

Rethinking Sound

Computer-assisted reading intervention with a phonics approach for deaf and hard of hearing children using cochlear implants or hearing aids

Cecilia Nakeva von Mentzer



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Till min familj

”Alfabetet är bara en ytterst obetydlig vågtopp på det ofantliga hav som utgör språket. Det lilla fåtalet bokstäver är ett intet i jämförelse med de oräkneliga ljud som de betecknar, och även ljuden är endast tillfälligtvis och slumpvis förnimbara antydningar om det egentliga underliggande språkets sammansatthet och mångtydighet och väldighet.”

”The alphabet is but an insignificant crest of a wave on the enormous sea that constitutes the language. The very few number of letters is nothing compared to the innumerable sounds that they represent, and even the sounds are only incidentally and randomly perceptible indications of the complexity and versatility and greatness of the real underlying language.”

Torgny Lindgren

PREFACE

My fascination in children's spoken and written language acquisition has for many years been the driving force in my work as a Speech Language Pathologist (SLP), although my initial interest in the profession came from my background as a classical singer. In this thesis work I wish to share some of the knowledge I have obtained in meeting children with difficulties acquiring language, and particularly in seeing children who are deaf and hard of hearing (DHH) using cochlear implants (CI) or hearing aids (HA). Besides the novelty of using a method that up till recent days has been exclusively directed to children with normal hearing (NH), this thesis is an endeavour to embrace the heterogeneity of the DHH population. With interest and respect in each individual DHH child's learning potential, I hope the results will inspire the professionals who work with these children. That is, to look beyond barriers and difficulties in the past, and meet the ever-changing pedagogical landscape for the DHH children with open minds and curious attitudes.

Cecilia Nakeva von Mentzer

April 2014

Abstract

In the present thesis, computer-assisted reading intervention with a phonics approach was examined in deaf and hard of hearing children (DHH) aged 5, 6 or 7 years old using cochlear implants, hearing aids or a combination of both. Children with normal hearing (NH) matched for non-verbal intelligence and age served as a reference group. Deaf and hard of hearing children constitute a heterogenetic population regarding cognitive and academic achievement. Many of them do not reach age appropriate levels in language and reading ability during their school years, with negative consequences for later training facilities and job opportunities. Finding relevant intervention methods to promote early language learning and literacy development that are easy to implement, is therefore of great importance. This thesis examined three aspects of cognitive ability (phonological processing skills (PhPS), lexical access and working memory capacity, WMC) and reading ability at three points in time; baseline 1 (B1), pre intervention (B2) and post intervention (PI). Additionally, it explored whether computer-assisted training delivered by means of the Internet in the children's homes, would be a useful and efficient method for the DHH population. The intervention was accomplished by a computer program originally developed to support reading development in children at risk of dyslexia.

In Study 1-II, intervention effects on PhPS were examined, that is, *phonological change* and cognitive predictors thereof. Group comparisons were made according to children's hearing status (DHH and NH). Tasks for cognitive abilities were assessed by means of a computer, with a test battery called the SIPS, that is, the Sound Information Processing System, as well as by pictures and letter cards. A phonological composite score was created by a unit-weighted procedure, that is, each variable of phonological processing skills was calculated in per cent and then summarized. Through the use of the phonological composite score, as well as conducting subgroup analyses in relation to this, we were able to discover patterns associated to children's cognitive abilities and the influence of demographic variables on phonological change. The results from study I and II replicated previous findings of weak PhPS and lexical access in DHH children, and comparable levels as in NH children on complex and visual working memory, WM. Further, results showed that all children improved their accuracy in phoneme-grapheme correspondence and output phonology as a function of the computer-assisted intervention. For the whole group of children, and specifically for children with CI, a lower initial phonological composite score was associated with a larger phonological change between B2 and PI. The influence of demographic variables was evident

in that children with weak initial PhPS were older when diagnosed and had had shorter time with CI. Further, letter knowledge was found to be a mediating factor for phonological change in DHH children with weak initial PhPS.

In Study III, NH children's and DHH children's reading ability was compared at B2 and at PI. Further, effects of the intervention were analyzed. Additionally, cognitive and demographic factors were analyzed in relation to reading improvement. Results showed equivalent levels in reading ability for both groups in the 5 and 6-year old children, but a higher proficiency in the NH 7-year olds at both test points. The intervention seemed successful for word decoding and passage comprehension. Additionally, there was a reduction of nonword decoding errors in both NH and DHH at PI. The DHH children's reading improvement was influenced by visual strategies whereas in the NH children reading improvement was influenced by PhPS and complex WM.

In Study IV, segmental and suprasegmental characteristics in nonword repetition and the connection to nonword decoding were examined in a subsample of the children; 11 children with NH and 11 children with bilateral CI at PI. The findings in Study IV provided several new insights. The syllable omissions and insertions in the CI-group in nonword repetition (NWR) reflected similarities as for phonological processes commonly seen in younger NH typically developing children. No significant difference was found between the groups in decoding accuracy, but differences were observed regarding error patterns. Phoneme deletions occurred almost exclusively in children with CI. The correlation analysis revealed that the ability to repeat consonant clusters had the strongest associations to nonword decoding in both groups. Study IV showed that more thorough and descriptive work on phonological skills in NWR and nonword decoding in children with CI is needed to shed light on decoding strategies in these children.

Overall, the results from the present thesis support the notion that offering a computer-assisted intervention program delivered at home, is an alternative way to support not only NH children with reading difficulties but also to support DHH children's phonological development and decoding proficiency.

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List of studies

This thesis is based on the studies reported in the following papers, referred to in the text by their respective Roman numerals.

- I. Nakeva von Mentzer, C., Lyxell, B. Sahlén, B. Wass, M., Lindgren, M. Ors, M., Kallioinen, P., & Uhlén, I. (2013). Computer-assisted training of phoneme-grapheme correspondence for children with hearing impairment: Effects on phonological processing skills. *International Journal of Pediatric Otorhinolaryngology*, 77(12), 2049-2057.

