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# SWEDISH CHILDREN WITH MODERATE HEARING LOSS – ON THE IMPORTANCE OF MONITORING AUDITORY AND EARLY SPEECH DEVELOPMENT THE FIRST THREE YEARS

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# SWEDISH CHILDREN WITH MODERATE HEARING LOSS – ON THE IMPORTANCE OF MONITORING AUDITORY AND EARLY SPEECH DEVELOPMENT THE FIRST THREE YEARS

## THESIS FOR DOCTORAL DEGREE (Ph.D.)

By


**Anna Persson**


The thesis will be defended in public at Karolinska Institutet, 4U Solen, Alfred Nobels Allé 8, Karolinska University Hospital Huddinge on Friday Dec 18th, 2020 at 9.00 a.m.

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To all children with hearing loss and their families, and to all professionals in the field of paediatric audiology

“I am only one, but I am one. I cannot do everything, but I can do something. And I will not let what I cannot do interfere with what I can do”.

*Edward Everett Hall*

# ABSTRACT

The overall aim of this thesis was to investigate the auditory and early speech development in a group of children born with moderate hearing loss (HL) who were fitted with hearing aids (HAs) before the age of 6 months, and to compare their development to a group of children with normal hearing (NH). More specific aims were to examine the impact of auditory variables on the early speech development in the children with HL and to validate a questionnaire of auditory development in the children with NH.

This PhD project consisted of four longitudinal studies. Participants included a group of children with moderate HL (n=11) and a reference group of children with NH (n=29). Data was collected at the ages of 10, 18, 34, 30 and 36 months with assessments of early speech development and auditory variables that were compared between groups. In **Study I,** the Swedish version of the LittlEARS® Auditory Questionnaire (LEAQ) was externally validated in children with NH. In **Study II,** HA use from first fitting to 36 months of age and the impact of hours of HA use on auditory development and functional performance were investigated. In **Study III,** consonant production in babbling and early speech at 10 and 18 months and consonant proficiency at 36 months were examined. In **Study IV,** expressive vocabulary between the ages of 18 to 30 months was investigated and compared between groups.

The Swedish version of the LEAQ showed equal validation properties to the original version albeit found to be similar in content to an inventory of vocabulary. The variability in hours of HA use was large and only two children reached full-time HA use at all ages. Auditory development was found similar between the groups but the children with HL presented with lower scores on functional auditory performance in noise at 30 and 36 months of age. The children with HL showed delays in their consonant production in babbling and early speech at 10 and 18 months compared to the children with NH. At 36 months, there was no significant difference between the groups on consonant proficiency. The number of produced words was similar between the groups at 18 months but at 24 months of age there was a gap which increased even further at the age of 30 months, disadvantaging the children with HL. The most prominent auditory variable found to impact the outcomes on the early speech measures was hours of HA use, meanwhile scores on auditory development, functional auditory performance, and aided audibility showed weaker relationships.

Despite early fitting with HAs the children with moderate HL showed delays in their early speech development and challenges in auditory functional performance in noise. The findings from this thesis suggest that careful monitoring at specific ages with appropriate methods of early speech development and auditory variables could help professionals to identify children at risk and work in a preventive way. This calls for cross-professional collaboration within the clinical setting. Attention is also needed to make sure that parents and other caregivers outside the clinic (e.g. preschool teachers) receive information so they can use appropriate strategies to reduce the risks of language delays in children with moderate HL. Albeit acknowledging the limitations to generalize the findings from this thesis to a larger population, the results and experiences from the longitudinal project are in line with current research that promotes monitoring of auditory and spoken language development the first three years in children with HL.

# SAMMANFATTNING

Det övergripande syftet med denna avhandling var att undersöka hörsel- och tidig talutveckling hos en grupp barn födda med måttlig hörselnedsättning (HNS) som fick hörapparater (HA) före 6 månaders ålder, och att jämföra deras utveckling med en grupp barn med normal hörsel (NH). Mer specifika mål var att undersöka ett antal så kallade hörselvariabler och deras påverkan på den tidiga talutvecklingen hos barnen med HNS samt att undersöka den externa validiteten av ett frågeformulär om hörselutveckling.

Doktorandprojekt omfattar fyra longitudinella studier. Deltagarna var en grupp barn med måttlig HNS (n=11) och en referensgrupp barn med NH (n=29). Datainsamlingen bestod av bedömningar av tidig talutveckling och hörselvariabler vid åldrarna 10, 18, 24, 30, och 36 månader som sedan jämfördes mellan grupperna. I **Studie I** verifierades den svenska versionen av LittlEARS® Auditory Questionnaire (LEAQ) genom data från barnen med NH. I **Studie II** undersöktes HA-användning från första anpassningen till 36 månaders ålder och effekten av användningstimmarna på hörselutveckling och funktionell lyssningsförmåga. I **Studie III** undersöktes konsonantproduktion, dels i joller och tal vid 10 och 18 månader, dels andel korrekta konsonantproduktioner vid 36 månaders ålder. I **Studie IV** undersöktes expressivt ordförråd mellan 18 och 30 månader och jämfördes mellan barnen med HNS och NH samt hur antalet ord påverkades av hörselvariablerna och antalet tryckstarka konsonanter vid 18 månaders ålder.

Den svenska versionen av LEAQ överensstämde med originalversionen, men visade att frågornas innehåll till stor del liknade en checklista för ordförråd. Variationen i timmar av HA-användning var stor och endast två barn nådde HA-användning på heltid vid samtliga åldrar. Hörsel-utvecklingen visade sig vara lika mellan grupperna men barnen med HL hade sämre funktionell hörselförmåga i buller vid 30 och 36 månaders ålder. Barnen med HL visade förseningar i sin konsonantproduktion i joller och tidigt tal vid 10 och 18 månader jämfört med barn med NH. Vid 3 års ålder fanns det ingen signifikant skillnad mellan grupperna när det gällde artikulation. Antalet producerade ord var lika mellan grupperna vid 18 månader men vid 24 månaders ålder fanns det ett gap som ökade ytterligare vid 30 månaders ålder, till barnen med HNS nackdel. Den mest framträdande hörselvariabeln som påverkade resultaten på de tidiga talmåtten var antal timmar med HA-användning, medan hörselutveckling, funktionell hörselförmåga och hörbarheten i barnens HA påvisade svagare korrelationer.

Trots tidig anpassning av HA visade barnen med måttlig HNS förseningar i deras tidiga talutveckling och utmaningar gällande funktionell hörselförmåga i buller. Resultaten från denna avhandling tyder på att regelbunden uppföljning vid specifika åldrar med lämpliga metoder för tidig talutveckling och hörselvariabler skulle kunna fånga upp tidiga avvikelser så man kan arbeta förebyggande. Detta kräver tvärprofessionellt samarbete inom den kliniska verksamheten. Uppmärksamhet behövs också riktas till barnens föräldrar och andra vårdgivare utanför kliniken (till exempel förskolelärare) så att lämpliga strategier för att minska risken för språksvårigheter hos barn med måttlig HNS implementeras. Trots begränsningarna att kunna generalisera resultaten från denna avhandling på grund av det begränsade deltagarantalet så är resultaten och erfarenheterna från detta longitudinella projekt i linje med övrig aktuell forskning som förespråkar uppföljning av hörsel- och talspråksutveckling de tre första åren hos barn med HNS.

# LIST OF SCIENTIFIC PAPERS

I. **Persson A**, Miniscalco C, Lohmander A & Flynn T. (2019). Validation of the Swedish version of the LittlEARS® Auditory Questionnaire in children with normal hearing - a longitudinal study. *International Journal of Audiology, 58(10), 635-642.*

II. **Persson A**, Al-Khatib D & Flynn T. (2020). Hearing aid use, auditory development and auditory functional performance in Swedish children with moderate hearing loss during the first 3 years. *American Journal of Audiology, 29(3), 436-449.*

III. **Persson A**, Flynn T, Miniscalco C & Lohmander A. Impact of auditory variables on consonant production in babbling and early speech in children with moderate hearing loss – a longitudinal study. *Manuscript.*

IV. **Persson A**, Marklund U, Flynn T & Lohmander A. Expressive vocabulary development in children with moderate hearing loss - the impact of auditory variables and early consonant production. *Manuscript.*

Scientific paper not included in the thesis:

Marklund U, Lacerda F, **Persson A** & Lohmander A. (2018). The development of a vocabulary for PEEPS-SE-profiles of early expressive phonological skills for Swedish. *Clinical Linguistics & Phonetics*, 32(9), 844-859. doi: 10.1080/02699206.2018.1445780.

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# LIST OF ABBREVIATIONS

| | |
|---|---|
| CB | Canonical Babbling |
| CI | Cochlear Implant |
| dB HL | Decibel Hearing Level |
| EI | Early Intervention |
| HA | Hearing Aid |
| HL | Hearing Loss |
| HoH | Hard of Hearing |
| JCIH | Joint Committee of Infant Hearing |
| LEAQ | LittlEARS® Auditory Questionnaire |
| NH | Normal Hearing |
| UNHS | Universal Newborn Hearing Screening |
| PCC-A | Percentage of Consonants Correct – Adjusted for age |
| PEACH | Parent´s Evaluation of Aural/Oral Performance in Children |
| SECDI | MacArthur-Bates Communicative Developmental Inventory, Swedish adaptation |
| SET | Special Education Teacher |
| SII | Speech Intelligibility Index |
| SLP | Speech Language Pathologist |
| TD | Typical Development |
| WCM-SE | Word Complexity Measure, Swedish adaptation |
| WHO | World Health Organization |

# 1 INTRODUCTION

Hearing plays an essential role in typical development of speech and language skills. Access to acoustic input is a fundamental condition from which spoken language learning takes place (Kuhl, 2004). For children who are born with hearing loss (HL), this constitutes a risk for delays in their speech and language development, and furthermore, poor academic and social development (Ching et al., 2013; Moeller & Tomblin, 2015; Yoshinaga-Itano et al., 2001). The group of children with HL is often referred to as heterogeneous and the consequences of a HL bring additional factors affecting their spoken language development compared to children with normal hearing (NH).

The introduction of universal newborn hearing screening (UNHS) changed the scene for generations of children born with HL to come. UNHS was rolled-out in Sweden in the late 1990´s and was implemented at a national level in 2007. Early detection and diagnosis of HL followed by early intervention (EI) services (e.g. "habilitation" in Swedish) have led to improved spoken language outcomes in children with HL although at a population level they are still 1-2 standard deviations below children with NH (Ching et al., 2017; Moeller & Tomblin, 2015; Yoshinaga-Itano et al., 2017). These results mainly derive from international research. According to the Swedish Agency for Health Technology Assessment and Assessment of Social Services there is limited scientific evidence that earlier detection and commencement of habilitation promotes communication and language development in Swedish children (SBU, 2020). Existing evidence on spoken language development of Swedish children with sensorineural HL has been dominated by research of children with profound HL using cochlear implants (CIs) (Asp et al., 2015; Karltorp et al., 2020; Löfkvist et al., 2014) meanwhile studies of children with mild to moderate HL are sparse (Borg et al., 2007; Stübner et al., 2020), and even more so with regards to children under 3 years of age (Löfkvist et al., 2019). In 1990, a national quality register (e.g. Hörselbarnsregistret) started to collect data of children with CIs, which was updated in 2014. The second update of the register began in 2016 to also include children with a permanent HL in at least one ear with a four-frequency pure-tone-average of >29 dB HL. The current version of the register starts collecting data at 3 years, leaving data on an important period of early auditory and spoken language development at loss. One of the major goals with EI of children with HL is to promote opportunities for them to develop effective communication. Therefore, it is important to identify children at risk for speech and language delay and/or disorder as early as possible. Research in the field of speech and language pathology has found several variables in young children to predict later speech and language development in children with typical development. However, studies on these predictors in children with HL are limited. Therefore, the rationale for this thesis was to investigate auditory and early speech development from this perspective, and to examine the impact of auditory variables identified in research to affect speech and language outcomes in children with HL.

# 2 BACKGROUND

In a broad sense, *language* can be defined as a set of symbols people use to communicate, and *speech* as one way of expressing language. Other ways to express language are through sign language and writing. This doctoral project aimed to investigate the early speech development in children with HL from birth to 3 years. Throughout this thesis, the term “early speech development” refers to the development of early use and proficiency of consonants produced in babbling and early speech, as well as early use of words. Albeit word production is also included in the development of language, this thesis has focused on the number of words that parents have reported that their children use. The term “spoken language” is used when referring to research that have included methods that measure both speech and language outcomes.

## 2.1 EARLY SPEECH DEVELOPMENT

In typically developing (TD) children there is large variability in the early spoken language development (Stoel-Gammon, 2011). Thus, there is a prevalence of 6-7% of preschool-aged children demonstrating developmental language impairments (Law et al., 2000). Spoken language development is closely intertwined with auditory development. As HL constitutes a risk factor it is important to rule out if observed difficulties in early speech development are caused by the lack of access to auditory input, or something else (or a combination). Difficulties in the development of spoken language in children with HL may not only be due to the HL itself (Hansson et al., 2000) which can make it challenging to diagnose language impairments in this clinical group. One way to investigate this is to examine the variables found to predict later spoken language outcomes in the children with HL, as well as controlling for auditory variables. The following paragraphs will describe the auditory and early speech development separately and the consequences a HL may have on these areas of development.

### 2.1.1 Typical speech development 0-3 years

With regards to consonant production, the typical development is a gradual process starting from around 6 months of age up to 6-7 years of age. During the first 3 years, the speech development often comprises of inconsistent productions including omissions and substitutions (Vihman & Greenlee, 1987). The age at which children acquire consonants of the ambient language may vary between studies, mainly because different criteria of what is required “for a consonant to be acquired” have been applied. There is, however, a consensus in the literature that there are milestones in early speech development most TD children follow, and to which children develop at their own rate (Morgan & Wren, 2018). Before the appearance of the first speech sounds, infants spend months practicing their speech through cooing, grunting, gurling, laughing and making different crying sounds, moving on to more variegated babbling. First to develop are the vowel-like sounds, which proceed the production of the consonant-like. Early babbling development is often described in four stages with overlap: phonation (0-2 months), primitive articulation (1-4 months), expansion (3-8 months) and canonical (5-10 months) (Morgan & Wren, 2018; Oller et al., 1999). In the final stage, infants begin to produce sequences of well-formed consonants and vowels with rapid transition, defined as canonical babbling (CB) (Oller et al., 1999). The most typical consonants in CB are anterior stops, first t/d and then p/b (McCune & Vihman, 2001). The onset of CB by 10 months is robust and seen in the majority of TD infants regardless of socio-economic environment, as well as in infants born premature. However, late onset of CB has been found in clinical groups of children with HL and unrepaired cleft palate (Chapman, 2003; Lohmander et al., 2017a; Stoel-Gammon, 1986). Late onset of CB has also been associated with later speech and language disabilities.

Research investigating different aspects of early vocalizations have found that use of oral stop consonants with anterior placement, and "true" consonants are amongst the most stable variables to predict later speech and language development (McCune & Vihman, 2001; Stoel-Gammon, 2011). True consonants are speech sounds that cannot be confused with sounds that fall into other categories such as vowels, or aspects of speech production such as perceived breath or onset of vowels, thus excluding glides and glottals (Stoel-Gammon, 1988). The most typical sounds in CB correspond largely with the speech sounds in early words (McCune & Vihman, 2001). With ample listening experiences the sounds and words connect to meaning, commencing the building of vocabulary. The first spoken word/s usually appears around 12 months of age, predominantly consisting of nouns with a simple syllable structure (Cox Eriksson, 2014; Vihman, 2014). The following period from 12 to 18 months is characterized by a slow development where single words are learned without any visible structure or organization. These words mainly consist of nouns and common words used in the interaction of young children, the size of the expressive vocabulary being about 25-50 words. After the appearance of the first 50 words, there is an acceleration of rapid growth often referred as the "vocabulary spurt" (Fenson et al., 1993; Stoel-Gammon, 2011). This rapid growth occurs around 18-20 months of age. However, all children do not demonstrate this period of rapid acceleration (D´Odorico et al., 2001) and the individual variability in terms of number of words produced around 2 years of age is large (Berglund & Eriksson, 2000a; Pine et al., 1996), ranging from less than 300 words to more than 500 words (Stoel-Gammon, 2011). Example of factors associated with this large variability in TD children under 3 years of age are higher levels of parental education (Cox Eriksson, 2014) and parental contingency (Lieberman et al., 2019; Tamis-LeMonda et al., 2014). Observational studies have demonstrated that contingency in the communication between a child and the caregiver, like responding fast to a child´s utterance, is associated with vocabulary size (Marklund et al., 2015). This sensitivity in early communication between parents and their children has in turn been found to be strongly correlated to the level of maternal education (Cox Eriksson, 2014). Studies targeting low-income families have found that level of parental education can be overcome through structured intervention around parent-child communication strategies (Leffel & Suskind, 2013). Children with NH that do not follow the milestones in typical early speech development may eventually be diagnosed with speech and/or language disorders. One of the groups identified to be at risk for delays in their spoken language development is children with HL.

