to auditory information has motivated the development and provision of Early Hearing Detection and Intervention (EHDI) programs. Initial recommendations for Universal Newborn Hearing Screening were met with criticism based on both the absence of comprehensive detection and intervention programs to support children who are hard of hearing (CHH) and a lack of evidence supporting benefits of such programs (Bess & Paradise, 1994). These criticisms and concerns influenced research evaluating the effectiveness of comprehensive EHDI programs. When considered as an overall predictor, the benefits of early hearing detection and intervention have been demonstrated (Kennedy et al., 2006; Moeller, 2000; Vohr et al., 2008; Wake et al., 2016; Yoshinaga-Itano et al., 2001; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; see Yoshinaga-Itano, 2003a for a review) and some work has revealed age-appropriate language achievement on certain measures of language ability in CHH who received early intervention **(**Fulcher, Purcell, Baker & Munro, 2012; Moeller, 2000; Stika et al., 2014**)**.

Recent work has moved beyond considering early hearing detection and intervention as a single factor to begin studying how specific aspects of early detection and intervention relate to language outcomes. Outcomes for Childhood Hearing Loss (OCHL), a well-controlled accelerated longitudinal study, recently explored various factors that predict outcomes in a sample of 317 CHH who were fitted with hearing aids. The OCHL project explored language achievement using composite measures of language ability from 2 to 6 years of age and identified that while earlier hearing aid fitting was associated with improved language outcomes, it was not the only factor that predicted performance. Quantity (amount of caregiver speech), quality (directing versus eliciting utterances), amplification dosage (the residual difference between unamplified and amplified hearing levels, or rSII), amount of hearing aid use (both hours per day, and months with the hearing aid) and severity of hearing loss all contributed both to language performance, as well as growth over time (Ambrose et al., 2014; Tomblin et al., 2015). Previous work has also explored the contributions of some of these factors to the outcomes of CHH who are fitted with hearing aids (Ambrose et al., 2014; Ching & Dillon, 2013; Ching et al., 2010; Walker et al., 2015), but have not considered all of them in such a large single sample.

This recent research underscores the role that additional factors may play in a child's language development and overall outcome. However, it has yet to consider the child's communicative development *prior to* hearing aid fitting as a predictive factor, despite recognition of the importance of early communicative experiences to eventual language outcome and its use as a rationale for the provision of early auditory access through EHDI programs. It is currently unknown how CHH are able to perceive or process suboptimal speech signals that have not been amplified with hearing aids and use it to develop oral language. In regions with EHDI programs, the goal is for CHH, whose families elect amplification, to be fitted with hearing aids promptly following confirmation of permanent hearing loss and parent readiness to proceed with fitting (Bagatto et al., 2010; JCIH, 2007 within 1 month of diagnosis; SAC, 2010). Although CHH are identified and provided with hearing aids as early as possible, they are still faced with forming the perceptual and cognitive basis for their linguistic system for a number of months prior to personal amplification. The degree of progress in communication development made during unamplified periods of development may impact the degree of benefit hearing aids are able to provide. Evaluating the role of language and communication ability prior to receipt of hearing aids in eventual language outcome is crucial for understanding how CHH are able to use the amplified input provided by the hearing aids and for developing theories regarding the mechanism by which intervention with hearing aids impacts the language outcomes of CHH.

Purpose

The present work addresses the role that the time prior to intervention with hearing aids plays in the language outcomes of CHH by examining their language performance at the time of hearing aid fitting and its relationship with language performance after exposure to amplification. Three main questions were addressed: 1) To what extent does hearing loss impact language performance prior to hearing aid intervention? 2) Can hearing aids ameliorate these effects? If so, 3) What factors predict the magnitude of change in language performance after exposure to hearing aids?

2.2 Methods

This retrospective cohort analysis examined CHH serviced by the Ontario Infant Hearing Program (OIHP) who were born in 2008 and 2011. Following recommendations set out by the Joint Committee on Infant Hearing and Speech Audiology Canada (Bagatto et al., 2010; JCIH, 2007; SAC, 2010), newborns in Ontario are universally screened for hearing loss and provided with appropriate family-centered follow-up services to confirm the presence of permanent hearing loss and provide intervention (e.g., hearing aids, communication development) according to the family's choices. The screening, audiological assessment, hearing aid fitting and audiological outcome measurement components of the OIHP are based on evidence-based protocols that are implemented province-wide. Due to the nature of oral communication development services, specific protocols for intervention do not currently exist in Ontario, however, children identified with permanent hearing loss through the OIHP are routinely assessed using the *Preschool Language Scale, 4th edition* (PLS-4; Zimmerman, Steiner & Pond, 2002) to track their progress. Those data are entered into a provincial database.

Data collection

Data were extracted from the OIHP clinical management database for the 2008 and 2011 birth cohorts. OIHP protocol mandates the collection of basic demographic data (sex, birth date, gestational age), audiological information (hearing thresholds, hearing aid fitting date, audibility provided by the hearing aid), outcome assessment information (test results and scores), as well as information regarding appointment dates and complicating factors, for entry into a de-identified database. All information included in the database was obtained over the course of the child's care by their clinical service providers (e.g., Audiologist, Speech-Language Pathologist) who follow the relevant provincial protocols. Given the clinical nature of the services provided, the features of the communication development intervention received by children in our sample were variable and reflected the decision making of the OIHP clinicians.

Participants

Children within the database from the 2008 and 2011 birth cohorts had been identified as having permanent childhood hearing loss in at least one ear and had been fitted with hearing aids at some point during their care within the OIHP. Both children who were typically developing and children who had other comorbidities were included in the sample. Data from children were included in the current analyses if they had a PLS-4 language assessment conducted before or within two months of their first hearing aid fitting. These broad inclusion criteria resulted in an extremely variable sample (see Table 1 for details). The mean age at which children were fitted with hearing aids in the sample was 21.92 months (*SD* =16.54), with an average hearing loss severity of 47.46 dB HL better-ear four pure tone average (BE-4PTA; described in further detail below). It is important to note that the mean age of hearing aid fitting in the sample is older than JCIH benchmarks for children identified in the first few months of life (i.e., fitted by 6 months). This is representative of our inclusion of children with comorbidities who, for a variety of reasons, may have received their hearing aids later, not of a failure in implementation of the OIHP. Comorbidity was dichotomized as present or absent in our study (described below), but the comorbidities flag is not restricted to medical, neurological, or developmental diagnoses. In the OIHP database, a comorbidity may be entered for the presence of medical issues (e.g., cerebral palsy or Down syndrome) or a complex factor (e.g., family or psychosocial challenges, inconsistent hearing aid use, Children's Aid involvement). Similarly, our sample included children with unilateral hearing loss (represented by a BE-4PTA of less than 25 dB HL) who receive amplification as well as children with profound bilateral hearing loss (BE-4PTA greater

Table 1: Summary of Participant Characteristics

	Group 1 (*N* = 47)			Group 2 (*N* = 19)		
Variable	*n*	*M*	*SD*	*n*	*M*	*SD*
Comorbidity						
Present	11			4		
Absent	36			15		
BE4PTA (dB)		47.46	22.93		50.79	21.31
Age at Fit (months)		21.92	16.54		14.81	10.47
PLS-4 percentile rank Pre-Amplification						
Auditory Comprehension Scale		35.85[a]	32.37		34.47[b]	31.47
Expressive Communication Scale		40.42	28.71		41.74	28.79
PLS-4 percentile rank Post-Amplification						
Auditory Comprehension Scale					39.11[b]	34.89
Expressive Communication Scale					44.84	35.01

Note. BE4PTA= Better Ear 4-Pure Tone Average. PLS-4 = Preschool Language Scale, 4th edition.