- II. Nakeva von Mentzer, C., Lyxell, B. Sahlén, B. Dahlström, Ö., Lindgren, M. Ors, M., Kallioinen, P., Engström, E. & Uhlén, I. (2014). Predictors of phonological change in deaf and hard of hearing children who use cochlear implants or hearing aids. Submitted.

- III. Nakeva von Mentzer, C., Lyxell, B. Sahlén, B. Dahlström, Ö., Lindgren, M. Ors, M., Kallioinen, P. & Uhlén, I. (2014). Computer-assisted reading intervention with a phonics approach for children using cochlear implants or hearing aids. *Scandinavian Journal of Psychology* (in press).

- IV. Nakeva von Mentzer, C., Lyxell, B. Dahlström, Ö., & Sahlén, B. (2014). Segmental and suprasegmental properties in nonword repetition – An explorative study of the associations with nonword decoding in children with normal hearing and children with bilateral cochlear implants. Under revision.

List of abbreviations

ADHD	Attention deficit hyperactivity disorder
AVT	Auditory verbal therapy
B1	Baseline 1
B2	Baseline 2
CI	Cochlear implants
DHH	Deaf and hard of hearing
HA	Hearing aids
HH	Hard of hearing
HL	Hearing loss
HRF	Hörselskadades riksförbund (National Organisation for People with HL)
ICF	International Classification on Functioning disability and health
LI	Language impairment
LTM	Long-term memory
NHS	Neonatal Hearing Screening
ND	Neighbourhood density
NWR	Nonword repetition
pcc	Per cent consonants correct
PI	Post intervention
PhPS	Phonological processing skills
pnwc	Per cent nonwords correct
pnwe	Per cent nonword decoding errors
pnwt	Per cent nonword decoding trials

PTA	Pure tone average
ppc	Per cent phonemes correct
pvc	Per cent vowels correct
SBU	Statens beredning för medicinsk utvärdering, Swedish Council on Health Technology Assessment
SCR	Sentence completion and recall
SIPS	Sound information processing system
SL	Sign language
SLP	Speech language pathologist
SNHL	Sensorineural hearing loss
SOU	Statens offentliga utredningar. Swedish Government Official Reports
SPSM	Specialpedagogiska skolmyndigheten, National Agency for Special Needs Education and Schools
SSSL	Sign to support spoken language
WF	Word frequency
WM	Working memory
WMC	Working memory capacity

INTRODUCTION

United Nations Educational, Scientific, and Cultural Organization, UNESCO's theme for 2014 is "Equal Right, Equal Opportunity: Education and Disability".

This theme stresses the urgency of not just making education accessible but also inclusive for all (UNESCO, 2014). UNESCO highlights the need of strong efforts everywhere to uphold the right for education, to keep with article 24 in the United Nations (UN) convention (UNICEF, 2014) of the rights for persons with disabilities.

Reading ability is one of the most important tools in education. Reading ability enables the democratic right of each individual to participate in the society. In planning for deaf and hard of hearing (DHH) children's education we need to embrace the UN-convention article 24. Concretely, this means we need to address every individual DHH child's abilities and opportunities, and put in enough resources to enable them to reach their full potential.

The International Classification of Functioning, Disability and Health, ICF (Socialstyrelsen, 2014) broadens the perspective for Speech Language Pathologists (SLPs) who work with DHH children. With the ICF framework, SLPs may leave a strict medical view where intervention aims at correcting deviant body functions. Instead, ICF provides a tool where intervention goals incorporate environmental and personal contextual factors. Methods to improve activity and participation in children with communicative difficulties have recently been summarized in "Young direct", a report from the Children's Ombudsman (Barnombudsmannen, 2014). The report highlights the importance of using augmentative and alternative communication and to look into each individual child's communicative need. When performing an intervention study, as in the present thesis, where the same method is used in a heterogeneous group of children, it is of considerable importance to look at each child's starting level to appreciate the developmental steps taken.

The present thesis focuses on computer-assisted reading intervention with a phonics approach for DHH children 5, 6 and 7 years of age who are using cochlear implants (CI), hearing aids (HA), or a combination of both. This is, to the author's information the first time that a method, exclusively developed to support NH children's reading acquisition, and particularly children at risk for developmental dyslexia, is used in a study on the DHH population. The study could not have been accomplished had it not been for the development of technical

hearing devices during the last century.

A CI is a technical device that was first introduced for children with severe to profound sensorineural hearing loss (SNHL) in the late 1980's (Powell & Wilson, 2011). Although the primary goal of cochlear implantation is open set auditory-only speech understanding in everyday listening environments, the hearing, language and literacy outcome in children receiving CI is quite diverse (Peterson, Pisoni & Miyamoto, 2010). This makes intervention a natural element of many of these children's lives. Since the advent of CI a large body of research has been conducted on this population. Studies of children with mild-to-moderate HL using HA, on the other hand, have been limited in number and scope, although the HA have been at hand for a much longer period of time. Thus, less is known about the cognitive development and methods that promote optimal language and reading development in these children.

In the present thesis DHH children's cognitive abilities are examined, primarily phonological processing skills (PhPS), that is, how they deal with the sound patterns of spoken language. Additionally, DHH children's working memory capacity (WMC) is examined, that is, how they simultaneously process and store incoming information over a short period of time. Further, lexical access; that is, how the DHH children retrieve words from long-term memory, LTM, is investigated. Finally, DHH children's reading ability is inspected, mainly how they decode words and nonwords, and secondly, how they comprehend passages of written text pre and post intervention, with a computer-assisted program using a phonics approach.

The outline of the thesis

This thesis starts with an audiological chapter including sections regarding communication mode and support by SLPs. This is followed by a historical overview. My wish is to convey how historical events have shaped the present views on DHH children's communicative needs and learning potential. Chapter III focuses on cognitive development, including early language and reading development in typically developing children and in DHH children. Here, my main ambition is to acknowledge the complexity of language acquisition in typically developing children and the importance of recognizing how consequences of a hearing loss, (HL) come into play. Intervention methods to support language and reading development are also presented. Following a brief summary of the papers, I-IV, the findings on the cognitive skills, phonological processing, WMC, lexical access, and reading are discussed as well as the effects of the intervention. This is followed by a section on methodological issues in research on children with CI and HA. Finally, clinical implications

and some ideas for future research are suggested.