### 2.1.2 Implications on speech development for children with hearing loss

Infants who are born with a sensorineural HL have to various extent already missed out on auditory experiences at birth. Although amplification can alleviate the negative consequences of the HL, it cannot restore the damage inside the ear which in turn may affect the acuity of speech perception abilities (Tomblin et al., 2014). Studies of children with HL demonstrate similar outcomes to children with NH in terms of volubility in their early vocalizations but fewer utterances containing canonical syllables (Moeller et al., 2007a; Nathani et al., 2007). Children with HL is also one of the clinical groups that do not meet the milestone of CB by 10 months (Stoel-Gammon, 1988; Löfkvist et al., 2019). Recent research in children following cochlear implantation has demonstrated a rapid progress in their babbling and early speech development (Duchesne & Marschark, 2019; Karltorp et al., 2020; Schauwers et al., 2004) meanwhile the corresponding gains in children with moderate degree of HL after HA fitting are not as clear (Iyer & Oller, 2008; Löfkvist et al., 2019). In terms of consonant use, children with HL show similar development of vowels and bilabials to children with NH but are later in their production of alveolar stops and velars at 2 years of age (Ambrose et al., 2014). Children with HL are also more prone to delete final consonants in words (Ambrose et al., 2014) and to use fewer true consonants in their early speech (Stoel-Gammon, 1988), which is related to later word production.

Accordingly, research of expressive vocabulary in children with HL under 3 years of age have found that they produce significantly fewer words compared to children with NH (Mayne et al., 2000; Moeller et al., 2007b; Vohr et al., 2011). Although the children with HL increase their number of words with age, they do not show the same growth rate. The lower number of words are still present, even when socioeconomic status and comorbid neurodevelopmental disorders are controlled for (Vohr et al., 2011). More children with HL have also been found to perform below the $10^{th}$ percentile on vocabulary norms compared to children with NH (Vohr et al., 2011). Examples of factors associated with the development of expressive vocabulary in children with HL are maternal education, degree of HL and age at HA fitting (Ching et al., 2017) as well as absence of additional disabilities (Cupples et al., 2014; Yoshinaga-Itano et al., 2017).

As previously stated, the group of children with HL present with a large individual variability in their early development of spoken language (Nathani et al., 2008). Some studies describe children with HL to develop spoken language skills on par with their peers with NH by the age of 3 years (Sininger et al., 2010). However, most children with HL still show delays despite early fitting of HAs (Ambrose et al., 2014; Ching et al., 2013; Moeller et al., 2007a). As hearing is a fundamental component in the early speech development, the impact of auditory variables is important to investigate as improvements like fitting before 6 months of age has been found to alleviate the delays to some extent. Accordingly, the first step to mitigate the consequences of HL on spoken language development is the fitting of HAs with optimal aided audibility as early as possible (McCreery et al., 2013).

## 2.2 AUDITORY SYSTEM AND HEARING

### 2.2.1 Terminology

Hearing loss can be defined as a partial or total inability to hear and may occur in one (unilateral) or both ears (bilateral) (Northern & Downs, 2002). Other terminology used in the field of paediatric audiology are hard of hearing (HoH) and deaf. The term HoH refers to people who communicate through spoken language through the help of hearing amplification in accordance with their type and degree of HL (WHO, 2018). From a medical point of view, the term deaf refer to persons who have a HL greater than 90dB HL (Northern & Downs, 2002) but it can also refer to the choice of communicating through sign language and being a part of the deaf culture (Ladd, 2003). The latter definition is often spelled with a capital D to separate the medical term from the cultural identity. Sweden was the first country in the world to acknowledge Swedish sign language as a mother tongue (1981). Children with NH of deaf parents have legal right to receive instruction in sign language, but this is not the case for children with HL of parents with NH. However, as more than 90% of the parents of children born with HL are NH themselves, spoken language is often chosen as the primary communication mode (Mitchell & Karchmer, 2004). The goal of developing spoken language on par with peers with NH is now achievable for children with HL through the benefits of early detection and identification of HL in conjunction with prompt fitting with HAs and/or CIs (Ching et al., 2015; Sininger et al., 2010).

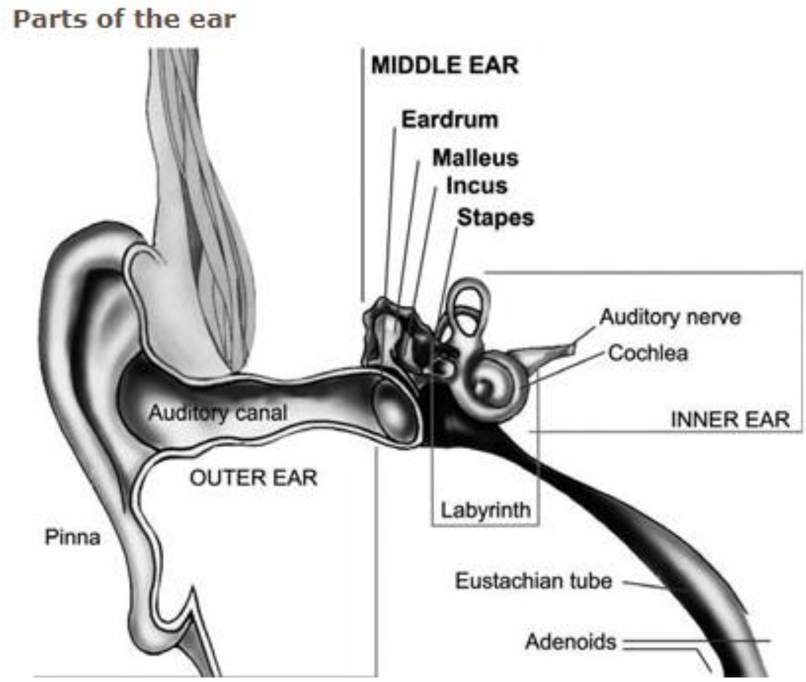
### 2.2.2 Development of the auditory system and hearing

The first step in the development of the auditory system takes place around the 10-12$^{th}$ week of gestation (Hall, 2000) and around the 25$^{th}$ week the auditory system becomes functional (Graven & Browne, 2008). The auditory system requires stimulation from the outside to develop. Examples of research bearing evidence of the listening experience during the time in utero are newborns´ preferences for the mother´s voice (DeCasper & Fifer, 1980) and low frequency sounds (Northern & Downs, 2002) as well as recognition of melodies played by the parents before birth (DeCasper et al., 1994). These findings indicate that infants born with HL may have missed out on important listening experiences already at birth.

### 2.2.3 Anatomy of the auditory system and how sound is perceived

The peripheral hearing system comprises the outer, middle, and inner ear (Figure 1). In typical hearing, sound waves are picked up by the outer ear and travel through the narrow passage of the ear canal which leads to the tympanic membrane. The incoming sounds make the tympanic membrane vibrate and sends the vibrations on to the small ossicles of malleus, incus and stapes in the middle ear. These bones increase the intensity of the sound vibrations and send them on to the snail shaped cochlea through the oval window in the inner ear. The cochlea is filled with fluid and thousands of tiny stereocilia. The vibration coming into the cochlea makes the hair cells move and bend. When they bend, electrical signals are sent via the hearing nerve to the brain. The brain perceives the sound. With meaningful listening experience, the infant brain will eventually learn to identify and understand these signals and attach meaning to them.

**Figure 1**. Illustration of the components of the ear. Reprinted with permission from the National Institute on Deafness and Other Communication Disorders (https://www.nidcd.nih.gov/health/)




## 2.2.4 Types and degrees of hearing loss

The main types of HL are conductive, sensorineural, or mixed (Katz et al., 2009). A conductive HL means that sounds are not conducted efficiently through the outer and/or middle ear (Katz et al. 2009). Sensorineural HL is the most common type of HL and refers to a damage in the inner ear or sensory organ (cochlea and associated structures) or the vestibular cochlear nerve (Northern & Downs, 2002). A mixed HL is a combined conductive and sensorineural HL.

Hearing loss can also be categorized in terms of degree and configuration. With normal hearing you are able to hear soft sounds without any difficulty, which is around 0-20dB HL. In terms of the degree, a HL can be either mild, moderate, severe, profound or total. Classifications of the degrees of HL as measured in decibels hearing level differ to some extent (Clarke, 1981; Katz et al., 2009). By the standards of the WHO, degree of HL are referred to as: slight (26-40dB HL), moderate (41-60dB HL), severe (61-80dB HL), and profound impairment including deafness (81dB HL or greater) (WHO, 2018). A disabling HL in children is by definition referred to a HL greater than 30 dB in the better hearing ear (>40 dB in adults) (WHO, 2018) although current research has found that slight and unilateral losses also impact children´s educational attainment (Huttunen et al., 2019; Lieu, 2013).

The shape of the HL or how it is visualized on the audiogram is called the configuration of the HL. The configuration of the HL depends on the shape of the audiogram and can be referred to as ski and reverse slope, U-shaped, or flat (Northern & Downs, 2002). Depending on the type, degree of HL, and configuration, children receive different kinds of amplification, e.g. bone-anchored, behind-the-ear acoustic HAs, CIs. The focus of this thesis is on children born with a bilateral sensorineural HL of moderate degree fitted with behind-the-ear amplification. This type of HA amplifies the sounds through the damaged inner ear, in contrast to a CI, which requires a surgical procedure to "bypass" the damage in the inner ear through electronic stimulation of the auditory nerve.

### 2.2.5 Prevalence and aetiology of hearing loss

The general prevalence estimates of permanent HL ranges from 1 to 3 in 1000 births (Berninger & Westling, 2011; Mehl & Thomson, 1998). According to a recent study in the Stockholm County, the overall prevalence of all categories of HL >20dB was 3.6/1000 in children 0-18 years of age (Uhlén et al., 2020). For children with sensorineural bilateral moderate to profound HL (>40 dB HL), the prevalence increased from 0.7 to 2.4/1000 from 1 to 18 years of age. For the target group in this thesis (e.g. children diagnosed <1 year of age with a bilateral sensorineural HL of >40dB HL) the prevalence was 0.4/1000. The prevalence numbers of the Stockholm County were found to not be significantly different to prevalence numbers in other high-income countries (Uhlén et al., 2020).

In terms of aetiology, HL can be either congenital or acquired, and temporary or permanent. It can be caused by a variety of factors including; genetic factors, infections during pregnancy in the mother, birth complications, trauma, certain medications, or toxins as well as due to exposure to noise and ageing. HL can also be part of a variety of syndromes which may contribute to the high percentage of children with HL having additional disabilities with numbers ranging between 20-40% (Cupples et al., 2014).

### 2.2.6 Auditory development

Already from birth, infants respond to loud sounds through a variety of behaviours like blinking, sucking and eye widening (Northern & Downs, 2002). Their responses to sounds and use of voice develops gradually in conjunction with other areas of development like fine- and gross motor skills. Contingent responses from interactive caretakers to the early vocalizations help the infant develop the process of the auditory feedback loop which is important for self-monitoring of own speech productions (Northern & Downs, 2002). The infant also learns to localize sounds from side-to-side, down, up and eventually from all directions around the age of 2 years (Katz et al., 2009).

Just like there are milestones in speech and language development, the same is true for auditory development. A simplified way of describing the clinical monitoring of auditory development from birth is through the four levels of detection, discrimination, identification, and comprehension (Cole & Flexer, 1999). The first and fundamental step is hearing a sound or not, referred to as the *detection* of sounds. The second step is *discrimination*, which can be described as the ability to recognize, compare, and distinguish between distinct and separate sounds. At birth, newborns can discriminate between sound levels and duration, but it is not until around 6 months of age they start showing preference for the phonemes of their ambient language (Kuhl, 2004). The ability to discriminate between sounds is often observed through the infants and toddlers attempt to localize between two or more sound sources. For a beginner listener, sounds that are more distinct from each other (e.g. /m/ to /ɕ/) are easier to discriminate compared to sounds that are closer in frequency (e.g. /m/ to /n/) (Estabrooks & Marlowe, 2000). Discriminating between sounds does not imply understanding of what is heard but is needed to develop the third step of *identification.* Identification of all frequencies along the speech spectrum is essential as the acuity of each speech sound (or phoneme) relies on the ability to perceive the various formants that constitutes each speech sound (Ling, 1988). Formants can be described as the frequency peaks in the speech spectrum which have a high degree of energy (Ling, 1988). Identification also refers to the ability to connect a sound or a word to an object or situation. Later in development, identification of words leads to the final step of *comprehension* at different levels, spanning from a single word to following simple directions ending at sentences with complex grammar. How far children with HL will develop along these levels depends to a large extent on the kind and degree of HL (Moeller & Tomblin, 2015). For example, if not intervened with amplification, a profound HL may hinder a child to even detect loud sounds when presented close to the ear. If detectable, the ability to discriminate between sounds also depends on the configuration of the HL making it more or less challenging to perceive sounds in continuous speech at various frequencies. It is common that high frequency sounds are more difficult to detect and discriminate from one another (e.g. /f, ɕ, s/), as well as voiceless consonants (e.g. /p, t, k/) (Cole & Flexer, 1999).

### 2.2.7 Implications on auditory development of children with hearing loss

In addition to the limited hearing caused by the HL itself, the pace of the steps in auditory development also depends on the quantity and quality of targeted stimulation of the auditory pathways. Although children with HL have demonstrated challenges in their auditory development, improved outcomes have been found for those fitted with HAs before the age of 6 months with access to EI services (Ching et al., 2017; McCreery et al., 2013; Yoshinaga-Itano, 1999). The goal is to reduce the time of auditory deprivation, as poorer and less auditory input may also have consequences on other functions than just speech development (e.g. cognitive function, auditory memory abilities). As pure observation of auditory development cannot control for the quality of the sounds that reach the brain, objective methods of hearing and aided audibility are warranted. Functional assessments of auditory development and performance are often evaluated through parental questionnaires, which are further discussed in later sections.

## 2.3 MANAGEMENT OF HEARING LOSS

### 2.3.1 Detection of hearing loss before and after UNHS and the consequences on spoken language development

Before the introduction of UNHS, one way to identify HL in children was through their delays speech and language (Stoel-Gammon & Otomo, 1986) and the mean age of HA amplification was 2-3 years (Ching et al., 2017). The UNHS changed the possibility for children born with HL to achieve an earlier access to sounds and spoken language through the fitting of HAs at only a few months of age. Shortly after UNHS was introduced, research on the benefits of early detection of HL on spoken language outcomes was inconsistent (Kennedy et al., 2006; Korver et al., 2010). Lately, it has been demonstrated that it is the age at fitting of HAs that has improved the spoken language outcomes rather than UNHS itself (Ching et al., 2017; Moeller &Tomblin, 2015). However, early fitting for children with slight to moderate HL would most likely not be possible without the UNHS, unless the HL was suspected to be hereditary.