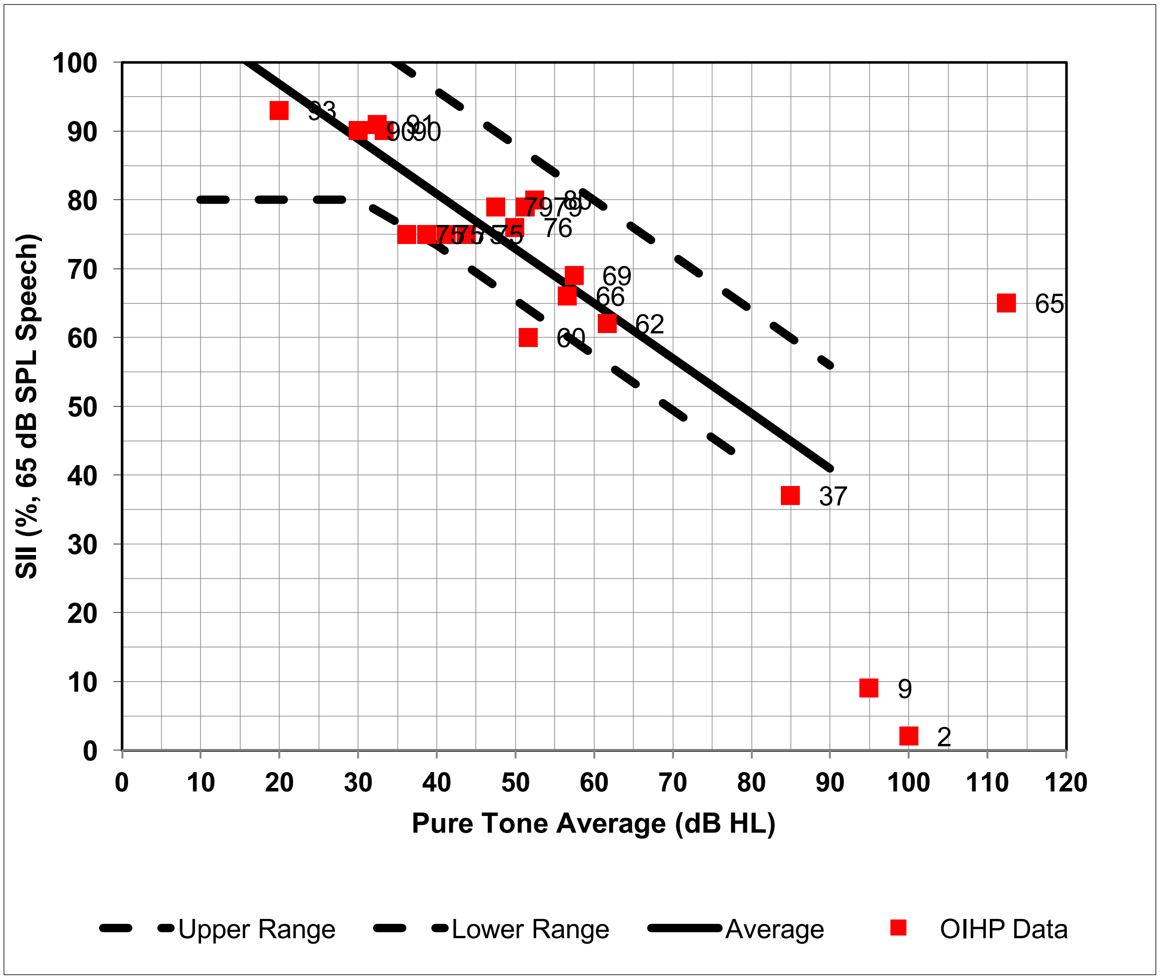
[a] *N* = 46

[b] *N* = 18

than 90 dB HL). Two children were excluded from our analyses because of discrepancies between the raw scores and the percentile ranks reported in the database that appeared to be due to data entry error and could not be resolved. One child was included but was missing scores for the PLS-4 Auditory Comprehension scale. The first author adjusted the percentile ranks of seven children when there was evidence that they had been calculated incorrectly. Types of errors that were adjusted included: using the Auditory Comprehension charts to calculate Expressive Communication percentiles, rounding a child's age to the next age band, calculating a child's percentile using corrected age rather than chronological age, and calculating a percentile using the wrong raw score. These adjustments were made to maintain consistency between children, and were made according to the PLS-4 examiner's manual.

Forty-seven children were included in our final analyses of language achievement at the time of hearing aid fitting. A subset of these (N = 19) had data available for a second analysis of language achievement both at the time of hearing aid fitting and at some time (greater than two months) after the initial hearing aid fitting. In accordance with the OIHP Protocol for the Provision of Amplification procedures (Bagatto et al., 2010), infants and children are considered candidates for amplification if the hearing loss is permanent and hearing thresholds for either ear are 30 dB HL or greater at any frequency between 500 and 4000 Hz. Amplification is provided based on ear-specific threshold estimates at 500 and 2000 Hz using the Desired Sensation Level Method (Scollie et al., 2005) and real-ear-to-coupler difference measurements (Bagatto et al., 2005, 2010). Given limitations in sample size, we were unable to statistically consider the unique effects of quality of the hearing aid fitting in the analysis. However, using normative values developed by Bagatto et al. (2016) to evaluate the audibility of speech provided by the hearing aids (e.g., speech intelligibility index, or SII) demonstrated that the audibility provided by hearing aids to the children in our sample fell within expected ranges in all but five cases (Figure 1). Four children had SIIs that fell below, and one child had values that fell above, the expected norms (see Fig. 1 for details). SIIs were calculated using an Average Input (65 dB SPL) level.

Figure 1: Hearing Aid Speech Intelligibility Index (SII) Values (N=19)



Outcome measures

Children's language achievement was evaluated using the PLS-4, an omnibus language measure containing scales for Auditory Comprehension and Expressive Communication that is suitable for children ranging from birth to 6 years, 11 months. Our analyses considered both percentile ranks and Progress Values (Zimmerman, Steiner & Pond, 2006) as outcome measures. Percentile ranks represent the percentage of children in the PLS-4 norming sample who performed below the individual child, thus providing an estimate of the child's ability relative to same-age peers. Progress Values, unlike percentile ranks, do not consider a child's achievement in relation to the PLS-4 norming sample and provide an index of progress on the test specific to the individual child. Raw scores provide similarly specific indices of performance, however they are problematic in that the difference between raw score points on the PLS-4 are not equal intervals whereas Progress Values are. For the purposes of our analyses, we chose to analyze both percentile ranks and Progress Values, which allowed us to consider their performance and subsequent growth relative to both the normative sample as well as themselves.

Language ability at the time of amplification was operationalized using either the percentile ranks or Progress Values from a PLS-4 assessment either before or within two months of a child's first hearing aid fitting. Similarly, language ability after amplification was considered to be either the percentile rank or Progress Values from a PLS-4 assessment conducted sometime greater than two months after a child was first fitted with hearing aids. It is important to note that the periods between first and second assessments were not the same for all children in our sample. Time between assessment periods was statistically controlled by creating standardized residuals for the change scores, which was used as the outcome measure in all analyses of language growth.

Data Extraction

We selected age, severity of hearing loss and presence/absence of comorbidity as relevant predictors for language achievement at the time of hearing aid fitting. Since the PLS-4 percentile ranks are calculated using age, only severity of hearing loss and presence/absence of comorbidity were included as predictors of language achievement at the time of amplification for analysis of PLS-4 percentiles.

Severity of hearing loss was operationalized as the BE-4PTA calculated during the audiological assessment closest in date (but not following) the child's first language assessment. The BE-4PTA is the average of a child's dB HL thresholds across 500, 1000, 2000 and 4000 Hz. Pure tone averages are calculated for each ear individually and the BE-4PTA is the pure tone average of the ear with the lowest dB HL threshold, that is, the better hearing ear or the ear that has the least amount of hearing loss.

Comorbidity was dichotomized due to the limitations of a regression analysis in using multiple categorical variables. Specifying whether or not a child has a comorbidity did not become a required field within the OIHP database until 2010. Therefore, for some time period of both the 2008 and 2011 birth cohorts examined, comorbidities may not have been identified. Therefore, it was not necessarily the case that the absence of a comorbidity entry was due to the absence of comorbidity. For the purpose of these analyses, *absent* comorbidities were operationalized as a comorbidity field that was either left blank, or specified "none."