BACKGROUND

AUDIOLOGICAL ASPECTS

This section starts with a brief description of the inner ear followed by an overview of prevalence, classification and definitions of hearing loss (HL). The present thesis examines children with sensorineural hearing loss (SNHL). Therefore the main focus will be to describe the consequences of SNHL.

Inner ear anatomy and function

The human cochlea is the receptive part of the auditory organ capable of exceptional sound discrimination, in terms of both frequency and intensity (Rebillard, Pujol & Irving, 2014). It is a mirror-shaped, fluid-filled, coiled, bony tube, 3–4 cm long, situated in the temporal bones (Rask-Andersen, Liu, Erixon et al., 2012). The human cochlea holds the structures of the auditory organ that convey mechanical acoustic energy through air and fluid to electrical bioenergy. This is made by means of the basilar membrane, which holds the receptor organ of Corti, and outer and inner hair cells ordered alongside it. There is considerable individual variation in the dimensions of the cochlea, which may explain the otosurgeon's difficulties sometimes to insert electronic arrays even in a normal cochlea (Erixon, Högstorp, Wadin et al., 2008), see Figure 1.



Figure 1. Corrosion cast showing variations in the anatomy of the human cochlea. From Erixon et al. (2008). Reprinted with permission from the author.

The human cochlea has a tonotopic organization, that is, specific places along the basilar membrane respond to certain frequencies, thus the basilar membrane acts like a frequency filter of the incoming auditory signal (Rebillard et al., 2014). The acoustic nerve (the 8th cranial nerve) carries the auditory information in the form of bioelectric outputs, so called action potentials that pass through five different relay nuclei in the brainstem (the lower part of the brain) and thalamus, which is essentially a relay station that conveys different kinds of sensory information connected to input from both ears, important for processing complex sounds and sound localization. Eventually the auditory information reaches the primary auditory cortex where auditory sensation occurs (Biacabe, Chevallier, Avan et al., 2001). One interesting feature in the auditory system is that, besides its rich afferent pathways ascending the central nervous system (CNS), the sensitivity of hair cells to physiological stimuli is controlled by the CNS via descending *efferent* nerve fibers. These have been found to play an important role in sensory integration (Rabbitt & Brownell, 2011).. These have been found to play an important role in sensory integration (Rabbitt & Brownell, 2011). Thus, efferent innervations modulate the inner ear's afferent inputs to meet the behavioral needs of the organism. For humans this is particularly important for sound localization in a noisy environment (Rabbitt & Brownell, 2011).

Hearing loss

According to the World Health Organization, HL is one of the six leading contributors to the global load of disease. Approximately 9% of the children worldwide have a disabling HL (> 40 dB HL), a number that varies greatly with socioeconomic status in the country (WHO, 2012). About half of disabling cases of HL are worldwide preventable (Paludetti, Conti, Nardo et al., 2012). Pure-tone audiometry is the most common behavioral assessment of an individuals hearing thresholds. It provides information about the peripheral hearing acuity for single frequency tones across the four key-frequencies for speech (0.5, 1, 2 and 4 kHz hearing level), and is illustrated as a graph, the audiogram. The most common pattern of HL, as indicated as a right-sided sloping curve, is reduced hearing in the higher frequencies which affects perception of speech sounds as for example /f/, and /s/. This type of HL is typically associated with aging or noise exposure (ASHA, 2011).

Hearing dysfunctions in children can be classified by type, degree, pattern, time of onset, etiology, as well as in relation to consequences on speech development (Paludetti et al., 2012;

Zahnert, 2011). In brief, HL can be divided into *conductive*, *sensorineural* (described in detail in the next chapter), *mixed* and *central types*. Mild hearing losses range from 26-40 dB, moderate from 41-60 dB, severe from 61-80 dB, and profound including deafness, from 81 dB or higher (Arlinger, 2007; WHO, 2014). Conductive HLs are mechanical in nature and include the disturbances that can arise along the sound conduction pathway (that is, the outer ear canal, the middle ear including the tympanic membrane and the three ear ossicles, the stapes, incus and stirrup). Maximum degree of reduction for a conductive HL is up to 60 dB HL (Zahnert, 2011). In many cases the HL can be restored by surgery or medical treatment, for example antibiotics in otitis media. Mixed HL is, as the name suggests, the co-occurrence of conductive and sensorineural HL. It is a quite common condition for children with sensorineural HL who have an ear infection. Central HLs are caused by neural dysfunction in the higher auditory pathways and the auditory cortex. One condition of particular interest for SLPs is central auditory processing disorder. Here, children have listening difficulties despite normal audiograms (Ferguson, Hall, Riley et al., 2011). Symptoms are, for example, reduced speech recognition in noise and poor sound localization. Since listening difficulties often occur in different clinical populations as for example specific language impairment and neuropsychiatric disorders, an interdisciplinary approach is recommended in diagnosing and treating these children.

Hearing losses present at birth are defined as *congenital* and those that appear after birth, as *acquired*. Furthermore, the definitions *prelingual* (before 2y) and *postlingual* (after 2y) are often used clinically in relation to whether the onset of HL is before or after spoken language acquisition (Arlinger, 2007). In the present thesis, I use the term congenital HL for children diagnosed before 1 year of age.