The demonstrated improvements in spoken language development of children with HL after UNHS, have pushed policies to decrease the recommendations of EI with HAs to even lower ages. The first recommendation was to accomplish detection of HL by 1 month of age, diagnosis of HL by 3 months, and intervention with HAs by 6 months of age (e.g. "1-3-6") (American Academy of Pediatrics, 2007). This was recently changed to "1-2-3" (American Academy of Pediatrics, 2019). In Sweden, UNHS is performed within a few days of life by the maternity hospitals, is free of charge and has an uptake of 98% (Berninger & Westling, 2011). Thus, the evidence on the impact that EI with HAs have on the spoken language development for Swedish children with slight-to-moderate degree is still lacking. The only source of evidence on Swedish children with HL found on the SBU website is Borg et al. (2007) who investigated speech and language in 4-6-year-old children with various types and degrees of HL <80dB HL. Borg et al. (2007) performed an extensive test battery of speech and language assessments that covered nearly 60% of the studied population. Their study also reported on several other factors like preschool placement and impact of signs to the speech and language outcomes. According to their findings with regards to the children with moderate HL, their weakest areas of development in comparison to the control group of children with NH were word production and phoneme discrimination at 4 years, and word and grammar perception at 6 years. They did not find any differences in speech and language scores in terms of gender in the children with HL, but girls in the group with NH performed slightly better. In their study group of 156 children with HL, 30% of the parents reported to use sign-support or sign language with their child. This was highly correlated to degree of HL, and there was a negative correlation between using sign-support to scores on the language measures. The children in the study were born between 2003-2004 and the median age for HA fitting was 3,5 years (Borg et al., 2007) which is an important aspect that needs to be considered when interpreting the results. The Borg et al. study contributed greatly to the status of spoken language abilities of Swedish preschool-aged children with HL. However, this further motivates the need for investigating the current status on speech and language skills in children with HL that have been detected from birth and received HAs in accordance with current recommendations (JCIH, 2019).

### 2.3.2 Early intervention of children with hearing loss: Terminology and content

The longer a HL remains unmanaged without appropriate and well-fitted HAs, the more cumulative the effects appear to be (Tomblin et al., 2015). The hypothesis is that the less time of insufficient auditory access and the longer duration with optimal amplification, the better the spoken language outcomes will be (Tomblin et al., 2015). In Sweden, children with HL is a clinical group identified to be at risk, and accordingly they (and their parents) are offered habilitation services from a range of professionals to alleviate the consequences of the HL on their communication, speech and language outcomes (Rikshandboken i barnhälsovård, 2020).

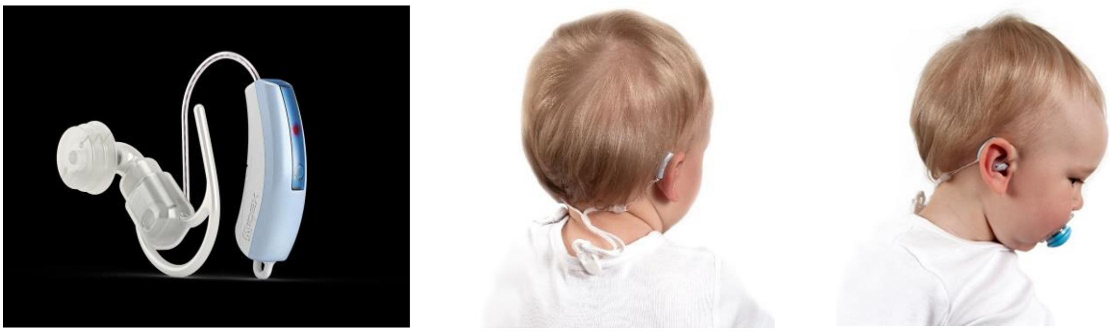
Early intervention (EI) is a term used to describe the services and support that are available for young children who are at risk of poor outcomes. In the field of paediatric audiology EI may include merely diagnosis and amplification of HAs by an audiologist or to incorporate broader services around communication, speech, and language, and psychosocial support from a cross-professional team. International studies on the impact of EI have found improved outcomes if enrolled in a "program" before the age of 6 months compared to after (Ching et al., 2017; Yoshinaga-Itano, 2003). The corresponding Swedish term for EI is "habilitation". The specific content of the Swedish habilitation services of children with HL may vary depending on local resources but is commonly incorporating support from a variety of professionals like audiologists, doctors, psychologists, social workers, special education teachers (SETs), speech and language pathologists (SLPs), sign language instructors, and technical engineers. The habilitation may consist of individual therapy sessions with varying content and time intervals and/or group activities. They may be performed in the clinic, in the home or at the ambient preschool/school of the child/youth. In terms of communication mode, the form may vary from spoken language through listening only, to a combination of supported signs to spoken Swedish and/or sign language depending on the needs and desired outcomes of the child and family. However, as previously mentioned spoken language is a desired outcome of most parents as 90% of parents are NH. There is, to the authors knowledge no "program" in Sweden with a fixed content for all children with HL and their parents. Services are rather recommended and offered, leaving it up to the parents how often and what kind of support they desire. Accordingly, this set-up makes it challenging to evaluate the effectiveness these services may have on spoken language outcomes, as well as controlling for equal care. The registrations to the national register (Hörselbarnsregistret) is optional and do not yet include measures of speech and language before the age of 3 years. However, the joint work around Hörselbarnsregistret has the potential to serve as an indicator of equal care, with the possibility of evaluating the Swedish habilitation services in a large scale. One area of collaboration could be to discuss the process of fitting and verification of HAs in the youngest age group.

### 2.3.3 Hearing aids: fitting and verification

Having a sensorineural HL typically results in an inconsistent and degraded speech signal but infants and toddlers are not able to tell audiologists what they hear. Due to this fact, verifying HA characteristics through prescriptive fitting strategies is important to ensure that the HAs are providing the child with the best aided audibility of spoken language and sounds as possible (Bagatto et al., 2010; 2011a; 2016; Tomblin et al., 2014). The HA fitting process typically begin from the derived hearing sensitivity results from electrophysiological assessments. Once these results are established, behavioural information is gathered to ensure appropriate fitting of amplification. The following text will in short describe the fitting and verification process developed and recommended for the youngest age-group of children with HL by the creators of the University of Western Ontario Pediatric Audiological Monitoring Protocol (UWO PedAMP) (Bagatto et al., 2011a) as used in this project.

The fitting process of HAs includes HA selection as well as choosing fitting strategy. The selection of which HA for the youngest age group depends on a variety of components, most important being the degree and type of the HL. Other important factors when fitting infants are that the HA is of relative size to the child´s ear and durable to manage the wearing and tearing them out. The advanced technology in digital HAs of today are usually programmed with a number of features, which depending on the needs of the child and family are available to connect by the audiologist. Examples of one the HAs worn by the participants with HL in this PhD project are found below in Figure 2, along with a short description how the sounds and speech reach the hearing system through the HA technology.

**Figure 2**. Hearing aid and hearing aids fitted on a child´s ears with a strap and clip attached in the clothing in case they fall out. Reprinted by the kind permission of Widex.



1. Sounds are picked up by the microphone in the HA
2. The acoustic sounds are converted to an electric signal
3. The signal is processed by a central unit (computer), which can highlight some sounds and attenuate others (called signal processing) depending on the fitting rationale
4. The processed signal is sent to the amplifier
5. The amplified signal is transmitted to the speaker (headphone)
6. The speaker converts the electrical signal back into an acoustic signal which is transported via the eardrum and middle ear to the inner ear

In terms of fitting strategies, there are two main rationales used in pediatric HA fittings that are validated. These are the Desired Sensation Level (DSL v5 mi/o) and National Acoustic Laboratories (NAL-NL2) of which the former was used in this project. DSL mi/o and NAL-NL2 provide a “prescription” for HA output which are based on the child´s age and hearing thresholds. These prescriptions provide targets for the HA output which can be used to optimize audibility of speech at soft, conversational, and loud input levels within an impaired or smaller dynamic range. Targets are also provided for maximum power output to ensure that the HA does not exceed loudness comfort levels.

First step in the verification process is to measure the Real-ear-to-coupler difference (RECD). RECDs are performed to account for the individuals unique ear canal or earmold characteristics in comparison to a coupler that simulates the ear canal of an average adult. Due to the difference in ear canal size between infants and adults (also due to growth), RECDs is an important measure to assure a proper fit to achieve appropriate amounts of amplification. If individual RECD measurements may not be performed, average RECD values exist for one-month age intervals through the age of five years. However, they do not take into account the uniqueness of the individuals ear canal and accordingly, these values have been found not to match as precise to the prescriptive targets when compared to measured RECD (McCreery et al., 2013) which could lead to over or under amplification.

Verifications of the HAs should also include measures of aided audibility to estimate how much of the incoming speech that might be audible based on the wearers hearing thresholds, configuration of the HL and the output of the amplification. One such method used by audiologists is called Speech Intelligibility Index (SII) and can be measured unaided or aided. The SII describes the proportion of the speech signal that is audible above the hearing threshold as is presented in values from 0 to 1 (ranging from complete inability to full audibility to access the speech spectrum). Normative SII values for a 65 dB input for children with a mean four-frequency pure-tone average of 45dB HL have been suggested by Bagatto et al. (2011a) to be around 70 (lower and upper 95% confidence interval range being 60 to 80). These normative values were obtained from HA fittings of 161 ears of infants and toddlers (Bagatto et al., 2011a). In general, the SII values decrease with increase in HL (Tomblin et al., 2014). Once the HAs have been verified, the next step is to validate the HAs. Validation refers to a subjective measure that captures the perceived benefit of the HA user. In the case of infants and toddlers, this is usually made through functional assessments that evaluates the child´s functional hearing in a variety of real-life listening environments.

### 2.3.4 Functional assessments

Infants and toddlers do not always provide the audiologist with consistent behavioural responses in the audiology booth. Therefore, subjective standardised measures of auditory behaviour based on parental observations in real-life settings can aid in evaluating the effectiveness of EI with HAs (Bagatto & Scollie, 2013). How children with HL perceive and respond to speech with their HAs may depend on several factors like for how long they have worn the HAs and how far they have come in their auditory development. Other factors affecting the functional outcomes are degree of HL and the (in)consistency of HA use. Results from ratings of auditory development and functional performance have found poorer outcomes in children with HL compared to children with NH (McCreery et al., 2015b). Better scores were found in children with more hours of HA use and greater aided audibility through their HAs. However, these auditory variables have not always been accounted for when investigating early speech development in children under 3 years of age.

### 2.3.5 Auditory variables

Auditory variables found to predict spoken language outcomes in children with HL are age at HA fitting (< 6 months of age) and degree of HL (the milder the better outcomes) (Ching et al., 2013; Fitzpatrick et al., 2011; Moeller & Tomblin, 2015; Yoshinaga-Itano, 1999). Statistical models of largescale studies including predictive factors like age at HA fitting, degree of HL, gender, additional disabilities and maternal education have been found to account for 40-70% of the variation in spoken language outcomes (Ching et al., 2107; Yoshinaga-Itano et al., 2017). Thus, the conclusion of these studies has highlighted the importance of including other factors that may impact spoken language development in children with HL (e.g. consistency in HA use and aided audibility). Accordingly, recent research has investigated hours of HA use and aided audibility (Moeller & Tomblin, 2015; McCreery et al., 2015a) which both have been demonstrated to vary greatly in the youngest age group (McCreery et al., 2015a; Muñoz & Hill, 2015; Walker et al., 2013) However, their impact on specific speech and language outcomes are less investigated (Moeller & Tomblin, 2015; Yoshinaga-Itano et al., 2017). Therefore, the impact of aided audibility and hours of HA use on early consonant use in babbling and early speech, consonant proficiency and expressive vocabulary is not clear, and in need of further investigations.

## 2.4 RATIONALE FOR THE THESIS

Since the implementation of UNHS, age at HA fitting in children has decreased. This has in international studies been found to have a positive impact on the speech and language development in children with HL albeit the population at large are still delayed in comparison to peers with NH (Ching et al., 2013; Moeller & Tomblin, 2015; Yoshinaga-Itano, 1999). Swedish research on outcomes of this kind in children with HAs under 3 years of age is limited (SBU, 2020). One reason for this may be the lack of appropriate assessments of children under 3 years of age, and that existing methods were previously not validated in Swedish. Therefore, a rationale for this study was to design and use a protocol including measurements at specific time periods that covered the ages from birth to 3years in the areas of auditory and early speech development. As there is little or no research outcomes of Swedish children on some of the measurements used in this project, a contemporary reference group of children with NH from the same geographical area was recruited for comparison.

# 3 AIMS

## 3.1 GENERAL AIM

The overall aim of this thesis was to investigate the auditory and early speech development in children with moderate HL amplified with HAs before the age of 6 months and to compare their development the first 3 years with a contemporary group of children with NH. As there is no national protocol for monitoring the auditory and early speech development in children with hearing loss from 0 to 3 years of, the purpose was to incorporate methods at specific target ages that have been demonstrated in international research to be valid and feasible to use in the monitoring of this target group. This also motivated the prospective and longitudinal design of this thesis.

## 3.2 SPECIFIC AIMS

The specific aims of this thesis were to:

- externally validate the Swedish version of the LittlEARS® Auditory Questionnaire of auditory development in a group of Swedish-speaking children with NH (Study I)
- investigate HA use in the children with HL and to examine the impact of hours of HA use on parental questionnaires of auditory development and functional auditory performance (Study II)
- examine the impact of auditory variables on consonant use in babbling and early speech in children with moderate HL and to compare their development to a group of children with NH (Study III)
- investigate and compare the expressive vocabulary in children with moderate HL in terms of number, growth and complexity to a group of children with NH. Other aims were to examine the impact of auditory variables and relationship between early consonant development to the number of target words produced (Study IV)

# 4 METHODS

## 4.1 PARTICIPANTS AND RECRUITMENT

The children with HL were recruited from the Hearing Habilitation Unit at Karolinska University Hospital. During the recruitment period for this project, 15 children with HL were eligible for inclusion of which parents of 14 children agreed to participate (2 girls, 12 boys). Three children with HL were excluded from the study before the age of 10 months due to improved hearing (n=1) and progressive HL resulting in CI (n=1) and suspected neurodevelopmental disorder (n=1). A reference group of children with NH were recruited from two child health care centres in the same geographical area (n=30, 15 girls, 15 boys). One child left the study at 13 months due to relocation. The common inclusionary criteria for both groups were that at least one of the caregivers needed to be a native speaker of Swedish and speak it to the child and no diagnosed syndrome known at the timepoint of recruitment. Demographic data of both groups are found in Table 1 and a flowchart over the participants for each study is found in Figure 3.

Children with HL were invited to take part in the study if they had been diagnosed with congenital bilateral sensorineural HL of moderate degree (41-60 dB HL), ICD-code H90.3 and been fitted with HAs before the age of 6 months. The first information about the project was given to the parents by a social worker, who asked for the permission of the the primary investigator (author of thesis) to contact them if interested. The primary investigator then called the parents on the phone and booked a meeting in person. The parents received both oral and written information and were given time to consider participation in the project. Date of study entry was the timepoint of received consent forms of both parents.

Children with NH were invited to take part if they had passed the UNHS at birth. To control for NH, the hearing of the children with NH was screened by a pediatric audiologist at 10, 18, 24 and 36 months of age (American National Standards Institute, 2010) at 20 dB HL at 500, 1000, 2000, 4000 Hz (MADSEN Astera2, GN Otometrics A/S Denmark). The same recruitment procedure and start of study entry was made for the children with NH, although it was a midwife at the health care centers who gave the first information about the project. Five of the participants with NH were recruited in form of a convenience selection (e.g. through colleagues and social network).