2.3 Results

Language Ability at Time of Hearing Aid Fitting

Hierarchical linear regression analyses were conducted on the Progress Values for the Auditory Comprehension and Expressive Communication scales, wherein predictors were entered into the analysis in the following order: Age, BE-4PTA, the interaction between Age and BE-4PTA, and Comorbidity. R2change was evaluated for each model, and the most parsimonious model was considered to be the last model to produce a significant improvement in explained variance. Details of these regression models are presented in Table 2.

The most parsimonious model for the prediction of Auditory Comprehension using Progress Values, was the model that included Age, BE-4PTA, and the interaction between Age and BE-4PTA, R2(adj) = 0.84, $F(3, 43) = 83.96$, $p < 0.05$. Within this model, both age and BE-4PTA were statistically significant predictors, but the interaction between these predictors did not significantly contribute to prediction.

Similarly, the most parsimonious model for the prediction of Expressive Communication using Progress Values, was the model that included Age, BE-4PTA, and the interaction between Age and BE-4PTA, R2(adj) = 0.85, $F(3, 43) = 88.79$, $p < 0.05$. In this model, however, only age was found to be a statistically significant predictor of Expressive Communication.

A second set of hierarchical linear regression analyses was conducted for the percentile ranks, wherein BE-4PTA and Comorbidity were entered in successive steps. BE-4PTA was a significant predictor for both Auditory Comprehension, R2(adj) = 0.19,

Table 2: Hierarchical Regression Model of Language Progress Value Prior to Hearing Aid Fitting

	PLS-4 Progress Value Pre-Amplification					
	Auditory Comprehension			Expressive Communication		
Predictor	R^2 (adj)	ΔR^2	b	R^2 (adj)	ΔR^2	b
Model 1	0.56***			0.70***		
Age			5.82***			4.33***
Model 2	0.84*	0.27***		0.85***	0.15**	
Age			6.81**			5.62**
BE4PTA			-1.66*			-0.66
Age* BE4PTA			-0.26			-0.03
Model 3	0.87	0.03		0.85***	0.00	
Age			6.68*			5.48**
BE-4PTA			-1.66*			-0.66
Comorbidity			12.97			14.52
Age* BE4PTA			-0.02			-0.03

Note. PLS-4 = Preschool Language Scale, 4th edition. Auditory Comprehension Scale, $N = 46$. Expressive Communication Scale, $N = 47$. BE4PTA= Better Ear 4-Pure Tone Average.

*$p < .05$. **$p < .01$. ***$p < .001$.

$F(1,45)=11.7$, $p<0.05$, and Expressive Communication scales, R2(adj) = 0.10, $F(1,45)=5.8$, $p<0.05$. Comorbidity was not a significant predictor for either of these dependent variables. Details of these regression models are presented in Table 3.

Change in Language Ability

There was no significant change in Auditory Comprehension and Expressive Communication percentile ranking after hearing aid fitting, $t(17)=0.5$, $p>0.05$ and $t(18)=-0.5$, $p>0.05$, although Progress Values did show significant growth, $t(17)=6.46$, $p<0.05$ and $t(18)=8.23$, $p<0.05$. Hierarchical linear regression analyses were conducted on the residualized Progress Value difference scores, using predictors entered in the following order: Progress Values from the first assessment, BE-4PTA, and the interaction between Progress Values from the first assessment and BE-4PTA. As was the case in evaluating initial language ability, the most parsimonious model was considered to be the last model that produced significant R2change. The most parsimonious model for residualized Auditory Comprehension Progress Value difference scores was the model that included first assessment Progress Values, BE-4PTA and the interaction between them. In this model, BE-4PTA and the interaction between Progress Value at first assessment and BE-4PTA were significant predictors, $F(3,14)=16.42$, $p<0.05$, explaining 70% of the variance. Our analyses did not produce a significant model of residualized Expressive Communication Progress Value change, $F(1,17)=2.201$, $p>0.05$ (see Table 4 for a summary).

Language Ability after Amplification

Final hierarchical regression analyses were conducted on Progress Value performance and percentile ranks after hearing aid fitting using BE-4PTA and Progress Value performance, or percentile rank, from first assessments as the second variable entered. Unlike previous analyses, the most parsimonious model was considered to be the model that significantly accounted for the most variance. We were not interested in R2change for these regressions, as we were interested in evaluating how the relation between BE-4PTA and language ability after hearing aid fitting changes when we account for the

Table 3: Hierarchical Regression Model of Language Percentile Rank Prior to Hearing Aid Fitting

	PLS-4 percentile rank Pre-Amplification					
	Auditory Comprehension			Expressive Communication		
Predictor	R^2 (adj)	ΔR^2	b	R^2 (adj)	ΔR^2	b
Model 1	0.19*			0.10		
BE4PTA			-0.64**			-0.42*
Model 2	0.17***	-0.02		0.08	-0.02	
BE4PTA			-0.64**			-0.42*
Comorbidity			2.37			2.03

Note. PLS-4 = Preschool Language Scale, 4th edition. Auditory Comprehension Scale, $N = 46$. Expressive Communication Scale, $N = 47$. BE4PTA= Better Ear 4-Pure Tone Average.

*$p < .05$. **$p < .01$. ***$p < .001$.

Table 4: Hierarchical Regression Model of Change in Language After Hearing Aid Fitting

	PLS-4 Progress Value Difference Score					
	Auditory Comprehension			Expressive Communication		
Predictor	R^2 (adj)	ΔR^2	b	R^2 (adj)	ΔR^2	b
Model 1	0.57***			0.06		
PLS-4 Progress Value Pre-Amplification			-0.29***			-0.13
Model 2	0.73***	0.16*				
PLS-4 Progress Value Pre-Amplification			0.14			
BE4PTA			2.56**			
Progress Value*BE4PTA			-0.01*			

Note. PLS-4 = Preschool Language Scale, 4th edition. Auditory Comprehension Scale, N = 19. Expressive Communication Scale, N = 20. BE4PTA= Better Ear 4-Pure Tone Average. Outcome scores were standardized residuals of PLS-4 Progress Values removing the variance due to time between the pre-amplification and post-amplification assessments.

*p < .05. **p < .01. ***p < .001.

variance contributed by initial language ability. In all cases, the most parsimonious model was the model that included both BE-4PTA and Progress Value (Auditory Comprehension: $F(1,16)=4.347$, $p>0.05$, Expressive Communication: $F(1,17)=3.326$, $p>0.05$) or percentile rank (Auditory Comprehension: $F(1,17)=3.326$, $p>0.05$, Expressive Communication: $F(1,17)=3.326$, $p>0.05$) as predictors. Unlike the regression models evaluating language ability prior to amplification, BE-4PTA was not a significant predictor for Auditory Comprehension Progress Value, Expressive Communication Progress Value, Auditory Comprehension percentile rank or Expressive Communication percentile rank in these models, whereas Progress Values and percentile ranks from initial language assessments were a significant predictor for all models except the model of Auditory Comprehension Progress Value post hearing aid fitting. Details of these regression models are included in Tables 5 and 6.

2.4 Discussion

Our results indicate that the severity of hearing loss impacted language achievement prior to hearing aid fitting and that this had lasting effects on language outcomes after hearing aid fitting in our sample. Although children continued to acquire language skills after receiving amplification (as indicated by significant Progress Value change for both language scales), they maintained the same standing relative to same-age peers that they had before receiving hearing aids. The amount of Progress Value growth on the Auditory Comprehension scale was significantly predicted by an interaction of severity of hearing loss and Progress Values at the time of amplification, such that children with greater severities of loss experienced the greatest amount of Progress Value growth, but high levels of initial auditory comprehension abilities attenuated this growth. The presence or absence of comorbidity never entered any of the models. This suggests that the greatest benefits of hearing aids was delivered to the children who were at greatest initial risk: those with more severe hearing losses and the worst initial language comprehension ability.