Sensorineural hearing loss

Sensorineural hearing loss is a multifaceted condition caused by a combined dysfunction of the cochlea, in particular the organ of Corti (such as loss of outer or inner hair cells) and the auditory nerve (such as auditory neuropathy, Ng, 2013). Sensorineural HL affects environmental hearing capacity, for example the ability to detect the sound of a car approaching from behind (so called warning sounds), appreciating sounds in nature and listening to music. These factors are closely connected to quality of life (Elberling & Worsøe, 2006). Perceptual consequences of SNHL include five dimensions; 1) Reduced audibility (requires a louder signal), 2) decreased frequency resolution (e.g., the ability to discriminate between vowels /i/ vs. /e/, or differentiate fricatives in the higher frequency range), 3) reduced

temporal resolution (that is, the ability to perceive fast transient speech sounds, consonants e.g. /p/, /t/, /k/ or clusters /skv/), 4) reduced dynamic range (that is, decreased sensibility to soft sounds and increased sensibility to strong sounds), and 5) impaired ability to localize sound sources and to discriminate pitch. This latter aspect is connected to binaural hearing, that is, listening with two ears (Elberling & Worsøe, 2006).

The prevalence of congenital, bilateral, permanent sensorineural hearing loss of 35 dB or more is estimated at 1.2 to 1.6 per 1000 live births in the Western world (Bamford, Uus & Davis, 2005; Zahnert, 2011), and increase as a result of meningitis, delayed onset of genetic HL or late diagnosis. Taken together, the estimated prevalence of SNHL in patients >18 years is 6 per 1000, but much higher in countries with lower socioeconomic status. Twenty to thirty per cent of the affected children have a profound SNHL, and approximately 30% of these children have an additional disability, most commonly cognitive impairment (Paludetti et al., 2012).

The etiology of SNHL is genetic in 25% of the cases, acquired in 18% (e.g. infectious, metabolic, toxic, and birth trauma), and indeterminate in > 50% of the cases (Zahnert, 2011). According to Zahnert (2011) 30% of the genetic HLs are due to congenital syndromes (e.g. Goldenhar and Waardenburg syndrome) and 70% are non-syndromic (the most common type is due to a genetic mutation that impairs the synthesis of the transmembrane proteins connexin 26 and 30).

Identification of HL has improved since the neonatal hearing screening (NHS) was introduced. In Europe, the UK started in 1992 and Linköping University hospital was the pioneer in 1995 in Sweden (Hergils, 1999). Since 2008 it is obligatory in the whole country (personal communication Uhlén, May 2014). The NHS procedure tests the responses of the outer hair cells by means of otoacoustic emissions within the child's first days of life. Normal responses indicate an essentially normal function of the auditory organ at 30 dB hearing threshold or less. Those cases, where no responses are obtained, are followed up by brainstem response audiometry. Other methods to evaluate children's hearing are with the Boel test at 7-9 months (Jonsell, 2011), indirectly at the language screening procedure at 2,5 or 3 years of age, with pure tone audiometry at 4 years (recommended) and at school start (SBU, 2012).

Audiological intervention

Early enrollment in audiological services is of utmost importance for children with SNHL. If a child with SNHL is identified < 6 months of age, there is a good chance for language development close to that of NH children (Moeller, 2000). Population based studies in countries that use the neonatal hearing screening (NHS) procedure, report that audiological assessment, enrollment in early support programs, and hearing aid fitting may be accomplished as early as 5, 10 and 16 weeks of age, which is much earlier compared to previous methods when NHS was not in use (in England, for example the mean age of identification in the past was 2 years of age). Often, children with severe HL are fitted with HA earlier than those with moderate HL (Uus & Bamford, 2006), and it is routine to first fit children who are candidates for CI with HA. It is very important with safe routines for follow up of children who have failed the screening. Here, some barriers may still remain, for example lack of service-system capacity, lack of provider knowledge, challenges to families in obtaining services, and information gaps (Shulman, Besculides, Saltzman et al., 2010). At the introduction of CI, criteria for receiving CI were profound HL in both ears (Karltorp, 2013). The most common situation worldwide at that time was unilateral CI. Since 2004 Swedish children with bilateral profound HL are provided CI in both ears. Further, since 2005 children with an asymmetric HL (profound in one ear and moderate in the other), receive a CI in one ear and a HA in the other, that is, have bimodal hearing. Still, another option since 2008 is hybrid hearing aimed for individuals with a profound HL above 1000 Hz (Erixon, 2014; Karltorp, 2013). In these cases the surgeon inserts a short electrode, thus preserving acoustical hearing in the lower frequency range.

Deaf and hard of hearing children in Sweden

Communication mode

In 1991, with the introduction of paediatric CI in Sweden, there was an initial phase of uncertainty regarding communication mode and the educational needs within the deaf population (Jacobsson, 2000; Svartholm, 2005; Uhlén, 2009). Since then, bilingualism (sign and spoken language) has been part of the political endeavour, which should include all DHH children. In 2006 the Swedish Ministry of Health appointed a one-man investigation of the Swedish sign language (SL), conducted by the former cabinet minister Danielsson (SOU, 2006). The investigation brought forward three perspectives on the importance of strengthening sign language. These were language policy, disability policy and democracy. The exact wording in the document about the benefits of the new HA that is, CI, was: "the

outcome of scientific studies that have been conducted in Sweden of children with CI and their language development points coherently to the importance that these children should be given access to sign language". The Swedish National Association of Hard of Hearing People (HRF, 2007) also emphasized that bilingualism was a right and necessity for all deaf children. HRF brought forward a proposition that education in sign to support spoken language (SSSL) or SL should be mandatory for all parents of DHH children (HRF, 2007; Rogell-Eklund, HRF, personal communication 2014-04-11). An attempt to grade the recommendations in the investigation came in a written comment from the Children's ombudsman (Barnombudsmannen, 2006; Johnson, e-mail communication 2014-05-08). They stated, that in general, the investigation would have benefitted from a more apparent child perspective. Further, they specified that it was not consistent with the Convention on the Rights of the Child (UNICEF, 2014) to put children in the same educational setting, just on account of a shared disability. In sum, there are, possibly, different solutions regarding which communication mode to choose for the DHH, and these will come naturally as the child develops. Even if, in theory bilingualism, that is sign and spoken language, seems as an optimal goal (although weak scientific support for the benefits have been reported, see Knoors & Marschark, 2012), this is presumably not a realistic goal for many DHH children, considering that 95% of the parents have normal hearing (Cole & Flexer, 2011).