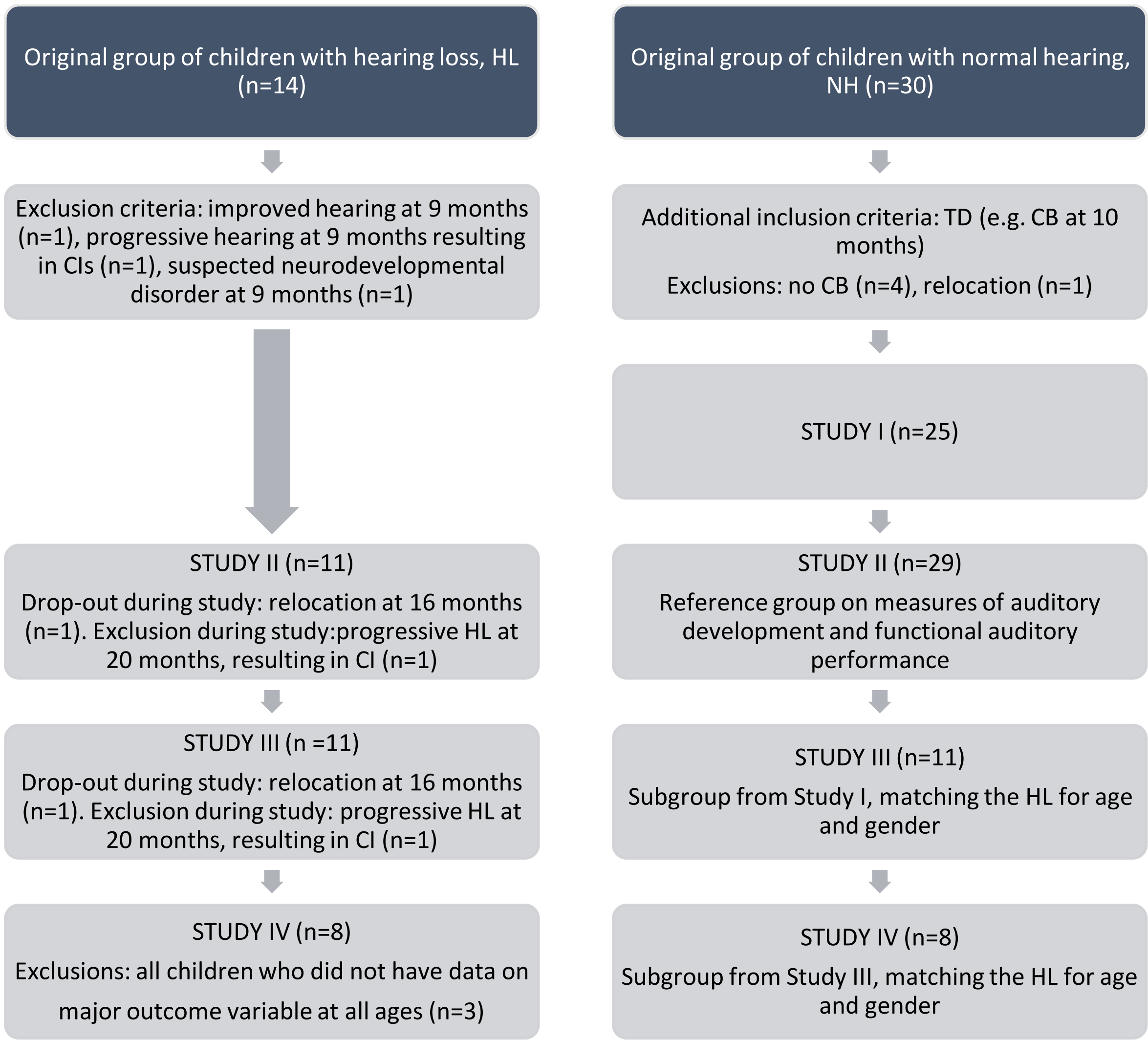
The educational level of the parents of both groups was higher compared to the mean of Swedish parents based on statistics that 50% of Swedish women and 38% of Swedish men have a university degree (Statistics Sweden, 2019). The number of mothers of the children with HL reported to have a university degree was 73% and fathers 45%. The corresponding number for the children with NH was 86% and 68%. The group mean was slightly lower for the parents of the children with HL but the difference was not significant according to Mann-Whitney U-test: mothers ($U$ (HL=11, NH = 29)=136.5, Z = -1.369, $p$ = 0.17); fathers ($U$ (HL=11, NH = 28)=120.0, Z = -1.576, $p$ = 0.12).

With regards to cognitive development, the difference in scores on the cognitive scale on the Bayley-III at 3 years of age was not significant between the groups: Mann-Whitney U-test: ($U$ (HL=8, NH = 29)=100.5, Z = -605, $p$ = 0.55).

**Table 1.** Demographic information of the children with hearing loss (CHL) and children with normal hearing (CNH) and their parents.

| | **CHL** | **CNH** |
|---|---|---|
| *Children (n)* | | |
| Girls | 1 | 15 |
| Boys | 10 | 14 |
| *Primary language in the home (n)* | | |
| Spoken Swedish | 11 | 29 |
| Additional language(s) | 0 | 6 |
| *Education level, mothers (n)* | | |
| Swedish elementary school (9 years) | 0 | 0 |
| Swedish college (12 years) | 3 | 3 |
| Higher education/University degree | 8 | 27 |
| *Education level, fathers (n)* | | |
| Swedish elementary school (9 years) | 1 | 0 |
| Swedish college (12 years) | 4 | 8 |
| Higher education/University degree | 6 | 20 |
| *Bayley-III score* | | |
| Cognitive scale *(mean, SD)* | 9.25 (1.98) | 10.10 (1.59) |

**Figure 3.** Flowchart of participants for each study



### 4.1.1 Study I

The children with NH were followed prospectively and longitudinally from timepoint of study entry (1-4 months) to 24 months. As one of the aims of this study was to investigate the construct of auditory development in this questionnaire in relation to a similar inventory of vocabulary development, one of the inclusionary criteria was for the children with NH to present with “typical language development”. In the absence of this kind of assessment, the presence/absence of CB at 10 months of age was used as it has been found to be a strong predictor of future speech and language development (Morgan & Wren, 2018; Oller et al., 1999; Stoel-Gammon, 2011). The assessment of CB was made through babbling observation which is explained in further detail in the Method section. The criteria of presence of CB led to the exclusion of four children, and as a fifth child relocated at 13 months, the final number of participants for this study was 25 children (Figure 3).

### 4.1.2 Study II

The children with HL were followed prospectively and longitudinally from the timepoint of first HA fit (2-6 months) to the age of 3 years. During the course of the study, five children were excluded due to improved hearing results/no HA use (n=1), progression in hearing resulting in CIs (n=2), relocation (n=1), and suspected neurodevelopmental disorder (n=1) (Figure 3). The mean age of HA fit of the remaining group of nine children was 4.6 months (range 3-6 months). Eight of the children were fit with Widex Baby 440, of which six changed to Oticon Sensei Pro 75 during the course of the study. One child was fit with Oticon Sensei Pro 75 and did not change HAs. The aetiology of the HL was unknown in seven of the participants, and two were confirmed as hereditary. All children used spoken Swedish as the primary language and two parents reported they used “some signs”.

### 4.1.3 Study III

In study III, the same nine participants with HL as in study II provided data at all timepoints. Additional data were included at 10 months of age for the child who relocated at 16 months, and data at 10 and 18 months but not 3 years for the child who received CIs at 20 months of age. The participants with HL were compared to a reference group of the children from study I (n=11), matching the children with HL for age and gender.

### 4.1.4 Study IV

In study IV, the participants were the same as in study III, with the exclusion of one child with HL who had missing data on the main variable outcome (expressive vocabulary) at two out of three data points. The final number of children with HL contributing with data at all data points were eight. This group was matched on age and gender to eight children with NH (n=8) from study III.

## 4.2 MATERIALS AND PROCEDURE

All four studies used a prospective longitudinal design with group comparison in study III and IV. A combination of standardised assessments performed onsite and parental questionnaires/inventories were used. Except from the hearing assessments and datalogging, the methods used in this project were not part of the regular clinical practice. The measurements and at what ages data were collected are found in Table 2.

All assessments were carried out at the Hearing Habilitation Unit, at Karolinska University hospital in facilities appropriate for the ambient test. All recordings in this project were made with high standard equipment (microphone Røde NT4, audio recorder TASCAM DR-22WL and video recorder Panasonic HC-V750). All parental questionnaires and inventories were given in paper format to be completed at home and returned in pre-stamped envelopes. Parents were offered reminders (email or text message) to complete and return to the primary investigator.

### 4.2.1 Hearing assessments

For the children with HL, behavioral audiograms at 6, 10, 18, 24, 30 and 36 months of age utilized behavioral observation audiometry, visual reinforcement audiometry, or conditioned play audiometry to obtain air and bone thresholds. Behavioral air and bone conduction thresholds were obtained at 250, 500, 1000, 2000 and 4000 Hz (MADSEN Astera2, GN Otometrics A/S Denmark) with insert earphones (Otometrics Oto insert ER-3A from Etymotic Research Inc.) and bone oscillator (Radioear B71). In cases where insert earphones could not be used, headphones were used (Radioear DD45, USA).

For the children with NH, the same equipment was used but they were screened at 20dB HL at the ages of 10, 18, 24 and 36 months. Due to resource constraints, screening could not be performed at 30 months. In cases where insert, nor headphones could be used, the screening was performed in a free-field setting not allowing for ear specific measures. Although assessment in free-field did not occur for any same child at repeated testings it cannot totally rule out the risk of a unilateral hearing loss for those children, albeit temporary.

**Table 2**. Included measurements and ages at data collection (marked with x). References to each of the measurements are found below the table.

| Purpose | Variable and measurement | 10 months | 18 months | 24 months | 30 months | 36 months |
|---|---|---|---|---|---|---|
| Hearing aid use and auditory functioning | Aided audibility<br>*SII* | x | x | x | x | x |
| | Hearing aid use<br>*Parental questionnaire of hours and situations*<br>*Datalogging of hours* | x<br>x | x<br>x | x<br>x | x<br>x | x<br>x |
| | Auditory development<br>*LEAQ** | x | x | x | | |
| | Functional Auditory Performance<br>*PEACH* | | x | x | x | x |
| Early speech development | Use of consonants<br>*Babbling observation* | x | x | | | |
| | Consonant proficiency<br>*SVANTE* | | | | | x |
| | Phonological complexity of words - *WCM-SE* | | x | x | x | |
| Communication and language development | Receptive language<br>*SECDI I - Words & Gestures* | x | | | | |
| | Expressive language<br>*SECDI II – Words & Sentences* | | x | x | x | |
| Cognitive development | Cognitive development<br>*Bayley – III* | | | | | x |

*The LEAQ was collected every other month from study start to 24 months of age in all participants.

Aided audibility: Speech Intelligibility Index (SII) (American National Standards Institute); Datalogging of hearing aids (Hearing Aid Manufacturer e.g. Oticon, Phonak, Widex); Parental questionnaire of hearing aid use (Created for this study); LEAQ = LittlEARS Auditory Questionnaire (Coninx et al., 2009; Khan, 2012); PEACH = Parents Evaluation of Aural/Oral Performance in Children (Ching & Hill, 2007; Brännström et al., 2014); Babbling observation (Lieberman & Lohmander, 2014; Lohmander et al., 2017a); SVANTE = SVenskt Artikulations- och NasalitetsTEst (Lohmander et al., 2017b); SECDI I & II= MacArthur Bates Communicative Developmental Inventory (Fenson et al.,1993; Eriksson & Berglund, 2000a; Berglund & Eriksson, 2000b); Bayley Scales of Development (Bayley, 2006; Stjernqvist et al., 2013); WCM-SE = Word Complexity Measure (Stoel-Gammon, 2010; Marklund et al., 2018).

### 4.2.2 Auditory variables

#### *4.2.2.1 Age at amplification and HA use*

Age at amplification and datalogging of the behind-the-ear HAs of each child were collected from their audiologist in the clinic. In a questionnaire created for this thesis, parents also reported number of HA use, scored on a scale divided in 3-hour brackets (e.g. 0-3 hours, 3-6 hours, 6-9 hours, and more than 9 hours). In addition to hours of use, parents were asked which situations and how often the HAs were used (e.g. play with parent, own play, play with other children, in public places, in the car, during mealtime, in preschool, TV/iPad/computer, with the alternatives “never, rarely, sometimes, often, always, don´t know”).

#### *4.2.2.2 Aided audibility*

Aided audibility was measured with the speech intelligibility index (SII). This measure ranges from 0 to 1, where 0 represents no audibility of the speech signal and 1 represents optimal audibility of the speech signal. SII was calculated with the Situational Hearing Aid Response Profile software (SHARP 1997, version 7) by entering the age and audiometric thresholds for each participant (air thresholds at 250, 500, 1000, 2000, 4000Hz) in the SHARP program. Aided SII was calculated for the participants, using the output of the HA for standard male speech signal (carrot passage) presented at 65dB SPL (average speech).

#### *4.2.2.3 Auditory development*

Depending on study entry, the parents of all children filled out the Swedish version of the LittlEARS® Auditory Questionnaire (LEAQ) in paper format every other month until their child turned 2 years. The LEAQ consist of 35 Y/N questions around responses to auditory stimuli (and speech). The original LEAQ had previously been back-to-back translated to Swedish (Persson & Rasmussen in cooperation with MED-EL, 2011) and evaluated by an audiologist student in 2012 (Khan, 2012).

#### *4.2.2.4 Functional auditory performance*

Parents of all children filled out the Swedish version of the Parent´s Evaluation of Aural/Oral Performance in Children (PEACH) every six months starting when the child was 18 months. The PEACH consists of 13 questions of functional auditory performance scored on a 4-point Likert scale. Functional auditory performance in children refers to how they hear and respond to sounds and speech in a variety of listening conditions. The 13 questions can also be divided into subscales of noise and quiet.

### 4.2.3 Methods for assessment of early speech development

#### *4.2.3.1 Measure for assessment of early consonant production in babbling*

A standardised observation of babbling and early consonant use was audio and video recorded by the author of this thesis when the children were 10 and 18 months old. Parents were instructed to play and interact with their child with a set of age-appropriate toys, positioned on a mattress. The about 45-minute long video recordings were later assessed by observation by a total of five experienced SLP´s according to a standardised manner (Lohmander et al. 2017a).

#### *4.2.3.2 Consonant proficiency measure*

Consonant proficiency was assessed by an experienced SLP with the Swedish Articulation and Nasality Test (SVANTE) (Lohmander et al., 2017b). Single word naming of 59 words with oral consonants and 5 with nasal were elicited by picture naming. Each target consonant is elicited in more than one word. The consonants are considered highly relevant in the development (six oral stops, three fricatives of which two are sibilants, and one nasal consonant) and are elicited in seven different words and positions (three initial, two medial, two final), except for /ɕ/ which is elicited in three words (initial only), and the nasal in five words (two initial, two medial and one final) (Lohmander et al., 2017b).

#### *4.2.3.3 Measure of size and growth of vocabulary development*

The Swedish version of the MacArthur-Bates Communicative Developmental Inventory (MCDI) – Words and Sentences (SECDI-II) was used to document expressive vocabulary development (Berglund & Eriksson, 2000b; Fenson et al., 1993). Parents were instructed to mark each item on the inventory they had heard their child produce spontaneously. The Words and Gestures (SECDI-I, receptive) (Eriksson & Berglund, 2000a; Fenson et al., 1993) was used in study I to compare the results of the scores on the LEAQ.

#### *4.2.3.4 Measure of word complexity*

The Swedish version of the Word Complexity Measure (WCM-SE; Marklund et al., 2018), originally developed for English by Stoel-Gammon (2010) was used to explore the complexity of the items that parents had reported on the SECDI-II. The WCM is a measure that calculates the phonological complexity of words or utterances based on a number of complexity parameters and is usually performed on live language samples.

### 4.2.4 Measure of typical cognitive development

To control for normal development, an assessment with Bayley Scales of Development (Bayley, 2006) was performed at 3 years by a paediatric psychologist. This assessment covered the areas of cognition, language (receptive and expressive), and motor skills (gross- and fine motor).

## 4.3 ANALYSIS

### 4.3.1 Measures of consonant use in babbling and consonant proficiency

Presence or absence of oral stops and dental / alveolar stops were collected from the assessment at 10 and 18 months. The consonants heard in at least two different occasions during the observation were noted and number of different true consonants counted for each participant. The same data was used for the decision of established consonants. In cases of uncertainty, a senior SLP with extensive experience with the method performed additional assessments. A SLP with more than 20 years of experience including extensive training in transcription, performed semi-narrow phonetic transcription of the SVANTE audio-video recordings using IPA and ExtIPA (IPA, 2008) of the consonants in SVANTE. The percentage of consonants correct adjusted for age (PCC-A) (Klintö et al., 2011) at 3 years, was calculated for each participant based on transcription of the target consonants. The proportion of children who had established the 10 target consonants /p, b, t, d, k, ɡ, f, s, ɕ, n/ were analyzed at the ages of 10, 18 months and 3 years. A consonant was defined to be established if heard in at least two productions at 10 and 18 months and at least 50% of the possible productions at 3 years.