Table 5: Hierarchical Regression Model of Language Progress Value After Hearing Aid Fitting

	PLS-4 Progress Value Post-Amplification					
	Auditory Comprehension			Expressive Communication		
Predictor	R^2 (adj)	ΔR^2	b	R^2 (adj)	ΔR^2	b
Model 1	0.16			0.11		
BE4PTA			-1.29			-1
Model 2	0.91***	0.75***		0.84***	0.73***	
BE4PTA			0.46			0.12
PLS-4 Progress Value Pre-Amplification			0.78***			0.93***

Note. PLS-4 = Preschool Language Scale, 4th edition. Auditory Comprehension Scale, N = 19. Expressive Communication Scale, N = 20. BE4PTA= Better Ear 4-Pure Tone Average.

*p < .05. **p < .01. ***p < .001.

Table 6: Hierarchical Regression Model of Language percentile rank After Hearing Aid Fitting

	PLS-4 percentile ranks Post-Amplification					
	Auditory Comprehension			Expressive Communication		
Predictor	R^2 (adj)	ΔR^2	b	R^2 (adj)	ΔR^2	b
Model 1	0.38			0.15		
BE4PTA			-0.99**			-0.63
Model 2	0. 50**	0.12		0.35*	0.20*	
BE4PTA			-0.57			-0.19
PLS-4 percentile rank Pre-Amplification			0.47·			0.63*

Note. PLS-4 = Preschool Language Scale, 4th edition. Auditory Comprehension Scale, N = 17. Expressive Communication Scale, N = 18. BE4PTA= Better Ear 4-Pure Tone Average.

*p < .05. **p < .01. ***p < .001.

In examining the results from Progress Values and percentile ranks, we propose that hearing aids benefit language outcome not by accelerating language achievement. Rather, based on these data, it appears that hearing aids may *preserve* language growth trajectories and protect them from declines associated with continued untreated hearing loss. The growth in Progress Value, therefore, is interpreted as a consequence of this preservation effect in that children with more severe hearing losses are protected by their hearing aids and the most severely-impaired children were able to acquire the skills necessary to maintain their standing relative to their peers with normal hearing.

Our preservation hypothesis is further supported by our finding that severity of hearing loss did not uniquely predict language achievement after amplification. In our sample, the relationship of hearing loss severity to language achievement after amplification was driven by its relationship with language at the time of amplification, rather than further effects. Tomblin et al. (2015) posited that either a period of rapid catch-up or protection from effects of severity might explain stable language performance across ages in children receiving intervention before 6 months of age. However, their work examined data collected after hearing aid fitting, thus they were unable to measure language ability prior to fitting. Our data supports the hypothesis that stable language performance for early fitted children is due to protection, rather than catch-up.

In proposing a preservation hypothesis, we emphasize the importance of the period of development prior to a child's hearing aid amplification in their eventual language outcomes. Studies that examine the language outcomes of children involved in EHDI programs have focused on providing evidence for the benefit of early amplification supported by EHDI programs as well as identifying factors that may improve language outcomes beyond amplification (Ambrose, VanDam, & Moeller 2014; Ching et al., 2010; Tomblin et al., 2015; Tomblin et al., 2014). However, these studies have not considered how language ability prior to amplification may impact eventual language outcomes. Our results suggest that language ability prior to hearing aid fitting is another factor that predicts eventual language outcomes. With the increasing prevalence of EHDI programs, research examining language outcomes in CHH should increase attention to the role of unamplified development.

Exploring the cognitive processes of CHH before amplification will inform our understanding of *how* CHH are able to learn spoken language using hearing aids as well as improve early identification and remediation of language deficits. A full understanding of hearing aid benefit depends upon understanding how CHH acquire language without intervention with hearing aids and how that changes with the introduction of amplification at various ages. It is currently unknown what cognitive strategies, if any, CHH use to compensate for their sensory deficits prior to being fitted with hearing aids. The ability to adopt compensatory strategies may differentially predict better, or worse, language outcomes for CHH and enable early identification of persistent language delays. Similarly, understanding the way in which CHH use auditory information prior to amplification may expose malleable factors early in development for these children that can maximize hearing aid benefit. If, as demonstrated here, hearing aids preserve pre-amplification language ability, then maximizing pre-amplification language ability may optimize outcomes. Research into the cognitive processes of CHH would provide stakeholders (speech-language pathologists, audiologists, educators, and caregivers) a starting point from which to begin intervening to maximize pre-amplification ability.

Our study also demonstrates the utility of using Progress or Growth Scale Values. Progress Values provided an index of language ability that allowed us to examine language growth without the limitations associated with raw scores, standard scores, or percentile rankings. Studying language change using raw scores is problematic in that the difference in ability required to score additional points on the PLS-4 (or any developmental measure) is not spaced at equal intervals. Furthermore, using raw scores to quantify language growth necessitates the use of difference scores, which compounds on measurement error associated with assessments at both time points. Progress Values, on the other hand, provide an equal interval unit of language growth that considers the standard error of measurement in its calculations, allowing identification of significant Progress Value change on a case-by-case basis. Concerns with standard scores and percentile ranking are immediately evident from our analysis: children in our study did not demonstrate significant percentile rank change. This lack of change is not evidence that growth did not occur; it is only evidence that the growth was not sufficient to alter children's standing relative to the norming sample (rather than relative to their own performance), that is, they demonstrated a typical rate of growth in their language skills between assessments. Progress Values are sufficiently sensitive to capture a child's change in her own performance, which, when considered in addition to percentile ranking, is especially informative.

Despite the benefits of using Progress Values, their use in CHH language outcome studies has not been adopted; standard scores are currently used for reporting results on standardized language assessments (Tomblin et al., 2014, 2015). This is, perhaps, due to the difficulty associated with calculating Progress or Growth Scale Values. The charts required to calculate the Progress Values for the PLS-4 were not included in the PLS-4 materials, but rather were later sold separately. However, due to difficulty in sales, the charts were never reprinted and are no longer available. Fortunately, Progress Values, renamed as Growth Scale Values are now being included in publications of the PLS-5 (Zimmerman, Steiner, & Pond, 2011). With their increasing availability, we encourage their adoption in the study of language growth and outcomes based on the aforementioned benefits.

As a retrospective study, our data included a number of limitations. First, known predictors of language (e.g., socio-economic status, characteristics of caregiver input, multilingual language environment) were not included in the database and thus were not available for consideration. Similarly, details about each child's communication development intervention (e.g., communication mode, type of intervention, frequency) were unknown and may have impacted our findings. Although the decision making for communication modality is multifactorial, surveys of communication modality choices suggest that between 87% and 96% of parents choose speech for either the sole communication modality or as a compliment to signed input (e.g., sign language or signed English; Crowe, Fordham, McLeod, & Ching 2014; Crowe, McLeod, McKinnon, & Ching, 2014; Li, Bain, & Steinberg, 2003). Thus, it is unlikely that the children in our sample were not receiving at least some degree of consistent speech input.

Additionally, we cannot fully account for differences between children in our sample who had PLS-4 assessments before hearing aid fitting and other OIHP children who did not. Of the 155 children in the OIHP database who had PLS-4 assessment data entered, only 48 of them had a PLS-4 assessment prior to their hearing aid fitting. Upon confirmation of hearing loss, OIHP protocol aims to begin intervention as soon as possible. It is possible that in some cases children were connected with communication development intervention, and thus had their language assessed before hearing aid fitting was complete, while other children did not have an opportunity to have a language assessment prior to the hearing aid fitting. Likewise, comorbidity was a poor predictor in our analyses perhaps due to our limitations in sample size or the variability in comorbidities due to the inclusion of both medical and complex factors. While we may expect that a medical diagnosis such as Down Syndrome would influence language development, it is less clear how a complex factor, such as Children's Aid involvement, may manifest in PLS-4 performance. This reinforces the importance of prospectively testing our preservation hypothesis on a larger sample of CHH in order to fully account for sample characteristics. We also had an insufficient sample size to consider the relationship of our predictors across different levels of hearing loss severity, as well as to include indicators of hearing aid quality (e.g., residual SII) and amount of hearing aid use per day, which have been demonstrated to impact language outcomes and trajectories after amplification (Tomblin et al., 2015). Given the limitations of our study, we are unable to draw definitive conclusions. However, our analyses have allowed us to generate hypotheses that warrant further research attention in a well-controlled prospective design and in larger samples.