Support by speech language pathologists

The prevalence of language impairment (LI) in children in the population at large is 6-8% during the preschool years (Leonard, 2000; Nettelbladt, 2007). Corresponding figures for deaf children using CI and children with a mild to moderate HL using HA are more difficult to obtain. At the least it should be the same as for children overall, but numbers up to 50% in research on a small number of children using HA, have been reported (Hansson, Sahlen & Mäki-Torkko, 2007). At present, there are approximately 4000 children in Sweden who use HA (Socialstyrelsen, 2009), and about 800 children below 18 years of age who have received CI (Barnplantorna, 2013). These children are enrolled in Audiological services from the age of identification and are followed up until the age of 18 years. Audiological services typically include Ear Nose Throat Audiology physicians, audiologists, special pedagogues, engineers and welfare officers but not by rule speech language pathologists, except in larger cities where SLP-programs exist, and in the CI-teams. Usually, DHH children are referred to SLPs from the child health care due to failed language screening at 2.5 years of age, or by the audiologist when the child shows signs of language delay. However, still in many cases, as

the SLPs do not meet the DHH child at first instance where intervention takes place, the chance of a DHH child receiving language assessment is largely dependent on other professionals' judgments.

The situation for children receiving CI is slightly different. In Sweden there are five CI-teams, these are situated in Gothenburg, Linköping, Lund, Stockholm and Uppsala. All teams offer support by SLPs. SLPs who work in the CI-teams assess and support the child's listening, communicative and spoken language development. Particular attention is given for congenitally deaf children during the first year, due to the importance of providing auditory stimulation when brain plasticity is high (Gordon, Wong, Valero et al., 2011a; Kral, Tillein, Heid et al., 2006). Since many deaf children have additional disabilities (for the various causes and syndromes associated with HL, see Cole & Flexer, 2011), the main intention with CI for these children is most often not spoken language ability. Rather, the support of the SLP in these cases is to provide the child, family and educational setting, facilitative advice for an optimal communicative development, which often includes the use of augmentative and alternative communication. SLPs work in close collaboration with teachers of the deaf, special pedagogues and social officers due to the complexity in these children's language and learning situation. Interdisciplinary work thus provides the best way to meet these children's needs.

Courses on DHH children's language development are given as part of SLPs undergraduate programs during the first year at the University. SLP programs are given in six places; Gothenburg, Linköping, Lund, Stockholm, Umeå and Uppsala. In Lund SLP students attend the same courses as the audiologists the first two years. Further on in the SLP's training, courses on DHH children's language development are given at an advanced level. Typically they give 1-2 credits (out of totally 160; four years of full studies). Deaf and hard of hearing children's language development is also acknowledged within the lessons on typical and atypical language development, e.g. as SSSL - courses. At Karolinska Institutet a two-semester course in Auditory Verbal Therapy (AVT) has been given since 2005. Approximately 50 participants (special pedagogues, SLPs, and a few audiologists) have finished the course (Löfkvist, personal communication April 2014). The variation in length of studies on DHH children's language development in the different SLP programs is quite substantial. Thus, it is difficult to obtain a comprehensive picture regarding the exact amount of training the SLPs receive in this area.

In sum, SLPs are needed at various places in the society to support DHH children's communicative, language and literacy development, for example at the hearing central, in the Audiological clinic, in language units, preschools and schools. SLPs need to improve the national guidelines regarding SLP students' basic training as well as inform professionals and policymakers regarding *how* SLPs may contribute in the habilitation and education of the DHH child. This is of particular importance due to many DHH individuals negative experience of oral training methods used in the past. With the use of the ICF framework, which stresses activity and participation, in combination with early intervention and modern technology, the situation, and consequently, the learning potential has changed dramatically for DHH children born today.

Listening through cochlear implants

A cochlear implant differs substantially from a hearing aid. Hearing aids amplify sounds but cochlear implants compensate for damaged parts in the inner ear. Further, CI bypass the impaired hair cells, and coded electrical signals stimulate different parts of the hearing nerve fibers, which send information to the auditory cortex in the brain (Cole & Flexer, 2011). Optimal mapping for speech perception is further secured by continuously evaluating psychoelectric parameters, that is, threshold levels, comfortable levels, dynamic range and electrode implant values throughout the first year of implant use (Henkin, Kaplan-Neeman, Muchnik et al., 2003). A CI delivers auditory stimulation but it does not restore the auditory perception to a normal level. Additionally, it does not deliver the entire speech spectrum in terms of the same fine acoustic-phonetic details or the rich spectral and temporal resolution as in acoustical hearing (Moore, 2008; Nittrouer, Caldwell, Lowenstein et al., 2012). Consequently, certain aspects of the speech signal are more difficult to incorporate in the child's language, for example consonant clusters and fragments with weaker amplitude.

There are several factors that influence how the child develops listening and spoken language through CI. Demographic variables are for example age at implant (Sharma, Dorman, Spahr et al., 2002; Sharma, Dorman & Spahr, 2002b), duration of deafness (Cole & Flexer, 2011, p 157), residual hearing and cause of hearing loss (De Barros, Roy, Amstutz Montadert et al., 2014; Philips, Maes, Keppler et al., 2014), daily use (Archbold, O'Donoghue & Nikolopoulos, 1998), whether the child uses bilateral implants, sequentially or simultaneously implanted, as well as time elapsed in between implantations (Cole & Flexer, 2011, p 158; Gordon, Jiwani & Papsin, 2011b). Personal and environmental factors that affect the outcome are whether the

child has an additional disability (Wakil, Fitzpatrick, Olds et al., 2014) and the amount of auditory stimulation that the child receives, that is, communication mode in the family and educational setting, as well as parents use of facilitative language techniques (Cruz, Quittner, Marker et al., 2013). Surgical variables have also been found to affect the outcome, for example insertion techniques, number of active electrodes in the cochlea, and placement adjacency of the electrode array to the auditory nerve (Addams-Williams, Munaweera, Coleman et al., 2011; Rask-Andersen et al., 2012).