### 4.3.3 Measure of phonological complexity score of words

For the exploration of calculating a complexity score of the items in SECDI-II, the onomatopoetic items were excluded as they are more prone to be irregularly produced resulting in large variations in score (Marklund et al., 2018). This resulted in a total number of possible words used for calculation in the SECDI-II to be 697, ranging in complexity scores of 0 to 12 (total complexity sum of all words = 2559, M = 3.6). The words were analyzed in respect to the dialect of the majority of people in the region where the participants were recruited. The items on the SECDI-II reported by the parents were analyzed based on the WCM-SE, and a complexity sum per word produced was calculated for each child.

## 4.4 RELIABILITY

For the babbling observations, carried out by a total of five SLPs, approximately 25% were re-assessed by a second SLP´s and in cases of substantial disagreement, a SLP and researcher with extensive experience performed the assessment. In terms of consonant proficiency (SVANTE) the SLP who assessed this data had an intra-transcriber reliability of 82% based on a material of over 100 3-year-olds. Twenty percent of the SVANTE data in this project was reassessed by a second trained transcriber with an exact inter-transcriber agreement, point by point of 85%.

In terms of the questionnaires and inventories, 60% of the SECDI inventories were cross-checked by a Master student in linguistics, 100% of the LEAQ questionnaires by a special education teacher (SET) experienced with the questionnaire, and 100% of the PEACH questionnaires by two clinical audiologists. No changes in scores were made for the SECDI, five LEAQ questionnaires were changed and one PEACH questionnaire was changed, each by one point.

## 4.5 STATISTICAL ANALYSIS

All data were analyzed in IBM SPSS Statistics Version 26 for Windows 18.0 software [Armonk, NY]. To adhere to best fit of the data, both parametric and non-parametric tests were used depending on the distribution of the data as well as due to the small study sample.

Study I: For study I a linear mixed model was used to generate a normative curve for the Swedish data and repeated measures ANOVA was used to investigate the effect of time (age). Pearson correlation coefficient was used to investigate the relationship between the LEAQ to SECDI and PEACH.

Study II: For study II, descriptive statistics were used to present a variety of data with regards to HA use. To describe the trajectory of HA use over time, within-subject linear regressions were made, and the mean of the individual slopes were calculated. Pearson correlation coefficient was also used to investigate the relationship between HA use to scores on auditory development and functional auditory performance.

Study III: Comparisons between the groups were performed with Fisher´s exact test. The number of different true consonants were calculated for each individual and compared at group level using Mann-Whitney U-test. PCC-A scores at 3 years of age were calculated and compared between the groups using Mann-Whitney U-test. Spearman´s rank correlations were used to investigate the relationship between the consonant variables to the auditory variables.

Study IV: Data in study IV is foremost presented in a descriptive manner (raw data and percentiles) for each group. Comparisons between groups were made through Mann-Whitney U tests. A two-way repeated measures ANOVA was performed with group as the between subject factor and age as the within-subjects factor to examine the growth in the number of words across ages. Spearman´s rank was used for correlation analysis between the different variables. The WCM-SE was used to calculate a mean complexity sum per word at each of the ages in all participants.

## 4.6 ETHICAL APPROVAL

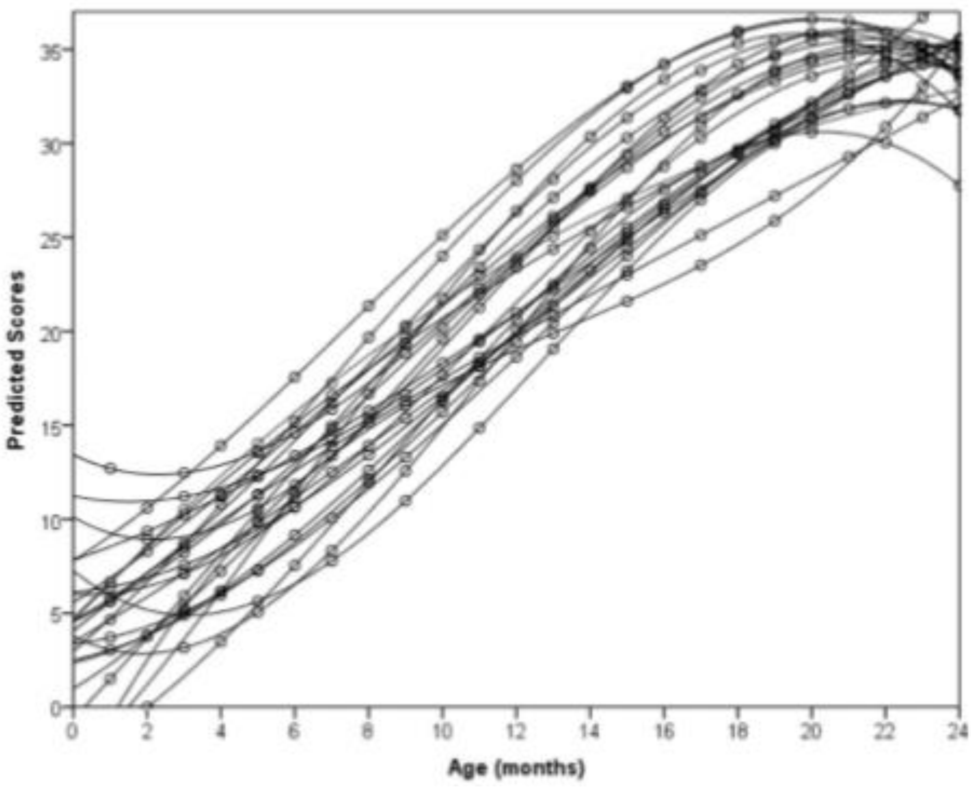
Ethical approval for the whole project was obtained from the Regional Ethical Review Board in Stockholm with Dnr.2014/1162-31/1. Information about the study was given orally as well as in writing and all parents signed a consent form. For studies II-IV an additional consent form was obtained from the parents of the children with HL to ask for permission to present individual data.

# 5 RESULTS

## 5.1 STUDY I

A total of 299 questionnaires from 25 children with NH were collected between the ages of 1 to 24 months. The individual growth curves increased with age for all children but two (at the ages of 16 and 20 months respectively). The mean LEAQ scores differed significantly between time points ($F(3.894, 93.467 = 368.304$, $p<0.0005$) and the Swedish normative curve was found to be similar to that of the original German version (Swedish model = 1.0026+2.1543*x-0.0298*x2, German version = 2.0651+2.2175*x-0.0376*x2). The association between timepoints within the LEAQ was strong and significant within the first 12 months ($r$=0.45 to 0.80, $p<0.01$) but weak and non-significant in relation to later timepoints of 12 to 24 months ($r$=0.023–0.39). A ceiling effect was indicated between 18-20 months of age. The correlations to SECDI were found moderate and significant at 10, 18 and 24 months and to PEACH at 18 months, but weak and non-existing at 24 months. The predicted development scores over time of each participant with NH (n=25) estimated with a mixed regression model is found in Figure 4.

**Figure 4**. Individual growth curves among the 25 participants with NH (linear regression mixed model). Reprinted with permission from *International Journal of Audiology*.



## 5.2 STUDY II

Hours of HA use according to datalogging varied greatly between the participants at all ages. The largest range in hours was found at 18 months (3-11.5 hours). The mean hours of HA use for the group increased with age from 7.5 hours per day at 10 months to 10.1 hours per day at 36 months (Figure 5). Parents reported that the most challenging age to maintain HA use was at 10 months and the most challenging situations were “meal-time”, “in the car”, “public places” and “own play”. The correlations between hours of HA use to parental ratings on auditory development (LEAQ) were weak at the corresponding ages but moderate between HA use at 10 months to LEAQ scores at 18 and 24 months. Hours of HA use at 18 months showed moderate to strong correlations to scores on the LEAQ at 24 months that were significant. Correlations between hours of HA use to functional auditory performance (PEACH) were all weak and non-significant. In terms of aided audibility (SII) the SII values did not show any relationship of significance to auditory development or functional auditory performance at any of the ages. However, the correlations between SII and functional auditory performance in noise showed higher correlations compared to the quiet and total scales (See Figure 6 for scores on SII over time). The children with HL (except P10 at 10 months) showed similar results on auditory development to the children with NH (Figure 7). The scores on auditory functional performance were also similar between the groups at all ages, albeit lower for the children with HL. When comparing the groups on the subscales in quiet and noise, the HL presented with lower scores on the noise scale at the ages of 30 and 36 months (see Figure 8 for results on the PEACH for both groups, and Table 3 for individual scores on the PEACH total score of the children with HL).

**Figure 5.** Average hours of HA use in the 11 children with HL over time according to datalogging measured at the ages of 10, 18, 24, 30 and 36 months. Note that P4 only has data at 10 and 18 months and P5 only has parent-reported hours at 10 months (9 hours/day).

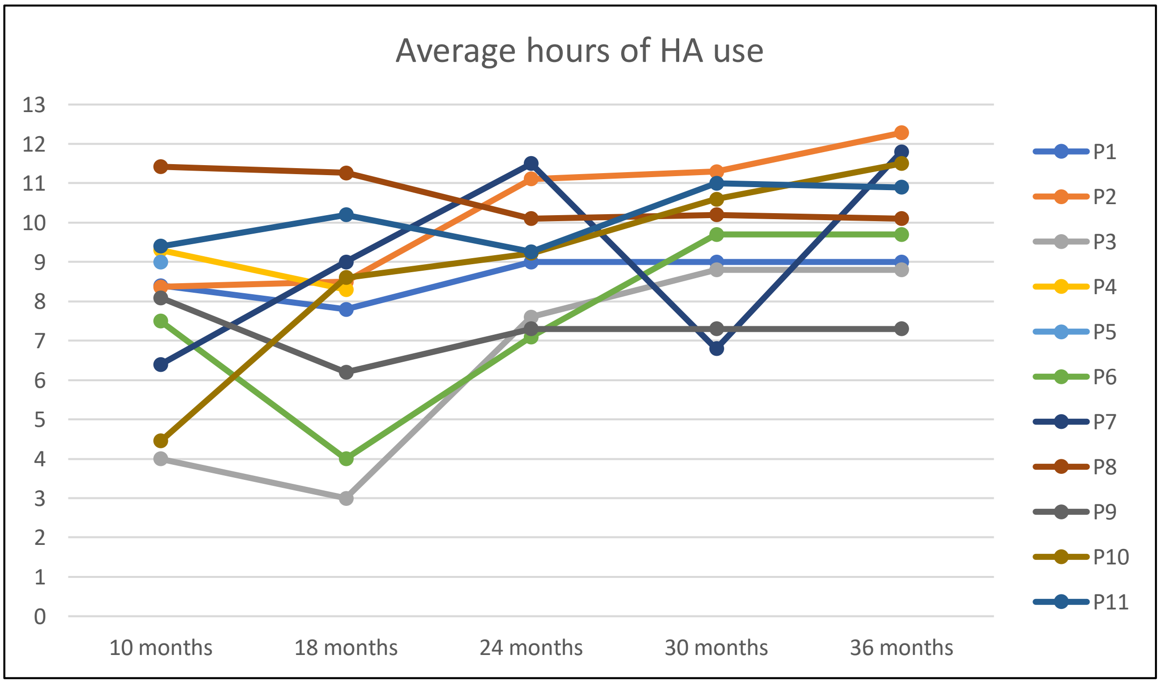


**Figure 6.** Speech Intelligibility Index values over time in 10 of the children with HL as SII values for P5 were missing. Note that P4 only has SII values at 10 and 18 months.

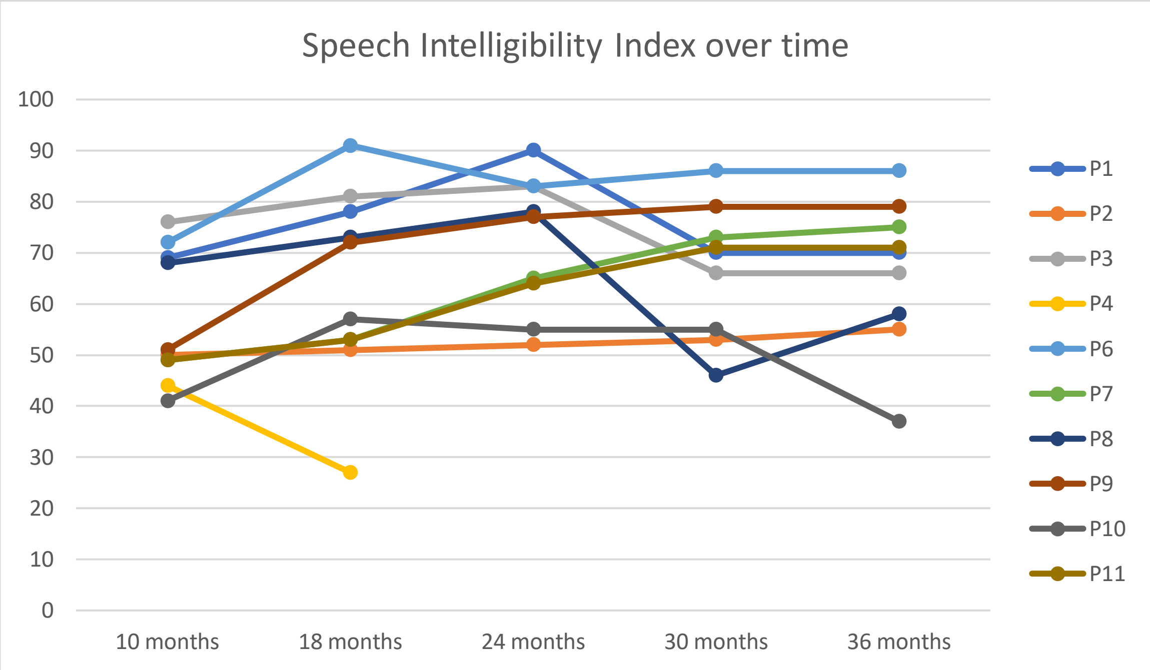



**Figure 7**. Mean LEAQ raw scores at 6, 10, 18 and 24 months of children with NH (n=29) and individual scores of the HL (n=11). Data is missing for P4 at 24 months, P5 at 18 and 24 months, and P10 at 6 months.

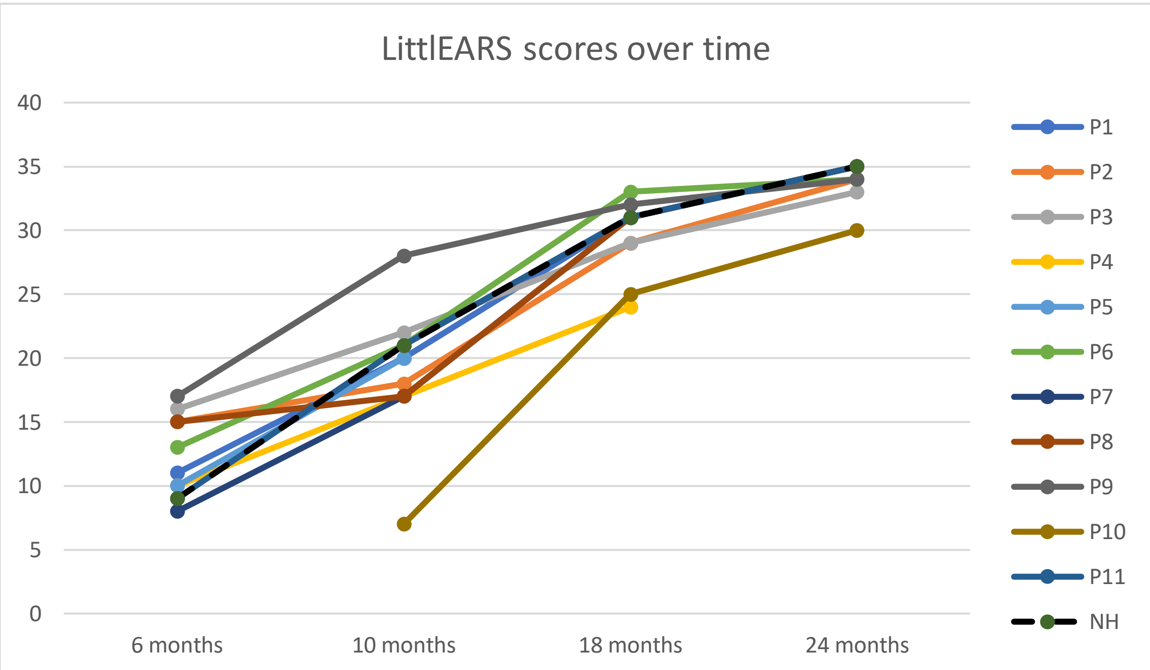


**Figure 8**. Boxplots of PEACH total scale (8a), noise subscale (8b) and quiet subscale (8c). X-axis represents the age in months and Y-axis the PEACH scores in percent. Children with HL in red (at 18 months, n=10, at 24, 30 months, n=8, 36 months n=9) and children with NH in green (at 18 and 24 months n=29, 30 months n=27, 36 months n=28).