2.5 Conclusions

Our retrospective cohort analysis represents a first attempt at studying the language outcomes of CHH in relation to their language ability prior to amplification. Despite some limitations, our data suggest that hearing aids preserve the language ability of CHH and enable them to follow a rate of language development typical of same-age peers without hearing loss. Further prospective research designs are needed to evaluate this preservation hypothesis.

Chapter 3

3 Discussion

The project described in Chapter 2 was a retrospective cohort analysis of the language abilities of CHH enrolled in the OIHP and was conceptualized to fulfill two goals: program evaluation and theory generation. This chapter describes in greater detail the implications of this project for the consideration of the OIHP specifically and Canadian EHDI program development broadly, as well as the implications of our analyses for clinical and theoretical understandings of hearing aid benefit.

3.1 Program Evaluation: EHDI Data Management and Language Outcomes in a Canadian Context

As Canadian provinces and territories move towards adopting provincial/territorial EHDI programs, Ontario is being increasingly presented with opportunities to serve as a leader. Evaluating the language outcomes of CHH first allows us to identify components of OIHP outcome assessment protocols that need to be addressed, and second, provides a demonstration of expected levels of achievement that can serve as a basis for comparison for other provinces and territories as they evaluate their developing programs. Finally, this project illustrates the utility of strong data maintenance and outcome assessment protocols and encourages their consideration in the development of future EHDI programs.

3.1.1 Outcome Assessment Protocols: Data Management Considerations

Conducting analyses of growth and change necessitated careful attention to the data included in the database and, through our data extraction process, highlighted data management practices that were problematic for research purposes. It is important to note that the OIHP uses a *clinical* data management program, and program evaluation is secondary to this purpose. In this regard, we make no conclusions about the ability of the current data management protocol to fulfill its clinical purpose. However, as this project demonstrates, evaluating program wide data offers a benefit to program provision. Future analyses will be limited by current data management protocols and, with program evaluation goals in mind, it is important to consider how effective current protocols are in facilitating data analysis.

We had access to both the 2008 and 2011 birth cohorts that were extracted from the database in 2012. 201 children were included in the 2008 database and 135 were included in the 2011 database. Of these children, 105 (born in 2008) and 50 (born in 2011) had a PLS-4 assessment conducted at any age. Only 48 children from both cohorts had their language assessed prior to their hearing aid fitting, and only 20 of these children had an additional assessment at some time greater than two months after their hearing aid fitting. While less than half of the children in the database were available to us for analysis, thus limiting the generalizability of our conclusions, it is promising to note that the children born in 2011 were being assessed at younger ages, suggesting that improvements are being made in language outcome assessment.

Additionally, there were discrepancies between regions, or clinicians, in regards to the scoring of the PLS-4. During the course of this project, the lead author identified inconsistencies between percentile calculation practices and instructions provided in the PLS-4 examiner manual. These discrepancies most often involved rounding a child's age to the nearest age band and using a corrected age adjustment for premature children, neither of which follows the standard scoring procedures required for this measure. Comparing children to either older, or younger, age bands can seriously impact the interpretability of results. A child compared to older children may appear to be more severely impaired than she truly is, whereas a child compared to younger children may appear to be less impaired. Ensuring standardized scoring procedures across regions and clinicians will be necessary for further program evaluations, but also in improving clinical decision making in regards to speech and language impairments.

Accounting for individual differences in PLS-4 performance was a challenge for this study. Information that is necessary to appropriately consider language achievement is not included in the OIHP database. For instance, information regarding language environment (monolingual or multilingual) and language modality (spoken, signed, or both) are not included in this database. Specifics regarding comorbidities or developmental delays are not fully accounted for in this database. Mandatory documentation of comorbidities was introduced in 2010, therefore the presence/absence of these was not necessarily fully documented for the 2008 cohort. Access to this information will facilitate interpretation of observed PLS-4 scores and degrees of individual variation, particularly in regards to understanding predictors of a child's performance relative to test norms. All of our regression models predicted more than 73% of the variance in either Progress Value performance at first or second assessment, as well as Progress Value change, despite having no more than two predictors in any model. Our model of Auditory Comprehension percentile rank performance, although significant, only accounted for 17% of the variance, leaving substantial variance unexplained. Coupled with the fact that CHH maintained their percentile rank between assessment periods, it is necessary to include other predictors in our analysis to more fully account for differences in ability in these children.

It is important to note that our access to OIHP databases was restricted to 2008 and 2011 cohorts. It is, therefore, impossible to identify if the issues noted here have been addressed as protocols have been adjusted, although there is some evidence to suggest improvement in practices (see above). Additionally, the OIHP is in the process of revising their communication assessment protocol. During this process, special attention paid to the issues documented here will facilitate later program evaluation. Furthermore, access to more recent cohorts would enable the identification of persistent data management issues, as well as underscore progress that has been made with protocol refinement.

3.1.2 Outcome Assessment Protocols: Measurement Considerations

Both the tools used to assess progress, as well as the scores used to report this progress heavily influence interpretation of speech and language achievement in CHH. The PLS-4 is currently the only language assessment measure used in the OIHP. As an omnibus tool, the PLS-4 may be useful in capturing a broad snapshot of a CHH's language ability but it is not overly sensitive to areas of specific concern for this clinical population. For instance, CHH have performed within normal limits on vocabulary assessments, but these same children have struggled on measures of verbal reasoning (Moeller, 2000).

Unlike the OCHL study, there are currently no measures included in the OIHP that are sensitive to specific speech and language impairments typical of CHH. For instance, outcome studies of CHH have demonstrated differences in performance across different measures. Moeller (2000) found that vocabulary was resilient to differences in predictors of outcome (age of intervention and levels of family involvement), but verbal reasoning skills were more vulnerable. Similarly, the OCHL study used composite measures of language ability and found subtle differences in language outcome, particularly in regards to both morphological accuracy as well as the morphological complexity of CHH's speech samples (Ambrose et al., 2015; Tomblin et al., 2015). Expansion of the OIHP language outcome assessment protocol to include additional measures would enable refined measures of program quality. Tracking known areas of difficulty for CHH would provide information on the OIHP's ability to benefit particularly problematic areas of development as well as provide information to tailor interventions on an individual basis.

How to best quantify change in performance on the PLS-4 was an area of particular concern for this study. While OIHP protocol necessitates that language should be assessed every six months, the time between assessments was not equal between children in our sample (perhaps due to differences in protocol uptake across OIHP regions) and we needed to statistically control for these differences by creating standardized residuals of the change scores. Residualizing our change scores involved removing the variance in the change scores due to time between assessments. Reducing the variability of our change scores resulted in less variance with which to run our future analyses. Methodological control, namely assessing language at equal time points, would have been preferable to statistical adjustments. While relying on residualized change scores enabled us to interpret our data in a meaningful way, it reduced the power of our subsequent analyses, which may have impacted our results, especially given our small sample size.

Selection of PLS-4 scores for the analyses also required careful consideration: raw score, standard score, and percentile rank are all available for the PLS-4. Percentile ranking, rather than standard score, was the norm-referenced score entered into the OIHP clinical database, and allowed for comparison of our sample to the PLS-4 norm sample. However, using percentile difference scores only provides information about a child's change *relative* to the norming sample, and is not sensitive to change in ability relative to the individual, nor is it reflective of the acquisition of additional skills. Since our research questions focused on changes in language abilities before and after hearing aid fitting, using percentiles was informative, but not sufficient for our analyses. Raw scores, the other score entered into the clinical database, are the most direct measure of a child's acquisition of skills, that is, every one-point increase corresponds to another correctly scored item on the test. However, using raw scores presented the opposite concern to that presented by using percentile ranks: the distance, or difficulty in acquiring skills measured by individual items is not equal-interval and therefore interpreting differences in change scores between individuals is not possible.