In sum, variation in listening and language skills is explained by many factors, where the complexity of the auditory organ is one. Additionally, it is of considerable importance to acknowledge the ears' connection to the brain and the interaction between HL and cognitive functions. Basically, better spoken-language outcome is seen when the majority of complicating factors are diminished. That is, for optimal development the child must be offered early intervention. Thus, for all DHH children this means early diagnosis and early fitting of hearing aids, and for congenitally deaf children particularly, this means early implantation.

Listening through hearing aids

As has been acknowledged previously, age at intervention and early access to sound are the variables that best predict the success of children with SNHL in using spoken language in everyday settings. The purpose of a hearing aid is to make sounds more audible to the listener and to activate neural circuits responsible for detecting and discriminating acoustic information (Sullivan, 2013). However, it cannot restore damaged hair cells. Consequently, the benefit from listening through hearing aids is usually a compromise between the five dimensions of hearing: audibility, dynamic range, frequency and temporal resolution, and binaural hearing (Elberling & Worsøe, 2006, p 73). The following requirements are put on hearing aids to provide the necessary audibility to develop speech and language. First, appropriately fitted, second, verified electro-acoustically and third, have real-ear probe microphone measures (Sullivan, 2013). Several interactive factors affect children's degree of success from a hearing aid, for example the child's residual hearing, the amount of time each day wearing hearing aids, the quality and quantity of auditory-based therapy and interaction in an "auditory world" created by family members, friends and therapists, as well as use of necessary additional technologies, such as FM systems to enable distance learning (Cole & Flexer, 2011, p 133). Sullivan (2013) points out the need of more research on fitting practices

in pediatric populations and the inappropriateness to rely on the outcome in the adult literature when interpreting the results for children. For example, regardless of hearing status, children's word learning rate has been found to be improved using signal processing with a broader bandwidth (Pittman, 2008). Further, there is a need to know more about how the hearing aid user can make better use of their amplified sound. One way to accomplish this is to perform studies on how speech recognition ability in noise may be affected by auditory training. In sum, the habilitation process of children with SNHL started out on how to provide access to auditory information. Now it is time to take into account how hearing aids may provide optimal stimulation in accordance with each individual child's auditory and cognitive development and to contextual demands.

Historical aspects with focus on communication, language and reading

In the following chapter I will focus on communicative aspects and language learning in the tutoring of DHH children during the 18th and 19th centuries. This is to enable a more thorough understanding regarding present discussions on bilingualism and educational methods for DHH children. For language learning I specially recognize the comprehensive work by Pauncefort Arrowsmith (1819), which was a strong contribution to deaf education in times past and parts of it could still be of value for teachers and SLPs meeting DHH children today. This is followed by more recent discussions on reading instruction and literacy acquirement from the late 1960's and onward, with the aim to understand how the present thesis relate itself in the fast changing landscape of deaf education.

Deaf education

Language and communicative development in DHH children has attracted the interest of SLPs, developmental psychologists, social anthropologists and educators since many years (Laes, 2011; Meadow, Greenberg, Erting et al., 1981; Nettelbladt & Samuelsson, 1998; Tamm, 1916). Many important events regarding DHH children's education took place during the middle of the 18th century. At that time, two main educational methods were practiced; the manual and the oralist approach. In short, these methods had two proponents; Charles-Michel de l'Épée from France and Samuel Heinicke from Germany. De l'Épée was a catholic priest who is regarded as the "father of deaf education". He developed an idiosyncratic gestural system that he connected to the French language, acknowledging the manual language used by deaf people to which he added grammatical functions. The main component that contributed to de l'Épée's fame was that he held his classrooms and methods available to

other educators. This enabled the manual approach to spread within France and to other countries (Monaghan, 2003; Pouncefort Arrowsmith, 1819). Heinicke, on the other hand, represented the oralist approach. He was an educator who stressed the importance for deaf people to be part of the hearing society, and thus implemented lip-reading strategies and use of spoken language in the classroom. Even if he did not advocate for DHH pupils to use sign, he did support his speech-oriented methods with fingerspelling techniques (Monaghan, 2003). After the establishment of formal schooling for DHH in Europe (e.g., in Paris; Institut National de Jeunes Sourds de Paris in 1760, and Leipzig; Electoral Saxon Institute for the Mutes and Other Persons Afflicted with Speech Defects in 1778) and in Sweden (Stockholm; Manilla school in 1808), the importance of speech as the first communication mode, and the high value of the sense of hearing for instruction became specifically emphasized, much as a result of the Milano congress 1880. Social policy in Sweden at that time highlighted (inspired by the French educator Montainer in 1870) that the DHH would need to adapt to the hearing culture via lip reading. Not until the middle of the 20th century, sign language (SL) was permitted in education after years of obscured subsistence (Bengtsson, 2005). As DHH children entered formal schooling, educators became increasingly aware of the challenges these children faced in acquiring spoken and written language (Power & Leigh, 2000). Since the DHH children varied in how they benefitted from spoken language input, school teachers basically used two methods; a combination of written language and SL, or spoken language (Svartholm, 2009).

Language tutoring of DHH children during the 19th century - Pouncefort Arrowsmith

In the comprehensive book “The art of instructing the infant deaf and dumb” inspired by de l’Epée, Pouncefort Arrowsmith (1819), gave detailed instruction on the language tutoring of deaf children, for example how to teach the alphabet, work with words and meaning, as well as with utterances and grammar. The main purpose of the book (which in many ways was a strong reaction to the asylums where most of the deaf education took place) was to increase public knowledge about the tutoring of deaf children. Pouncefort Arrowsmith stressed the importance of introducing a proper order of the different elements in instruction. For example, his advice was to work with writing and fingerspelling *before* introducing vocabulary work (word learning). Thus, in this two hundred year old document early introduction of written language for deaf pupils was emphasized. In the training with written words, capital letters were first introduced, followed by lower-case letters. Interesting to note is that Pouncefort Arrowsmith’s method included *not only* to present the whole word (compare whole-language

approach in reading, cf. Stanovich, 2000), but also to acknowledge its parts, that is the pupil should learn how to put the letters in the right order. Together with the curiosity of the child to learn more words, this work progressed in combination with the use of natural signs, which for many function words were easier.