**Figure 8a.** PEACH total score (noise and quiet subscales)

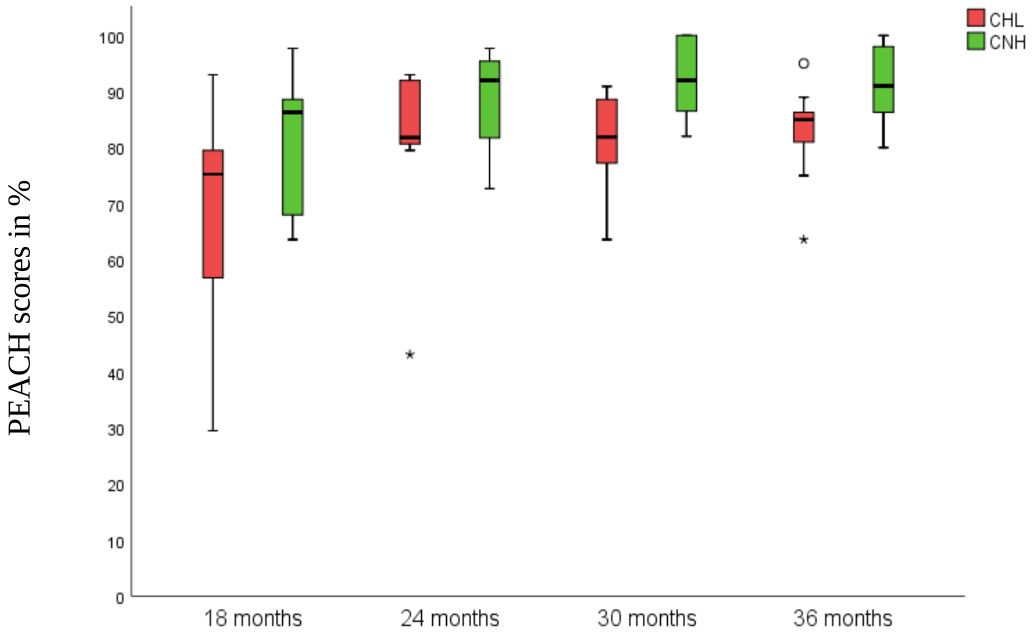



**Figure 8b.** PEACH noise subscale

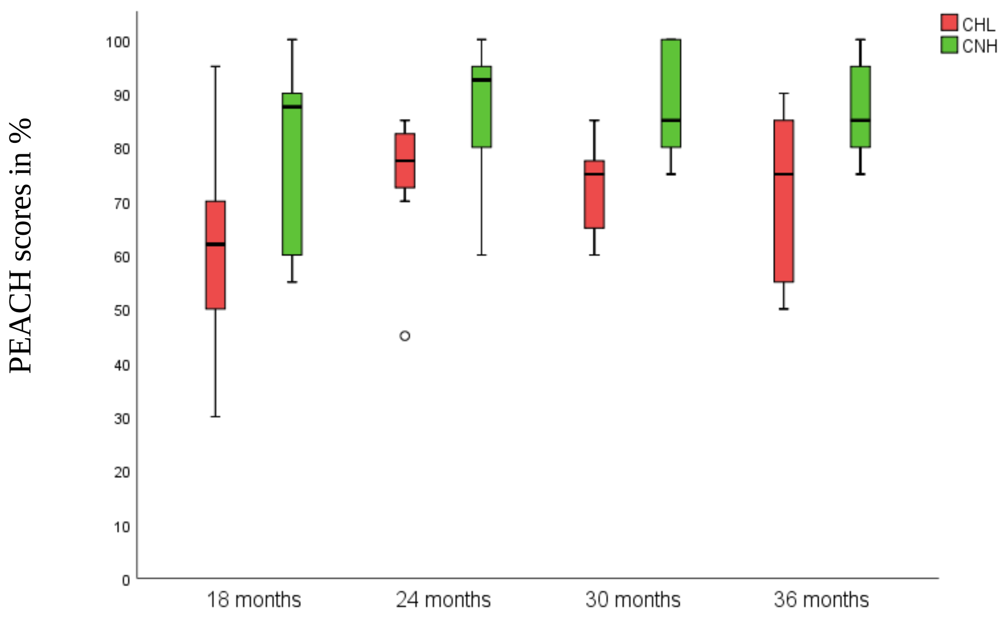
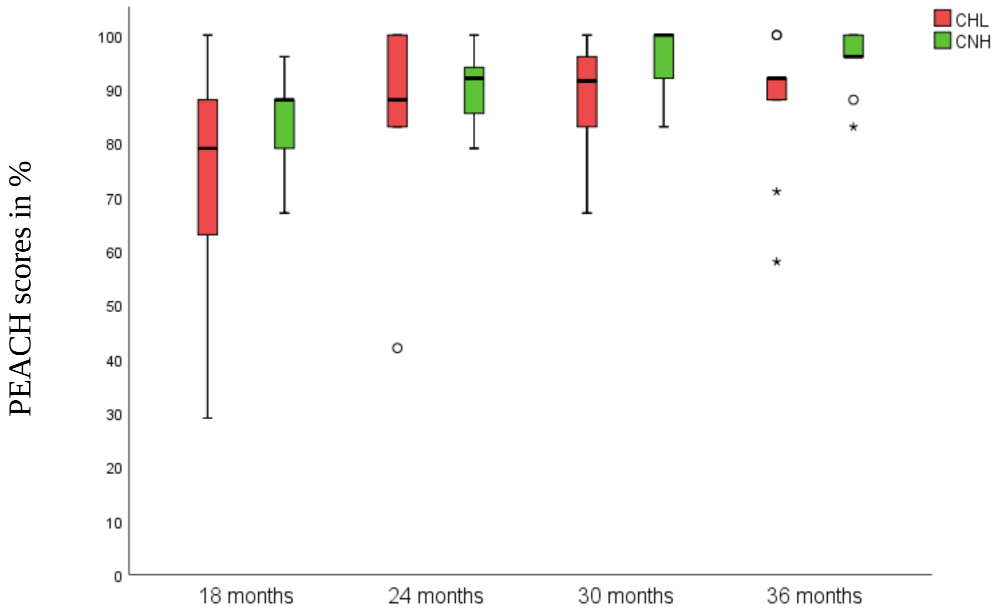


**Figure 8c**. PEACH quiet subscale




## 5.3 STUDY III

The difference in use of oral stops between the proportion children with HL and NH was significant at 10 months (45% vs. 100%), as was the use of and dental / alveolar stops (35% vs. 90%). At 18 months, the use of oral stops and dental / alveolar stops were still lower for the children with HL although the difference was not significant (oral stops 80% vs. 100%, dental / alveolar stops 70% vs. 100%) (Figure 9). In terms of number of different true consonants, the children with HL produced fewer true consonants compared to the children with NH at both 10 months, CHL (mean 0.5, range 0-2) and CNH (mean 1.3, range 1-3) (Mann-Whitney U: Z= -2.46, *p* = 0.01). At 18 months, CHL (mean 1.4, range 0-3) and CNH (mean 5.8, range 3-9) (Mann-Whitney U: Z= -3.73 *p* = 0.00) (Figure 10). The differences were significant and increased in strength at 18 months. The number of established consonants was substantially lower at 18 months in the children with HL, but at 3 years of age the differences in the establishment of the 10 target consonants included in the assessment (oral stops, three fricatives and one nasal consonant) were less than 10%. Consonant proficiency in terms of PCC at 3 years did not differ between the groups (Mann-Whitney-U: *Z*=-.41; *p* = 0.68). Hours of HA use at 10 months of age showed strong and significant correlations to the consonant variables at 10 months; oral stops (*rho* = 0.87, *p* = 0.01), dental / alveolar stops (*rho* = 0.81, *p* = 0.01), and number of different consonants (*rho* = 0.82, *p* = 0.01), and to PCC-A scores at 3 years (*rho* = 0.69, *p* = 0.04). Age at amplification and aided audibility showed weak to moderate correlations that were non-significant.

**Figure 9**. Percentage of children in each group using oral stops, and dental / alveolar stops at 10 and 18 months of age. At 10 months: CHL and CNH (n=11) respectively and at 18 months: CHL and CNH (n=10) respectively. P-values from Fischer´s exact test (*p < .01, **p < .05). Data from study III (in manuscript).

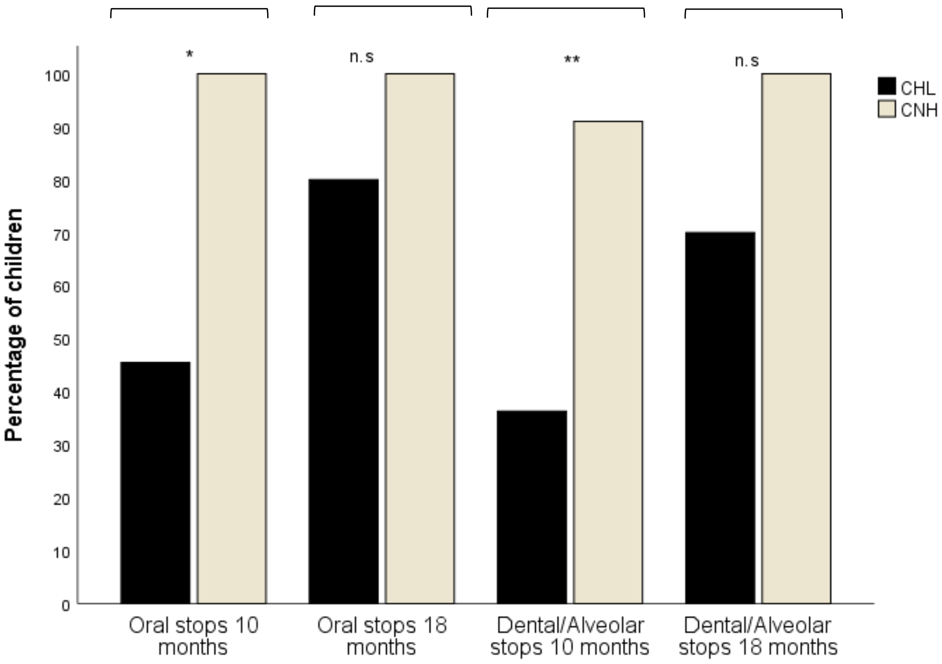
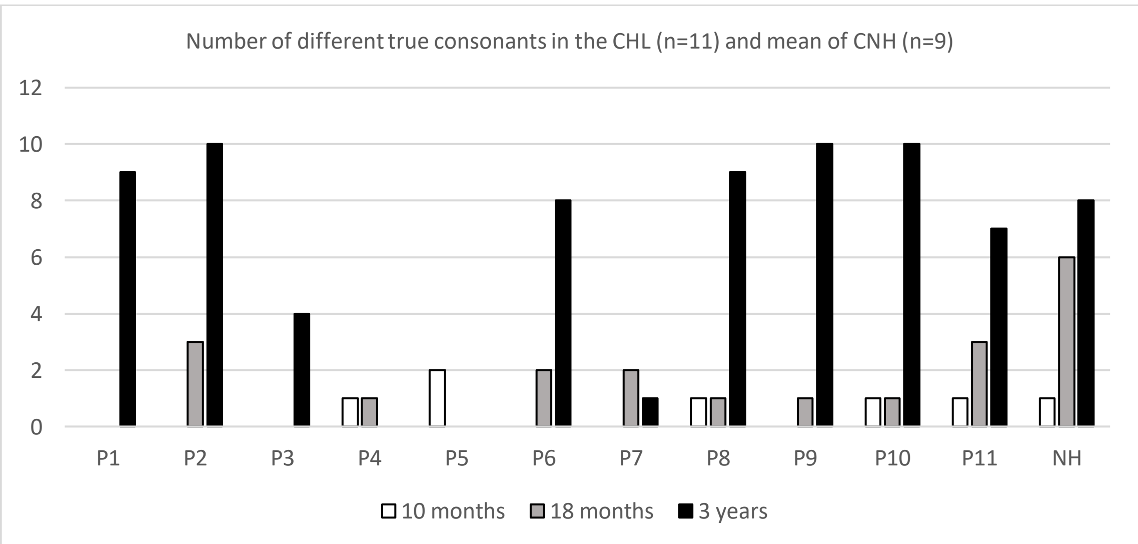


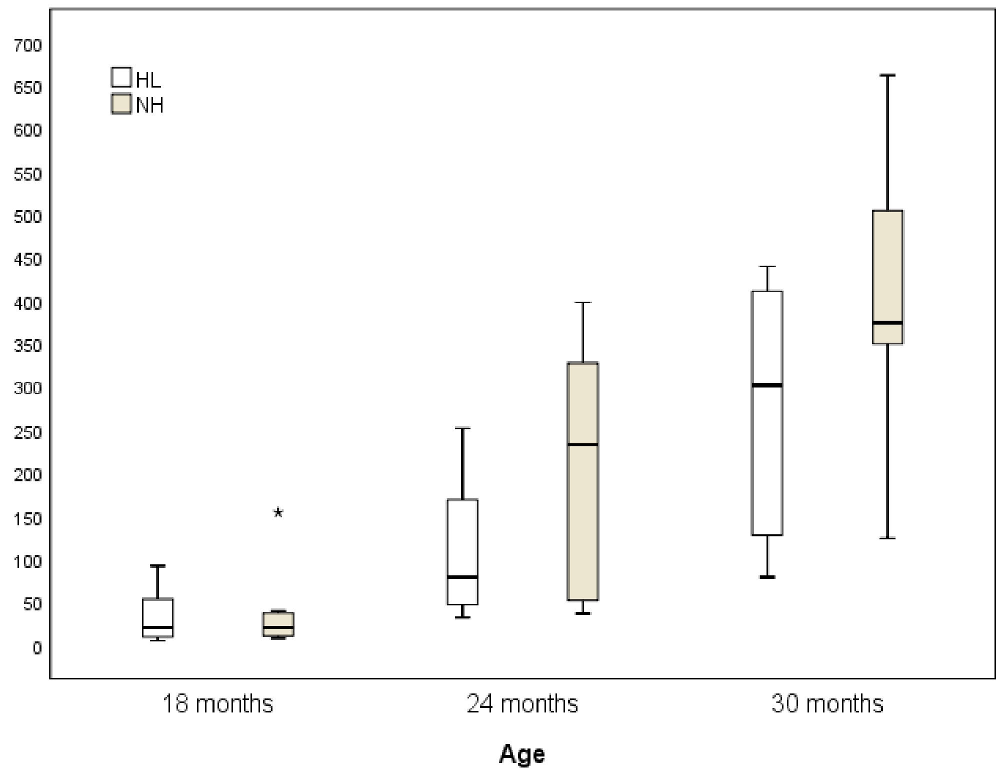

**Figure 10**. Number of different true consonants at 10 and 18 months and number of established consonants at 3 years (out of the 10 target consonants) in the children with HL (P1-11) and the corresponding mean numbers of the group of children with NH (n=11). The median number of different true consonants of the NH group are adjusted to fit the figure. Data is missing for P4 at the age of 3 years and for P5 at 18 months and 3 years, others being "0". Data from study III (in manuscript).