Progress Values (re-named Growth Scale Values in the PLS-5; Zimmerman et al., 2006, 2011) are scores that address the limitations of both percentile ranks and raw scores. Progress Values provide a standardized, equal-interval measurement of an individual child's change in performance on the PLS-4. A common qualm with the use of difference scores is that difference scores contain measurement error associated with both test administrations. Progress Value change analysis is facilitated through calculations of standard error of measurement associated with both assessments. Progress Value conversion tables, produced specifically for the PLS-4, provide the minimum Progress Value difference considered significant, unique to first and final Progress Value. That is, the amount of change in Progress Value that is significant is not linear across all initial Progress Values. Assessing *significant* individual improvement on the PLS-4 is therefore possible using Progress Values directly and does not require sophisticated statistical understanding. In this regard, Progress Values are not only useful to researchers concerned with measurement precision, they also provide a useful index of growth to servicing clinicians. Requiring clinicians to repeatedly administer standardized assessment to their clients does not need to be strictly a practice for program evaluation, as may be the case for percentile ranks and raw scores. As discussed earlier, percentile ranks are not especially sensitive, and raw score changes do not inform clinicians as to whether or not the child has made meaningful change. Progress Values may provide standardized evidence of whether or not a child is progressing and therefore provide insight for clinical decision-making. Highlighting the utility of Progress Values for both program evaluation and clinical decision making may facilitate their adoption by OIHP servicing clinicians.

Despite the benefits of using Progress Values, their use in research of speech and language development broadly, and speech-language outcomes in CHH specifically, has not been adopted, perhaps in part due to difficulty in obtaining the documents required for score conversion. Notably, the PLS-4 examiner's manual does not contain the required information to calculate a Progress Value, but rather this was contained in a separate tool sold separately at a later date. Due to difficulties with sale, the charts are now out of print entirely. The present work demonstrates the utility of using Progress Values in comparing speech and language growth in CHH and, with the inclusion of Growth Scale Values in the PLS-5 examiner's manual, the ease with which researchers and clinicians may use these scores is increasing.

3.1.3 Outcome Assessment Protocols: Conclusions

Using the OIHP database and language protocol to assess the language outcomes of CHH has highlighted important considerations in the development of OIHP language protocols, as well as the potential for these protocols to inform clinical decision making.

First, data collection procedures need to be more comprehensive and consistent in order to better understand the achievement of children in the OIHP. Without information concerning a child's language environment or communication modality, interpretation of PLS-4 scores are limited. Furthermore, agreement amongst regional sites needs to be reached in how to document PLS-4 information; it is impossible to compare children's performance when their scores were calculated using different methods. Having all sites follow the procedure outlined in the PLS-4 examiner's manual would be the most reasonable approach given that doing otherwise would invalidate the use of the test norms.

Second, using a single outcome measure only allows for very broad interpretations, and the PLS-4 is not especially sensitive to differences in ability in CHH. While the PLS-4 is useful in establishing that OIHP children are achieving outcomes consistent with other EHDI programs (Ching et al., 2010; Tomblin et al., 2015), the addition of speech-language outcome measures that are more sensitive to domains of difficulty for CHH would be more clinically informative in order to address these potential concerns. Additional measures would also be important in documenting how OIHP intervention protocols are able to succeed in facilitating specific communication challenges for CHH.

Finally, careful consideration of the types of scores used for the selected language measures can either impede or facilitate clinical utility. Training or workshops for clinicians working with the OIHP in how to use Growth Scale Values in their clinical practice could provide clinicians with an understanding of the extra benefit offered by this type of language outcome assessment, beyond program evaluation

3.1.4 Language Outcomes in a Canadian Context: Success of the OIHP

The goal of the OIHP is "to facilitate the affected child's development of communication skills and readiness for school" (Bagatto et al., 2010, pS71). While our interpretations are limited by reliance on a single omnibus measure, it is clear that the OIHP's diagnostic and intervention protocols are facilitating the achievement of this goal. On average, children in our sample performed at the 39th and 44th percentile on the Auditory Comprehension and Expressive Communication scales, respectively. While these percentiles are not significantly different from the PLS-4 normative mean of the 50th percentile for the Auditory Comprehension and Expressive Communication scales in our sample ($t(17)=-1.32$, $p>0.05$ and $t(18)=-0.642$,$p>0.05$ respectively), this is likely due to our limited sample size. Additional analyses of PLS-4 language assessment conducted on *all* children in the 2008 and 2011 cohorts who had their PLS-4 language assessed at any time *after* hearing aid fitting (but not necessarily before) demonstrates that these children are performing similar to those included in our sample (at the 31st and 33rd percentile), but these scores are significantly below the normative mean for both Auditory Comprehension and Expressive Communication scales ($t(106)=-5.41$, $p<0.05$ and $t(104)=-4.84$, $p<0.05$) and are not more than 1 *SD* below the mean ($t(106)=4.75$, $p<0.05$ and $t(104)=5.11$, $p<0.05$). These results are in line with CHH's performance on composite measures of language ability in the OCHL study (Tomblin et al., 2015) and CHH's performance in the Longitudinal Outcomes for Children with Hearing Loss (LOCHI) study in Australia (Ching et al., 2010). While children in the OCHL study did not meet criteria for a language impairment, they did, on average, score significantly below their normal hearing peers (Tomblin et al., 2015). Children in the LOCHI study were assessed on the PLS-4 at age three, and these children performed at, or below, 1 SD below the mean (Ching et al., 2010). This suggests that even though CHH enrolled in the OIHP are performing below the PLS-4 normative sample, their performance is appropriate when considered in relation to other EHDI programs. Despite our inability to assess particular areas of difficulty for CHH serviced with the OIHP, these early analyses suggest that, within the OIHP, CHH who wear hearing aids are achieving expected, and acceptable, levels of language performance.

It is also important to consider the OIHP's success in not only the language outcomes of the children they serve, but also in their dedication to continuous protocol improvement. The UWO PedAMP represents a thorough, evidence-based approach to evaluating the auditory behaviours of CHH. Although the current language assessment procedures have not been developed with the same level of rigor as the UWO PedAMP, researchers at the University of Western Ontario are currently working to develop an additional high quality language assessment protocol to accompany the UWO PedAMP. The OIHP's commitment to improvement is similarly evidenced in improvements in data management practices as described above (see 3.1.1 for details). In this regard, this project highlights Ontario's role as a leader in Canadian EHDI service provision and models the importance of program evaluation for other Canadian provinces and territories.

This project has important implications for other provinces and territories that are struggling to gain support for an EHDI program by providing both evidence of benefit, and a model for success. On average, the children serviced by the OIHP have been able to achieve overall age appropriate language outcomes. Our results also demonstrate that, prior to hearing aid fitting, severity of hearing loss negatively predicted continued declines in language performance relative to the PLS-4 norming sample. Importantly, these declines did not appear to be cumulative *after* children receive their hearing aids. Rather, hearing aid amplification appears to preserve language ability, and protect children from declines associated with their hearing losses. These benefits are the greatest for children at the greatest risk: those with the greatest severity of hearing loss, and the weakest pre-fitting language abilities. Our results therefore echo the sentiments of previous research, as well as the JCIH and CIHTF: early intervention is necessary for CHH to reach their full potential (Bagatto et al., 2010, 2016; Ching et al., 2010; JCIH 2013; Moeller et al., 2000; SAC, 2010; Thompson & Mehl, 1998; Tomblin et al., 2014, 2015; Wake et al., 2016; Yoshinaga-Itano, 2003b). This project provides evidence collected in a Canadian context, using an existing EHDI program, that early intervention is beneficial for children with hearing loss.