Speech training was advised to be performed using tactile (placing the student's fingers to feel the movement of the teacher's tongue and jaw, and vice versa), and visual support (written letters). Further, the importance of not being too harsh in speech instruction was emphasized since great difficulties would be expected. Lip-reading strategies basically included the same components, which meant use of tactile and visual support (Paucefort Arrowsmith, 1819).

Paucefort Arrowsmith was way ahead of his times in recognizing the importance of deaf children starting early in school *together* with typically developing children, as he wrote (1819, p. 85):

‘It is very extraordinary that this book of Abbé de l'Epée which was published in 1801, should have entirely disappeared and that there is not a single copy now to be met with. I am inclined to think that the work was suppressed; for if publicity had been giving to it, the deaf and dumb would have been educated common with other children, long before now’.

After having shed light on Paucefort Arrowsmith's contribution to deaf education during the 19th century I turn to more recent literature. Several factors contributed to the gradual increase of methods in deaf education today that earlier were exclusively directed to NH children (Kyle & Harris, 2011; Trezek & Hancock, 2013; Trezek & Malmgren, 2005; Trezek, Wang, Woods et al., 2007; Wang, Trezek, Luckner et al., 2008). These were, for example, increasing knowledge regarding the importance to utilize residual hearing during the 20th century (Cole & Flexer, 2011), and technical advancement, that is auditory methods were made possible to use with the development of conventional hearing aids and cochlear implants (Cole & Flexer, 2011; Levitt, 2007; Mudry & Dodele, 2000; Wheeler, Archbold, Hardie et al., 2009). As Marschark, Archbold, Grimes et al. claimed in 2007: “There has never been a better time to be a deaf child...or a parent or educator of one”. It should nevertheless be remembered that still in the 21st century, choice of communication mode and integration vs. segregation for DHH children are issues continuously debated (Powell & Wilson, 2011).

The times they are a-changin'

Recently, Knoors and Marschark (2012) acknowledged the gradually changing situation for different generations of children using cochlear implants in the Netherlands and the US. Since the health insurance and overall socio-economic status differ between these countries and Sweden, direct translations of all conclusions made in the mentioned report are not possible, but many aspects are worth considering for Swedish circumstances as well. Knoors and Marschark (2012) propose a need for a more differentiated view of the implementation of signed and spoken language in relation to children with possibly different language needs. They identify three different groups of CI-users: 1) deaf children born in the last five-year period who have received their implant before two years of age have the most favourable opportunity of spoken language acquisition, 2) deaf children born in the last ten year period, implanted at around 3-4 years of age have a less favourable prognosis of spoken language acquisition and would be in need of a combined approach with both auditory and visual communication, and 3) in secondary education approximately only 20 per cent of the deaf children have received an early implant and still require the kind of teaching methods set up within deaf education in the 1990's, that is with more focus on sign language.

Knoors and Marschark suggest that sign to support spoken language (SSSL) could be useful in the young ages, as well as in situations when the growing child faces challenging situations for example, in background noise, temporary equipment malfunctions or dead batteries. The National Association for Children with CI and/or HA (Barnplantorna) principally conveys a similar differentiated approach. They stress that the parents' knowledge regarding their child's communicative needs should guide their choice of communication. Further, they emphasize that the hearing technology represents a major change in what is a reasonable requirement to impose on deaf children's language development.

Reading acquisition

Initially, in the teaching of DHH children, there was a widespread view that acquiring reading skills would be a way to compensate for the difficulties deaf children faced in acquiring spoken language. However, soon it became evident that degree of hearing loss negatively affected literacy acquisition, particularly reading comprehension (Power & Leigh, 2000). During the 1950s-60s several teachers of the deaf had acknowledged this (Conrad, 1977). With Conrad's discouraging results that 75% of the deaf school-leavers (15-16½ years of age)

in Great Britain lacked functional reading skill, (the criterion set at a reading age of 11 years), the discussion regarding educational reading practice began to swing.

In 2008, the National Evaluation of deaf Swedish children's target achievement in the Swedish language was carried through (Hendar, 2008). This demonstrated that as many as 68% of children who attended special schools for the deaf (with varied use of technical hearing aids) did not reach age appropriate school-leaving certificate levels (16-17 years of age) in one or more subjects. Corresponding rates for children who attended schools for the hard of hearing were 44%, and for mainstreamed DHH children 32%, compared to 24% for the pupils in elementary school in 2006 (Hendar, 2008). Again, these differences obviously reflect the huge variation in demographic variables among the deaf and hearing impaired population.

The National Agency for Special Needs Education and Schools (Specialpedagogiska Skolmyndigheten; SPSM) has published guidelines concerning the support of literacy development in DHH (Roos, 2009). These guidelines stress the importance not to compare DHH children's literacy development to that of NH children. Instead one should find individualized solutions for each DHH child to develop language and literacy, preferably by using different alternative and augmentative communication such as sign language. Thus, the expectations from the SPSM on DHH children's reading ability are in sharp contrast to the reading levels found in scientific studies of Swedish children with CI (Lyxell, Sahlén, Wass et al., 2008; Lyxell, Wass, Sahlén et al., 2009; Wass, 2009). Perhaps this reflects a limited interaction between research and educational institutions, which could be improved by finding more arenas where the different representatives can meet and discuss.