### 5.4 STUDY IV

The number of reported "words produced" demonstrated with large ranges that increased per age, smallest found at the age of 18 months and largest at the age of 30 months, specifically in the children with NH (n=8). At 18 months, the median number of words were similar between the groups (*Median* HL = 20.5, NH = 20.5) but differed at the ages of 24 months (*Median* HL = 79, NH = 272) and at 30 months (*Median* HL = 302, NH = 375) (Figure 11). All children in both groups increased their number of words with age except for one child with NH between 24 to 30 months (albeit both scores were at the 70$^{th}$ percentile). For individual number of words produced in the children with HL at each of the ages, see Table 3.

**Figure 11**. Boxplots of number of produced words per group for children with HL and NH at the ages of 18, 24 and 30 months. The difference between the groups was not significant at any age (18 months: $U$ (HL = 8, NH = 8)=30.5, z = -.158, $p$ = 0.87, and at 24 and 30 months: $U$ (HL = 8, NH = 8) =19.0, z = -1.365, $p$ = 0.17). Data from study IV (in manuscript).




The growth curve of the children with NH was almost identical to a reference group from a databank of Swedish children (n=420, all boys) at all ages but lower for the children with HL at 24 and 30 months. A two-way repeated-measures ANOVA of the children with HL (n=8) and NH (n=8), with group as the between-subjects factor and age as the within subject factor revealed a significant increase in number of expressive words with age, $F$ (1, 74) = 109,679, $p$ = 0.00 (Figure 12). More children with HL performed at or below the 10$^{th}$ percentile at all ages. The correlations between number of words to complexity sum of all words was significant in both groups at all ages. The auditory variables with strongest correlations to number of produced words were hours of HA use at 10 and 30 months and scores on auditory behavior at all ages. There was a strong moderate and statistically significant correlation in the whole group between number of different true consonants at 18 months and number of produced words at 24 months.

**Figure 12**. Mean number of produced words on the SECDI-II of the children with HL (n=8), children with NH (n=8) and a reference group of Swedish children from the Wordbank (boys, n=420) at 18, 24, and 30 months. Data from study IV (in manuscript).

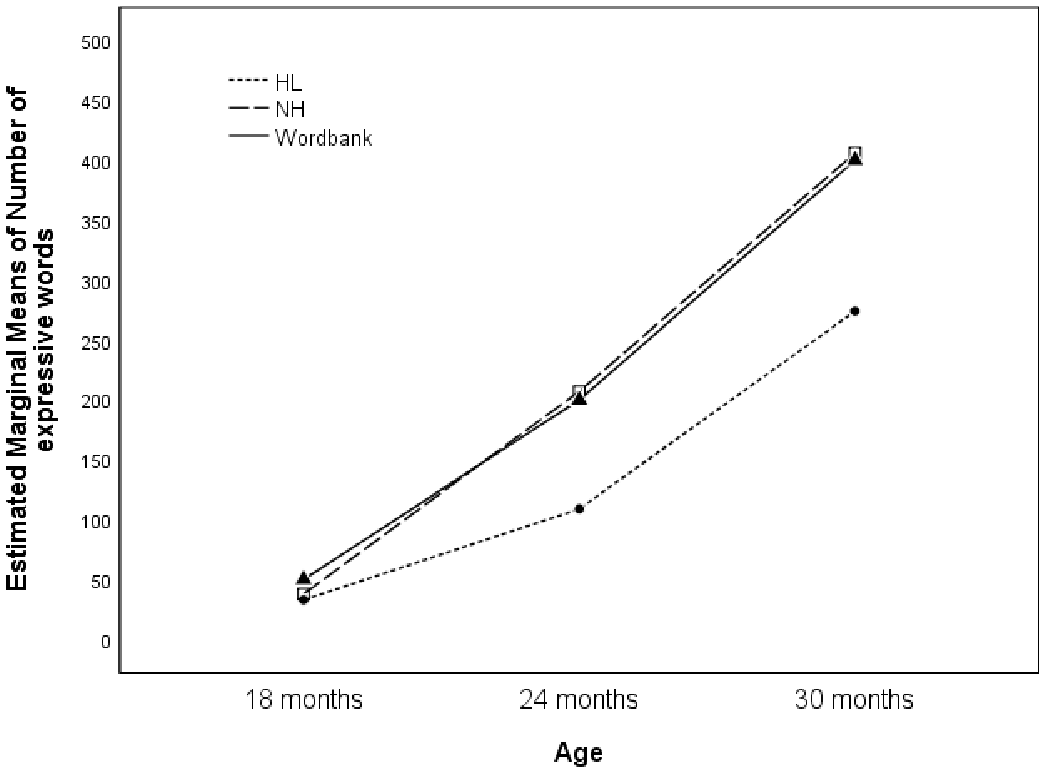


**Table 3**. Descriptive data of individual children with hearing loss (n=11). Unaided PTA: Better-ear four-frequency pure-tone average (250, 500, 1000, 2000 and 4000 Hz), mean dB HL over time measured at 10, 18, 24, 30 and 36 months. Hearing aid (HA) use was collected from datalogging of the HAs at the ages of 10, 18, 24, 30 and 36 months. Data of aided audibility (SII value) was calculated at the same ages. N/A means not applicable due to missing data. Data of SII at 10 months and all data at the remaining ages is missing for P5 as the child left the project at 16 months of age due to relocation. Data is also missing for P4 at 2, 2.5 and 3 years due to progressive HL at 20 months, resulting in CIs. P6 did not complete the Bayley Scales testing at 3 years. LittlEARS Auditory Questionnaire (LEAQ), Parent´s Evaluation of Aural/Oral Performance in Children (PEACH). Number of different true consonants assessed from Babbling observations (Lieberman & Lohmander, 2014; Lohmander et al., 2017). Number of words from parental inventory SECDI- II (Berglund & Eriksson, 2000a). Parental education (1= elementary school, 2= college, 3= university). Data on cognitive and expressive language scales are from Bayley Scales of Development at 3 years (Bayley, 2006).

| | **P1** | **P2** | **P3** | **P4** | **P5** | **P6** | **P7** | **P8** | **P9** | **P10** | **P11** |
|---|---|---|---|---|---|---|---|---|---|---|---|
| Gender (Boy/Girl) | B | B | B | B | B | B | B | B | G | B | B |
| Etiology | N/A | N/A | Hereditary | N/A | N/A | N/A | N/A | N/A | Hereditary | N/A | N/A |
| Age at diagnosis (months) | 2 | 2 | 1 | 2 | 3 | 3 | 3 | 2 | 2 | 2 | 4 |
| Better ear mean 4F PTA (dB HL)* | 44.8 | 48.1 | 44.6 | 50.5 | 45.0 | 37.3 | 43.5 | 40.0 | 44.8 | 58.8 | 44.8 |
| Age at HA fitting (months) | 3 | 4 | 3 | 2 | 4 | 5 | 4 | 5 | 3 | 6 | 5 |
| Average hours of HA use/day (mean over time) | 8.6 | 10.3 | 6.5 | 8.8 | 9.0** | 7.6 | 9.1 | 10.6 | 7.2 | 8.8 | 10.1 |
| Aided audibility (mean over time) | 75 | 52 | 74 | 35 | N/A | 83 | 63 | 65 | 72 | 49 | 62 |
| LEAQ score - *10 months* | 20 | 18 | 21 | 17 | 20 | 21 | 17 | 17 | 28 | 7 | 21 |
| *18 months* | 31 | 29 | 29 | 24 | N/A | 33 | 30 | 31 | 25 | 25 | 32 |
| *24 months* | 35 | 34 | 33 | N/A | N/A | 33 | 34 | 35 | 34 | 28 | 35 |

| | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| PEACH score - *18 months* | 77% | 80% | 64% | 30% | N/A | 93% | 39% | 75% | 76% | 57% | 86% |
| *24 months* | 82% | 80% | 82% | N/A | N/A | 93% | 43% | 93% | 82% | 91% | 84% |
| *30 months* | 80% | 91% | 82% | N/A | N/A | 89% | 64% | 82% | N/A | 75% | 89% |
| *36 months* | 82% | 86% | 89% | N/A | N/A | 86% | 64% | 84% | 95% | 100% | 75% |
| Oral stops (0 or 1)*** - *10 months* | 0 | 0 | 0 | 1 | 1 | 0 | 0 | 1 | 0 | 1 | 1 |
| Oral stops (0 or 1)*** - *18 months* | 1 | 1 | 0 | 1 | N/A | 1 | 1 | 1 | 1 | 1 | 1 |
| Dental /Alveolar stops (0 or 1)*** – *10 months* | 0 | 0 | 0 | 1 | 1 | 0 | 0 | 1 | 0 | 0 | 1 |
| Dental /Alveolar stops (0 or 1)*** – *18 months* | 0 | 0 | 0 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 |
| No of different true cons – *10 months* | 0 | 0 | 0 | 1 | 2 | 0 | 0 | 1 | 0 | 1 | 1 |
| No of different true cons – *18 months* | 0 | 3 | 0 | 1 | N/A | 2 | 2 | 1 | 1 | 1 | 3 |
| PCC-A (3 years) | 98% | 95% | 48% | N/A | N/A | 87% | 37% | 100% | 95% | 97% | 77% |
| Number of words - *18 months* | 83 | 4 | 15 | 6 | N/A | 26 | 6 | 10 | 275 | 5 | 68 |
| Number of words - *24 months* | 241 | 140 | 26 | N/A | N/A | 66 | 34 | 41 | N/A | 77 | 177 |
| Number of words - *30 months* | 398 | 404 | 69 | N/A | N/A | 152 | 87 | 310 | N/A | 277 | 431 |
| Parental education - *Mother* | 2 | 3 | 2 | 3 | 3 | 2 | 3 | 3 | 3 | 3 | 3 |
| Parental education - *Father* | 2 | 3 | 1 | 2 | 3 | 2 | 3 | 3 | 2 | 3 | 3 |
| *Cognitive scale (3 years)* | 10 | 10 | 7 | N/A | N/A | N/A | 7 | 11 | 10 | 7 | 12 |
| *Expressive language (3 years)* | 23 | 23 | 12 | N/A | N/A | N/A | 16 | 28 | 21 | 18 | 24 |

*Note:* *None of the children except P6 differed more than 5 dB HL in hearing threshold between the ears (P6 right 46.0, left 37.3) **Based on parental report (scoring range 9-12 hours) at 10 months ***0/1 = absence/presence

# 6 DISCUSSION

The overall aim of this thesis was to investigate the early auditory and speech development in children born with moderate HL and to compare their outcomes to a reference group of children with NH. Specific aims were to examine the impact of auditory variables on the early speech development in the children with HL and to validate a questionnaire of auditory development in the children with NH. As there is no national protocol for monitoring the early auditory and speech development the first three years, the measurements used in this project were inspired by international large-scale research of the paediatric population with HL using HAs (Bagatto et al., 2011a; Ching et al., 2013; Moeller & Tomblin, 2015).

## 6.1 HEARING AID USE AND AIDED AUDIBILITY

### 6.1.1 Hearing aid use

The children with HL in this study were fitted with HAs at or before the age of 6 months, which was in line with the prevailing recommendations at the timepoint this study began (e.g. "1-3-6") (JCIH, 2007). They also showed large variation in their hours of HA use, which has been reported in several studies (Moeller et al., 2009; Muñoz et al., 2015; Walker et al., 2013). However, the mean hours of HA use in this Swedish group of children was around 3 hours higher than previously reported in children under 2 years of age (Walker et al., 2013). In contrast to previous studies of HA use in infants and toddlers, this study reported hours collected from datalogging as opposed to parent-reported hours. When comparing parent-reported hours to hours from datalogging, parents have been found to overestimate the hours of HA use (Moeller et al., 2009; Walker et al., 2015). Furthermore, previous research has demonstrated that children with mild-to-moderate HL demonstrate a more inconsistent HA use (when compared to children with profound HL). Therefore, the results regarding hours of HA use from this Swedish group of children are indicating a positive trend. A possible reason for this outcome may be due to study bias (albeit positive), as the parents of the participating children performed additional questionnaires around own estimations of HA use and situations over a long period of time. Thus, it would have been interesting to compare these results of hours of HA use with a group of children who followed regular practice.

Despite the overall higher hours of HA use, there were only two children who wore their HAs full-time (based on 9 hours/day) (Paruthi et al., 2016) at all ages (Figure 5). This indicates that there are improvements to be made in the intervention around HA use in the children under 3 years of age. Motivation around consistent HA use should be a main priority as hours of HA use was found to be the most prominent auditory variable showing the strongest correlations with significance on several of the investigated speech variables. Albeit the small study sample, another finding in line with previous research on larger groups of children was that the participants in this study with the lowest HA use were children of parents with the lowest educational levels (Marnane & Ching, 2016; Walker et al., 2015). As level of parental education has been a factor that can be overcome with intervention with regards to vocabulary development, there is ample belief that this could be true for intervention to increase HA use as well.

### 6.1.2 Aided audibility

Aided audibility provided by the HAs have been found to be associated with better speech and language scores in older children (Tomblin et al., 2014) but specific effects of amplification are rarely described in studies of speech and language outcomes of children under the age of 3 years (McCreery et al., 2015a). This motivated the exploration of the relationship between aided audibility to consonant use and expressive vocabulary in this cohort of children.

Measuring aided audibility is important, but complex in nature (McCreery & Walker, 2017). As research on the impact of aided audibility on speech and language outcomes in infants and toddlers is sparse (Moeller & Tomblin, 2015) the results from the SII measurements in this project were challenging to interpret. However, in a study by McCreery et al. (2015a) where SII was controlled for, they found that approximately 35% of the values of 288 children (age range 5-110 months) with mild to severe HL were below the average SII value according to normative values based on degree of HL (Bagatto et al., 2011a). In terms of impact on speech outcomes, the findings from the SII measurements in the current PhD project showed correlations from non-existing to moderate that in main part were non-significant. The main reason for this these low correlations may to a large part be due to the small number of participants. Therefore, the non-significant correlations do not have to imply that the aided audibility may not have affected the early speech development of individual children. For example, two of the three children with SII values within normative values at all ages were also those with the lowest number of hours of HA use. Although not as prominent as the low hours of HA use at 10 months, these factors in combination may have contributed to the low presence of oral stops and dental / alveolar stops found at 10 months. The lowest mean value of SII for the whole group was found at 10 months when less than 50% of the children had values within expected norms (Figure 6). Furthermore, it may be that aided audibility is less sensitive to measures of speech in children under 3 years of age as the effect of this measure has been found to increase as children´s duration of HA use increase (Tomblin et al., 2014). Although the children may have been underfit, their auditory development was within normal limits, as measured by the LEAQ (Coninx et al., 2009; Persson et al., 2019). The number of children with SII values within expected values increased with age, as did average hours of HA use which may have affected the increase in number of established consonants, that will be described in section 6.3.1.

It should be noted that the SII measurement is dependent on hearing thresholds and achieving reliable responses from infants and toddlers is challenging (Katz et al., 2009). Accordingly, SII values derived from estimated thresholds have been found to result in lower values. This was also the case in this study for three children at the age of 10 months. All children showed stable hearing thresholds across all ages except the child who progressed and received first CI at the age of 26 months (P4). This child also had the lowest SII value, which is in line with normative values of children with more severe losses (Tomblin et al., 2014). The SII values were also lower for the child with the highest thresholds (P10). Due to the small number of participants, the findings of the measures of aided audibility from this study are not able to state what impact of significance they may have on the early speech variables in general. However, the findings indicate that the levels of aided audibility may have been “good enough” and that hours of HA use may be more important in children with moderate degree of HL. In terms of clinical implementation, the measurements of aided audibility were found to be clinically feasible and the findings from this study motivate data collection on a larger number of children to gain more knowledge on the effect aided audibility may have on early speech development.

## 6.2 VALIDATION OF THE LITTLEARS AUDITORY QUESTIONNAIRE AND OUTCOMES ON FUNCTIONAL ASSESSMENTS

### 6.2.1 Auditory development

One of the aims with this thesis was to externally validate the Swedish version of the LEAQ which is a questionnaire that monitors auditory development the first 2 years (Coninx et al., 2009). If considered to be valid, the LEAQ could evaluate auditory development in the youngest age group and complement the Swedish version of the PEACH (Brännström et al., 2014) to aid in the validation process of the HA fitting. The LEAQ and the PEACH are two of the highest ranked functional assessments (Bagatto et al., 2011b) and used in recommended protocols of audiological monitoring in the paediatric population (Bagatto et al., 2011a). The following paragraph will discuss the findings from the validation of the Swedish LEAQ as well as the outcomes on this measure of both groups of children.