While language assessment protocols need refinement within the OIHP, the protocols that are currently in place serve as a template for other provinces to adopt, and from which to learn. Furthermore, the achievement of CHH enrolled in the OIHP are both expected and acceptable, based on other EHDI outcome research that has used the PLS-4 as a language assessment tool. We recommend, therefore, that provinces in the planning stages of EHDI program development look to the OIHP for a program model with demonstrated success.

3.2 Theory Generation: A Preservation Hypothesis of Hearing Aid Benefits for CHH

A second goal of this project was to explore contributions of pre-hearing aid fitting language abilities to post-fitting outcomes. Given demands for evidence of EHDI benefit in order to guide program development (Bess & Paradise, 1994), research with a CHH population has emphasized benefits of EHDI programs in comparison to regions without EHDI programs, as well as identifying factors that may predict the variability of speech and language outcomes in children within EHDI programs.

There is sufficient evidence to demonstrate that EHDI programs benefit CHH and work is starting to identify the variables that influence outcome variability in these samples (Ambrose et al., 2015; Ambrose, VanDam & Moeller, 2014; Moeller, 2000; Tomblin et al., 2014, 2015). As work coming from the OCHL study repeatedly demonstrates, and as discussed in Chapter 1, early hearing aid fitting is not the only component to successful speech and language development for CHH. Quality of hearing aid fit and features of caregiver input predict both language growth, and overall outcome, after hearing aid fitting (Tomblin et al., 2015). Additionally, severity of hearing loss did not continue to predict language growth, or outcomes, after fitting when these additional factors were controlled (Tomblin et al., 2015), which is consistent with our suggestion that hearing aids protect the developing language system from further declines associated with hearing loss. The OCHL work, however, did not explore language abilities prior to amplification and, as this project illustrates, the pre-amplification abilities of CHH *is* one of the variables that influences individual outcomes. As research in EHDI programming moves away from asking whether or not EHDI programs benefit CHH and towards asking what factors predict and maximizes benefits for children, a full account of these factors necessitates considering the role of pre-hearing aid fitting language ability and development.

The speech and language development of infants born with congenital hearing loss is largely understudied. *How* infants with PCHL are able to learn from unamplified auditory information is relatively unknown, although a number of studies have examined the speech perception, word learning, and multisensory integration skills of children who use Cochlear Implants (CI; Bergeson, Houston, & Miyamoto, 2010; Horn, Houston, & Miyamoto, 2007; Houston et al., 2012; Rouger, Lagleyre, & Fraysse, 2007). Children who receive CIs, however, are not representative of the entire CHH population, and represent a group of children with severe to profound HL who do not benefit from hearing aids. Studying CHH with consideration to the entire spectrum of hearing loss severities allows for the identification of different cognitive effects, and understanding how the infant mind learns from varying degrees of degraded input.

This project represents a first step in addressing these questions. Our suggestion that hearing aids benefit CHH by preserving language ability is based on a small, extremely heterogeneous, sample. Further testing of this hypothesis is needed.

3.2.1 Testing a Preservation Hypothesis: Future Directions

Providing a conclusive explanation for the mechanism by which hearing aids benefit CHH was hindered by the retrospective nature of this study, restrictions in sample size, and measurement limitations that have raised the possibility of our data being sample specific. Future, prospective research is needed to confirm, or refute, our preservation hypothesis.

Future work will benefit from careful consideration of information to which we did not have access in our databases. For instance, features of a child's language environment such as caregiver speech, which is related to socio-economic status (SES) or maternal education, is known to impact language ability (Hoff, 2006, 2013), even in CHH (Ambrose et al., 2014; Tomblin et al., 2015). It may be the case that preservation of language ability is a mechanism of benefit *only* for a specific subset of CHH such as those without other risk factors, such as low SES, and that the mechanism of hearing aid benefit interacts with other factors. Additionally, preservation of language ability may not be the case for *all* CHH and may change in response to differing levels of hearing loss severity. Conducting linear regressions such as those done in this study assumes a linear relationship between severity of loss and language ability. Nonlinear regression analyses, or more complex modeling techniques may identify a more nuanced severity and ability relation than was captured here. Conducting these analyses, however, requires a much larger sample size than that to which we had access, as well as larger numbers of children across the spectrum of hearing loss severity.

Our conceptualization of ability, as well as growth, needs to be expanded in future testing of this hypothesis. In this project, ability was assessed using a single, omnibus language measure, the PLS-4. Analysis of the Auditory Comprehension and Expressive Communication scales revealed slightly different results. Although severity of hearing loss was not associated with additional percentile rank decline on either scale, the relation of Progress Value growth differed for the two scales. Progress Value change was only related to severity of hearing loss and initial performance for the Auditory Comprehension scale, suggesting that the benefit provided by hearing aids may be different for the two domains. Despite these differences, the skills assessed within each scale are still quite variable, and these subdomains may be differentially affected by amplification as well. For instance, the Auditory Comprehension scale does not isolate differences between receptive vocabulary and sentence comprehension. Our reliance on an omnibus measure limited our ability to identify with precision what areas of language development are impacted in what way by hearing aid fitting. Further testing of our preservation hypothesis will benefit from consideration of overall language ability *and* specific domains of language, in particular, specific domains within Auditory Comprehension.

Our null findings for a relation of Expressive Communication Progress Value change may represent language domain differences in hearing aid benefit, but may alternatively be an artifact of our conceptualization of Progress Value change. Change scores were calculated using only one assessment after hearing aid fitting and we are unable to consider the distal benefits of hearing aid usage. Given the nature of the relation between Auditory Comprehension and Expressive Communication, it is possible that improvements in Auditory Comprehension may manifest themselves before benefits in Expressive Communication, and therefore the relation we observed between Progress Value Growth, hearing loss severity, and initial language ability, may be observable at later ages. Using only two assessment periods, operationalized as pre-hearing aid fitting and post-hearing aid fitting, are additionally problematic when we consider the overall developmental trajectory of language development, and unpacking the implications of a preservation hypothesis.

Preservation necessarily suggests that hearing aids are halting a downward trajectory. This project provided some indirect evidence that this may be the case; hearing loss severity attenuated the increases in PLS-4 Progress Values and raw scores associated with increasing age before hearing aid fitting, and that relation was no longer present after fitting. However, this project did not *directly* evaluate language trajectory – doing so would require *multiple assessments* both before and after hearing aid fitting, and modeling growth rates, as well as relative change in growth rate. If a preservation hypothesis is truly the mechanism by which hearing aids benefit CHH, then we would expect an initial falling behind, or decline, in standing relative to normal hearing peers followed by a significant shift in inter-assessment growth after fitting. The variables of interest in these analyses are not differences between percentile ranks, but the differences between the changes in percentile ranks before, and after amplification. Given the repeated administration of the PLS-4 every 6 months is mandated in the current OIHP protocol, analyses of this nature may be possible. However, conducting this analysis requires access to cohorts born in years when regular PLS-4 testing was fully implemented. The issues surrounding conceptualizing growth trajectory highlights the additional concern in how best to measure language development. The limitations of using the PLS-4 have been considered above, however, it is important to highlight that standardized measures may not be particularly sensitive to the subtle changes in development that occur in the first two years of life, and may not be sensitive enough to detect differences in initial trajectories this early in development. Future research examining a preservation hypothesis will benefit from not only considering benefit in relation to changes in developmental trajectory, but also use evidence from sensitive measures of speech and language processing, perhaps including sophisticated neuro-imaging techniques.

Exploring a preservation hypothesis in greater depth is valuable not only in the confirmation or refutation of the hypothesis itself. *If* the preservation of language ability is confirmed to be a mechanism of hearing aid benefit, there are a number of significant clinical and theoretical implications that follow and are worthy of study.

3.2.2 Preservation as Benefit: Implications for Clinicians

Understanding how the infant mind uses unamplified input, and similarly reconciles unamplified input is of direct clinical importance because this knowledge facilitates an understanding of *how* a hearing aid benefits CHH. For instance, if our preservation hypothesis holds under more rigorous testing, what does this mean for interventionists?

First, this project demonstrated that early amplification is critical for language outcomes. EHDI programs aim to have children fitted with hearing aids promptly following permanent hearing loss confirmation and parent readiness (Bagatto et al., 2010, 2016; JCIH 2007; SAC, 2010) and thus it is not feasible to recommend hearing aid fitting any earlier. In this regard, this project re-iterates the benefits of early hearing aid fitting as recommended by the JCIH and CIHTF.

Rather than advocate for even earlier fitting protocols, this project suggests that careful attention must be paid to the developmental period prior to hearing aid fitting. If hearing aids protect against speech and language declines associated with hearing loss severity, then CHH are without protection until hearing aid fitting. Exploration of additional factors that protect against the language declines associated with hearing loss severity *before* fitting will facilitate early interventions to maximize speech and language outcomes. For example, prior to amplification, careful counseling and training for caregivers to provide optimal speech and language input for CHH may capitalize on the preservation provided by hearing aids.

Beyond facilitating earlier interventions, a preservation hypothesis has implications for clinical planning. If it is the case that hearing aids stabilize language performance relative to other children, and this language performance is measurable early in development, clinicians may use this information in determining long-term goals for the child, anticipating future challenges, and beginning to prepare parents for navigating helpful additional communicative and academic supports.

3.2.3 Preservation as Benefit: Implications for the Infant Speech Perception Literature

This project raised many more questions that it answered. Research surrounding infant language development and speech perception argues that early auditory experiences are foundational for later language development (Jansson-Verkasalo et al., 2010; Kuhl et al., 2008; Moon et al., 2012; Tsao et al., 2004). In this regard, this project re-affirms the importance of early auditory experience: unamplified language achievement is protected from further declines associated with hearing loss severity. However, this project did not offer any insights into the cognitive or neurological mechanisms by which preservation is possible. If, with additional work investigating a preservation hypothesis, it becomes clear that preservation is a mechanism by which hearing aids benefit language outcomes of CHH, this poses interesting implications for the role of early auditory input in language development, and understandings of sensitive periods to auditory information.

A preservation hypothesis necessitates that the auditory and linguistic deficits associated with PCHL early in development do not result in cascading deficits in later language development *if* the child receives appropriate, timely, intervention. In fact, the results of this project suggest the opposite: Progress Value analyses revealed that the children who grew the most in their auditory comprehension abilities after hearing aid fitting were those who were initially at the greatest risk, that is, those children with the greatest hearing loss severity and the lowest Auditory Comprehension Progress Values before fitting. How is the infant mind able to overcome what appears to be an initial downward trajectory? One possible explanation is that the increased amplification provided by hearing aids is sufficiently *different* from the auditory input children receive without amplification as to increase sensitivity to the previously missed auditory information. This difference, as provided by periods of use with a hearing aid and periods of use without, may increase the perceptual salience of certain auditory information. Cognitively, increased sensitivity to previously missed information may facilitate differential statistical extraction of this information.

Considering hearing aid benefit in regards to differences in levels of amplification is relatively new to the CHH outcome literature. Tomblin et al. (2014) have recently advocated for using a residual-speech intelligibility index (rSII; see Chapter 1 for a discussion) to precisely explore differences in amplification levels because rSII captures the magnitude of increase in aided SII accounting for aided SII's relation with unaided SII. rSII, therefore, may be considered as a unit of *dosage* – high rSII values indicate that hearing aids are providing large gains in access to auditory information, and low rSII indicates that the hearing aid is providing a smaller increase. Speech and language benefits, therefore, may not be related to either unamplified or amplified SII, if growth associated with hearing aid use is related to increased access, salience, or sensitivity, to auditory input. Rather, we would expect that rSII would be the logical predictor of growth rates, which is exactly what the OCHL study found (Tomblin et al., 2015). In the OCHL study, rSII predicted growth on composite scores of speech and language ability independently of severity of hearing loss, and independently of amplified SII. This suggests that having access to 90% of the speech spectrum is not *necessarily* better for *improving* speech and language outcomes than having access to only 35% of the speech spectrum. That is, we may expect a child with access to 90% of the speech spectrum to perform better on standardized language assessments than a child with access to 35% of the speech spectrum, but that the first child may *change* less than the second pre- and post-hearing aid fitting if the boost (rSII) for the child with an amplified SII of 35% is greater than the rSII of the child with an amplified SII of 90%.

rSII's role in predicting growth in language outcomes has interesting implications for cognitive and neurological theories of infant language acquisition. Recall that hearing loss is not a language disorder *per se*, but rather a sensory impairment with implications for the cognitive processes of language acquisition. Studying how infants with PCHL are able to learn from degraded auditory input, and consequently reconcile amplified input, provides insights into how a *typical mind* handles *atypical input*. If change in access to speech input (indexed by rSII) is causally associated with change in language performance, then this suggests that the infant is sensitive to these changes and is able to exploit them in some way.

Infants are particularly sensitive to the statistical properties of language information contained in their environment (see Chapter 1 for a discussion). For instance, infants can learn to discriminate between phonemes if the acoustic variance follows a bimodal distribution (suggesting two canonical sounds), but fail to discriminate the *same phonemes* if the acoustic variance falls in a unimodal distribution (suggesting allophonic differences; Maye et al., 2002). These findings have been extended to visually articulated information. Using the *same* phonetic distributions (unimodal) but presented with either unimodal or bimodal articulatory cues, infants continue to discriminate phonemes in the bimodal, but not the unimodal conditions (Teinonen, Aslin, Alku, & Csibra, 2008). As it pertains to PCHL, the increased access to the speech spectrum may provide access to previously missed auditory information and facilitate the statistical extraction of this information. As the differences between amplified and unamplified input (rSII) increase, the perceptual salience of missed information may increase, highlighting what was missed and facilitating learning from this information.

Exploring both the clinical and theoretical implications of a preservation hypothesis highlights the interdependence of these two disciplines. Identifying ways to maximize language ability prior to amplification in order to optimize hearing aid benefit rests on understanding how infants process unamplified input. Exploring how infants with PCHL process both unamplified and amplified input necessitates the consideration of functional outcomes. Proposing a preservation hypothesis, and considering the ways in which preservation may be the mechanism of benefit for CHH who wear hearing aids, provides a starting point to begin evaluating these various hypotheses and guide future research in CHH.

3.3 Conclusions

This project was a retrospective cohort analysis of clinical data collected and managed by the Ontario Infant Hearing Program. Our project aimed to evaluate the language outcomes children serviced by the OIHP as well as address theoretical gaps in the infant speech perception and CHH literatures. Our results indicate that increasingly severe hearing loss negatively predicts both language ability relative to same-age peers and level of language ability in CHH at the time of hearing aid fitting. Furthermore, in our sample, these children did not change in their relative standing from the time of amplification to sometime afterwards, despite significant growth in language skills. Skill acquisition in Auditory Comprehension was greatest for those considered to be at greatest risk: the infants and children with the most severe hearing losses, and the weakest language abilities. The non-significant changes in relative standing from the time of hearing aid fitting to afterwards are interpreted as evidence that hearing aids may be preserving the initial language ability of CHH. This suggestion was further supported by analyses of factors predicting language ability after fitting: severity of hearing loss only contributed to language ability after fitting via its relation with language ability prior to fitting, rather than continued effects.

This work, while limited in its ability to provide definitive explanations of the mechanism by which hearing aids benefit children with hearing loss, highlights the utility of considering pre-amplification language ability from numerous perspectives. Studying pre-amplification language abilities is necessary for accurately conceptualizing benefit, highlighting malleable factors prior to hearing aid fitting that may benefit eventual outcome, informing theoretical understanding of infant speech perception, and facilitating evaluations of EHDI programs. It is therefore imperative that future research continues to explore this period of development in CHH, and that all stakeholders in the development of a child with hearing loss consider this period in their interactions with the affected child.

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