The many previous external validations of LEAQ translations have evaluated the psychometric properties of the LEAQ with a cross-sectional design. The goal of this study was to contribute to the existing research with a longitudinal design. The result that the children with NH increased their LEAQ scores with age was in line with previous studies (Coninx et al., 2009; García-Negro et al., 2016; Geal-Dor et al., 2011). The findings from study II that the children with HL presented with similar outcomes as the NH was more surprising, but only to a certain extent as other studies have questioned the sensitivity of the LEAQ for children with milder HL (Bagatto et al., 2016; Ganek et al., 2020). This may also be partly explained by the fact that the LEAQ was created in 2003 intended to be used in the follow-up of children aged 0-2 years with severe to profound HL who receive CIs (Coninx et al., 2003). Accordingly, the recommended use of this tool for children with similar degree of HL as the current study group with moderate HL would be to calculate the scores based on chronological age instead of “hearing age” (e.g. the age from timepoint of first HA fit to test date). Comparing results based on chronological age is also relevant as the majority of children with moderate HL go to mainstream preschool and will be compared to children with NH. In evaluations of audiological measures for the youngest age group, the LEAQ has received high ratings for meeting the accepted standards in terms of conceptual clarity, convergent validity, respondent and administrative burden, and test retest (Bagatto et al., 2011b). These ratings were also confirmed in the current validation by the increase in scores between age months and the compliance with returning the questionnaires over time. Thus, the LEAQ has been found to be lower with regards to discriminant validity. Therefore, the LEAQ was investigated in relationship to other questionnaires hypothesized to be similar in content. The correlations between the Swedish LEAQ and the Swedish CDI were moderate to strong and significant (Persson et al., 2019). These findings question the construct validity of the LEAQ as the content seem similar to a measure of vocabulary. Accordingly, the derived score from the LEAQ should be interpreted with the awareness of the emphasis on language development rather than taking the score as a marker of auditory development, the second half in particular. Furthermore, by face validity, there are few questions around discrimination which would be of interest if the goal is to monitor auditory development. On the other hand, as it is measuring auditory development but also early language skills, incoherent scoring of individual children may be an early indicator of communicative and language delays. The LEAQ may therefore be useful in discussions in multidisciplinary teams. In sum, the LEAQ is an acknowledged tool and the Swedish version can now be used to monitor the early auditory and communicative/language development the first 2 years. The LEAQ is clinically feasible, with ample possibilities of comparing data of children with all degrees of HL.

### 6.2.2 Functional auditory performance

The children with HL as a group presented with lower scores on functional auditory performance as measured by the PEACH at all ages compared to the children with NH. This adds to previous research on parental ratings with the same instrument on larger groups of children (Ching et al., 2015; McCreery et al., 2015b). However, the only significant difference between the groups was on the noise subscale at the ages of 2,5 and 3 years. The reasons for these results may be several. The first reason may be due to the small number of participants with large variability in outcomes at 18 and 24 months. However, the large variation found was quite surprising as this cohort of children were homogeneous in terms of degree of HL and close in age of HA fitting. On the other hand, the PEACH scoring is situational, and could be affected by in what situation the parents scored their child´s functional performance. Consequently, another reason may be bias in scoring by the parents of the two groups. Parents of children with HL are likely to be affected by the awareness that children with HL (in general) has limited hearing. Therefore, a future recommendation in the use of this instrument could be to instruct the parents to score their child in two different occasions, e.g. one with HAs on, and one without. Another improvement for future studies would be to have the parents report on which of them who filled out the questionnaire as parents may score differently. Individual differences (in both groups of children) may also be due to variations in the children´s attention abilities, as the scoring of auditory performance is based on observed behaviours of the child in real-world settings.

The findings that the children with HL in this project had lower scores on the noise subscale indicate that they to some degree may be facing challenges to listen in noise. Despite early amplification and seemingly sufficient levels of aided audibility, it can still not make up for the consequences posed by the sensorineural HL (McCreery et al., 2015b). Preschool is one of the places where Swedish children spend many hours, which demonstrated to be noisy (Persson Waye et al., 2018). In Sweden, more than 90% of children are enrolled in mainstream preschools by the age of 18 months, and today children with HL are no exception. All children with HL in this study had started mainstream preschools by the age of 18 months. Therefore, information of the child´s functional performance in different environments could complement the observations made in the clinic. The creators of the PEACH have made a teacher version of this instrument called the Teachers Evaluation of Aural/Oral Performance in Children (TEACH) (Ching & Hill, 2007). Swedish experience with the TEACH has been found to be a good complement to the PEACH as the functional performance in children tended to be scored differently between parents and teachers (Trygg, 2015). The most likely reason for this was found to be due to the different listening environments at home and in the preschools/schools (Trygg, 2015).

The PEACH scores did not show any significant relationship to aided audibility at any of the ages. It may have been that this group of children with moderate HAs had sufficient auditory gain to demonstrate functional performance similar to the children with NH. Thus, considering that none of the HAs of the children in this study were using advanced features like telecoil, motivates a discussion around how listening in noise may be alleviated for the youngest age group of children with HL. The advancements in technology are constantly moving forward. With regards to the escalating risk of not being able to perceive speech and acquire new words, introducing remote microphone technology in addition to teaching appropriate strategies of talking at close distance could be beneficial for individual children with moderate HL. The experience with the PEACH in this project is that it is clinically feasible and useful in the individual monitoring of children with moderate HL. Recurrent use of functional measures like the LEAQ and the PEACH may raise the awareness of parents to their child´s prerequisites when it comes to listening in different environments. As the PEACH is based on parental observations of their child´s functional auditory performance, it may also capture additional information around perceptual abilities that may affect overall learning.

## 6.3 EARLY SPEECH DEVELOPMENT THE FIRST THREE YEARS IN CHILDREN WITH HEARING LOSS

### 6.3.1 Early consonant production and use of early speech

Despite early fitting with HAs the children with moderate HL in this study showed delays in their consonant production in babbling and early speech compared to the children with NH at 10 and 18 months. This is consistent with previous findings in children with severe HL (Moeller et al., 2007a; 2007b; Stoel-Gammon, 1988). However, on a positive note, the difference on important precursors (e.g. oral stops and dental / alveolar stops) became non-significant at 18 months. This indicates that the children with HL were able to “narrow the gap” at an earlier age compared to previous studies (Ambrose et al., 2014; Sininger et al., 2010). Thus, although there was an increase in the presence of oral stops and dental / alveolar stops from 10 to 18 months, the difference in number of different true consonants to the children with NH was still present at 18 months. This may have affected the following development of establishing consonants. At 3 years, however, the children with HL showed similar consonant proficiency (PCC-A) as the children with NH. This was also a positive finding as the restraints in auditory perception may result in a degraded speech signal, affecting the auditory feedback loop that is needed for monitoring of articulation skills (Stoel-Gammon, 2011). Challenges with articulation in children with HL have previously been found in children with profound HL (Stoel-Gammon, 1988) and in older (Swedish) children with HL (Borg et al., 2007). The assessments on consonant proficiency in this project were analysed with an approach that adjusted for age in terms of consonant production. Based on all the assessments on consonant proficiency with the SVANTE (HL n=9, NH n=29) the number of elicited words on the SVANTE test were similar between the groups, albeit the quality of productions of full words were by face validity of the transcriptions more accurate for the children with NH. The combination of results and reflections on overall data on consonant proficiency, motivates monitoring at later ages as a similar score to the children with NH at 3 years of age does not ensure that later outcomes are as typical.

The investigated consonant variables are the ones considered relevant and important to predict later speech and language skills in TD children (Stoel-Gammon, 2011; Vihman, 2014). In this project, these predictive variables at 10 and 18 months did not show any strong correlations to consonant proficiency at 3 years for the children with HL, and not to outcomes of expressive vocabulary either. Interestingly, when the children with NH were included in the analysis this led to significant correlations. One reason for the lack of association in the group with HL may have been the small number of participants. Another reason may be that the predictive variables are not as sensitive in children with HL as there are confounding factors affecting their speech development. Accordingly, hours of HA use showed strong correlations to early consonant use. This correlation has not been widely illustrated in previous research (Löfkvist et al., 2019). It should be further highlighted that the children with lowest hours of HA use were the same who did not show presence of any oral stops, dental / alveolar stops or true consonants by the age of 18 months as well as scores below the mean on both consonant proficiency at 3 years and number of words at 24 and 30 months of age. Although the results are in need of larger study groups to confirm the findings, they indicate that early detection and diagnosis of HL, and even fitting with HAs at the recommended age, is not enough for all children with moderate HL to be on par with their peers with NH. Therefore, the additional importance of achieving full-time HA use from first fit cannot be underestimated as it may alleviate the risk of future speech and language delays.

Another important factor to take into consideration when discussing early speech development is that it develops in contingent interactions with their caregivers (Tamis-LeMonda et al., 2014). In the first year of life, the amount of speech the infants are exposed to have shown to be more related to the early processing of speech than relation to age-dependent maturational factors (Marklund et al., 2019). The quality and quantity of parental input is also affected by maternal education (Ching et al., 2015), which in the case of this study was higher than in the population at large, albeit with individual differences. The experience from the recordings of babbling observations in the present study was that parents to children who vocalized less also spoke less to their child which is supported by research (Ambrose et al., 2015; Hoff-Ginsberg, 1994). Another observation was that parents to the children with HL used more direct communication strategies to their children which has also been found in previous studies (Ambrose et al., 2015; Roberts et al., 2018). The findings around the delays in early speech development in the children with HL in combination with the rationale for guiding parents around qualitative communication strategies, motivates the use of instruments that can identify children at risk as early as possible. This may be particularly important for children with mild to moderate HL as the style of language input has been found to vary depending on the potential risk for delays (Nittrouer, 2019). Therefore, it is important to acknowledge delays in early speech of individual children as opposed to rely on the milder degree of HL and the child´s ability to close the gap with time. The babbling observations used in this project is a valid and feasible method (Lieberman & Lohmander, 2014; Lohmander et al., 2017) that could serve as an effective assessment followed by guidance around early speech and communication strategies and hence serve several purposes that meet the need of the child, parents and professionals.

### 6.3.2 Expressive vocabulary development

The findings that expressive vocabulary outcomes were similar between the groups at 18 months was not surprising as the variability in this development of all children has been extensively reported, with number of produced words generally being less than 50 (Berglund & Eriksson, 1999; Fenson et al., 1993; Stoel-Gammon, 2011). The gap in number of words that increased with age between the groups is also congruent with existing findings (Mayne et al., 2000; Yoshinaga-Itano, 2017). Previous studies have identified predictive factors that impact expressive vocabulary development (e.g. degree of HL, absence of additional disabilities, and maternal education) (Yoshinaga-Itano et al., 2017). What this study added to previous findings was the impact of HA use, which showed significant relationships to number of words produced. It was primarily HA use at the ages of 10 and 30 months that were found to have strong correlations with number of words produced at 30 months. It should be noted that the ages of HA use should be thought of as the hours of HA use before the previous timepoint. That is, in this case from fitting to 10 months, and between the ages of 24 to 30 months. These findings indicate that listening experience from as early as possible is an important factor that may affect the acquisition of early vocabulary. This supports research of the first years as being specifically important for the development of future speech skills (Kuhl, 2004). Also, during vocabulary acquisition, new words are added based on already acquired words (Cox Eriksson, 2014). This may have affected the rate in development of produced words in the children with HL between 24 to 30 months. Another reason for the lower number of produced words in the children with HL may have been due to the lower number of different true consonants, which has been suggested by Moeller et al. (2007b). As mentioned in the previous paragraph on early consonant production, the hypothesis that number of different true consonants at 18 months would predict later outcomes on expressive vocabulary was not found in the group of children with HL alone. The correlations only reached levels of significance when the children with NH were added in the analysis. Albeit associations between the two were indicated for individual children, this finding adds further to the discussion whether the predictive variables found in the development of TD children apply to the same extent in children with HL.

In sum, the large heterogeneity in speech and language outcomes in children with all types and degrees of HL has been well documented in research. Regular monitoring of children with profound HL with CIs has been implemented for the past decade. The findings from this study recommend that children with HL of milder degree should be monitored in a similar way.

## 6.4 STRENGTHS AND LIMITATIONS

The main challenge with the outcomes of this doctoral project is the limited number of participants with lack of possibilities of generalizing the findings to a larger population. The small number of participants became a fact despite that recruitment took place in the largest region in Sweden over a whole year. Also, an extension was made to invite participants from

Uppsala, but no children were found to fit the inclusionary criteria. The inclusionary criteria of the study may be reflected upon as strict, but as this project served the purpose to validate a tool in Swedish and to investigate the impact of several variables on the early speech, including more variables was believed to challenge the interpretation of the findings even further. In terms of variables affecting outcomes, it could have been valuable to control for hours of intervention as this is a variable that have shown to effect speech outcomes in children. The project could also have included additional measures of communication and language development.

Thus, the prospective and longitudinal study design with few missing data points over a three-year-period is a strength. Another strength was the inclusion of a reference group of children with NH from the same geographical area. The reference group showed to be representative in terms of development to a larger population. For example, they showed both individual variation and similar group outcomes of expressive vocabulary to other large data. Furthermore, the project controlled for predictors like cognitive development and parental education which were similar between the groups. An interesting finding was that despite the small number of participants, individual patterns were seen when adding all the variables together. However, the group was too small to state any conclusions at a population level. Thus, this clinical research project has gathered extensive experience with methods that are feasible to implement in the Swedish habilitation services of infants and toddlers with HL which could be seen as a value itself.

# 7 CONCLUSIONS AND IMPLICATIONS

Despite early fitting with HAs this group of children with HL showed delays in their consonant production in babbling and early speech compared to the children with NH at 10 and 18 months. This calls for a recommendation to introduce standardised observations of early speech and language development, that could be performed to both evaluate, but also to guide parents how to stimulate this development through contingent communication with their child. The babbling observations used in this project, could as a suggestion, serve this purpose.

The most prominent auditory variable that had an impact on early speech development was hours of HA use. Despite that it is the work of the audiologist to fit and verify the fitting of HAs, other professions like SLPs and SETs can contribute by guiding parents around strategies and motivate parents to reach consistency in full-time HA use. The use of validated parental questionnaires could help parents become aware of the auditory development and functional auditory performance of their child, and accordingly report their observations to their team of professionals. The Swedish version of the LEAQ is now translated, evaluated and feasible to be used in a Swedish context in the youngest age group. However, the findings question the construct validity of this tool as the content was similar to a measure of vocabulary. Depending on the responses for each question and not only the total score, additional information around a child´s early understanding and use of spoken language can be gathered and used in cross-professional discussions of audiologists, SLPs and SETs.

The affected ability to listen in noise as captured by the functional auditory performance measure (PEACH) also calls for collaboration between audiologists, engineers, SETs, parents and teachers of the children to make sure that the children with HL are able to hear as optimal as possible in their homes but also during their time in preschool. Considering the slower development of expressive vocabulary, attention is also needed to teach and promote appropriate strategies to maximize communication and spoken language stimulation, both in the home and in educational settings.

It is widely known that professionals have large impact on parent´s expectations and desired outcomes on their children (Moeller et al., 2013). Therefore, the habilitation services need to be evidence-based, feasible and cost-effective. As evidence around the effects of specific methods that promote typical spoken language development is lacking, monitoring early predictors of future speech and language development at particular time points and/or specific ages, may serve as a starting point for the planning of individualised habilitation plans.

The current project incorporated measures that monitored the effects of early fitting with HAs that to the authors knowledge, are not part of a national protocol but may be performed in local services. The methods used are all available in Swedish and clinically feasible. Regular monitoring of children with profound HL with CIs has been implemented for the past decade. The findings from this study recommend that children with HL of milder degree should be monitored early in a similar way. This could serve the purpose to identify the children at risk of future delays and provide new evidence of the impact of EI on auditory and early speech development in Swedish children.

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