In sum, cochlear implants and hearing aids are technical devices that enable auditory stimulation to children who are DHH. They have positive impact for many of the areas of these children's development, for example, language and literacy skills. Nevertheless, individual factors are important to consider in relation to children's attitudes and satisfaction with the devices. Together the families, children, educators, medical professionals, scientists and policymakers need to share their knowledge to enable an optimal learning situation for each individual DHH child. One way to move forward is to consider intervention methods in DHH children that have been fruitfully implemented for typically developing children as well as for other clinical populations. These could be different kinds of cognitive interventions (Helland, Tjus, Hovden et al., 2011; Latham, Patston & Tippett, 2013; Oei & Patterson, 2013) and cognitive stimulation in the children's homes (Cates, Dreyer, Berkule et al., 2012) as well

as in the classroom (Grenner, Åkerlund, Johansson et al., 2014)

Cognition and language

As a brief introduction to the chapter of cognitive development I acknowledge the work ‘Thinking and Language’ by the Russian psychologist Lev S Vygotsky (Vygotsky, 1934, 1999). Although Vygotsky only reached an age of 37, he has influenced a great number of people working with child development (Piaget, 2000; Vygotsky, 1999). Today, cognitive psychology embraces several intellectual processes, such as attention, perception, learning, memory, language, problem solving, reasoning, and thinking (Eysenck & Keane, 2005). Vygotsky (1999) elaborated on several of these when he discussed children’s mental development. For example, he described the human consciousness as undifferentiated, separate functions during infancy, the perceptual development and refinement during childhood, and the development of memory during school age. The most important and influential parts of ‘Thinking and Language’ in recent times, is Vygotsky’s *sociocultural* approach embedded in *dialect* theory. Above all, this approach enabled Vygotsky to overthrow the cognitive revolution that was started by Piaget (Newcombe, 2013) by addressing the *social function of language*, which creates new meaning and supports different ways of thinking. Moreover, Vygotsky acknowledged the *changing relationship* between thinking and language through development, both quantitatively and qualitatively, that is, their developmental paths *meet and separate* continuously: ‘When the line of thinking intersects the line of language, thinking becomes lingual and language becomes intellectual’ (Vygotsky, 1999).

COGNITIVE DEVELOPMENT

As have been previously acknowledged, cognitive psychology refers to several different processes including attention, perception, learning, memory, language, problem solving, reasoning, and thinking (Eysenck & Keane, 2005). This chapter starts with a section on language acquisition followed by a presentation of the constructs that I have studied in the four papers, that is, phonological processing with an emphasis on phonological output, phonological representations and phonological working memory; lexical access, visual and complex working memory, and reading. In this chapter, I explain the constructs, address typical cognitive development and consequences for children who have a sensorineural hearing loss (SNHL) and use cochlear implants or hearing aids.

LANGUAGE ACQUISITION

To provide an overview of the language development in the participating children in the present thesis I will take early speech perception and production in normal hearing children as a starting point. Following this, I present possible effects of SNHL on speech perception and production in general. For limitations on speech processing due to the restrictions of listening through CI and HA I refer to the chapter on Audiological aspects.

Speech perception

Encased in the mother's womb, low-pass filtered speech sounds can be heard by the fetus' auditory system from about the 5th gestational month (Cole & Flexer, 2011; Porcaro, Zappasodi, Barbati et al., 2006; Querleu, Renard, Versyp et al., 1988). Experimental studies have shown that newborn infants detect prosodic cues in language (Sambeth, Ruohio, Alku et al., 2008), discriminate between familiar and novel voices (Kisilevsky, Hains, Lee et al., 2003), attend to and produce intonation and rhythmic characteristics of the ambient language (Hohle, Bijeljac-Babic, Herold et al., 2009; Mampe, Friederici, Christophe et al., 2009). Altogether, these findings suggest that suprasegmental patterns of the speech signal have formed early memory traces in auditory cortex. Even in utero, although sounds are quite softened, language experience affects infants' phonetic perception, shown as a sensitivity to native and non-native vowels in neonates, as young as 30 h of age (Moon, Lagercrantz & Kuhl, 2013). Together, these perceptual fundamentals gradually become imprinted by the phonology of the native language, first in the vowel system at 6 months, followed by the consonant system at 10 months (Kuhl, 1994; Kuhl, Stevens, Hayashi et al., 2006) and then with the stress pattern during the second part of the first year of life (Skoruppa, Cristia,

Peperkamp et al., 2011). Children at this age combine prosodic preferences with phonotactic information to break into the speech stream and detect word-boundaries (Clark, 2009), which is associated to word learning capacity (Junge, Kooijman, Hagoort et al., 2012; Werker & Yeung, 2005). Child-directed speech, a way of acoustically highlighting lexical items, and repeating sentences (linguistically dress children's early experience with the world), are probably an important help for children to detect word boundaries in the speech stream (see Ecological theory of language acquisition, Lacerda et al., 2004). Early sensory, perceptual and language experience alters the neural properties in the auditory cortical network, referred to as plasticity, which is greatest during the first year of life (Cardon, Campbell & Sharma, 2012; Sanes & Bao, 2009). At one year of life the cortex have developed all its six layers, as a result of an overabundance of synapses (Cardon et al., 2012). This process gradually slows down in visual, auditory and prefrontal cortex, where neurons undergo a pruning phase, that is, extraneous neurons and their synapses are eliminated from the respective sensory system, if not functionally necessary (Cardon et al., 2012). In the following, I will address how a SNHL may affect development of early speech perception.

The effects of SNHL on development of speech perception

Plasticity

Sharma (2002a-c, 2009) and Kral et al. (2006) has contributed to our current understanding of cortical reorganisation in deaf children using CI and the positive effects of early implantation. Using auditory evoked potential paradigms Sharma's studies showed that children who were implanted before 3.5 years of age displayed similar patterns of P1 (an early action potential normally observed 100 ms after onset of a speech stimuli) as typically developing children after 6 months of CI use. The longer the duration of deafness, the more abnormal cortical response latencies to speech were observed in the children. Further, plasticity of the auditory cortical areas was strongly diminished after 7 years of age. Since P1 reflects the accumulated sum of delays in synaptic propagation through the peripheral and cortical auditory pathways, which shortens during childhood development, the diminished delay patterns are a sign of cortical maturation (Sharma, 2002a-c). Mechanisms explaining the diminished plasticity with age are for example cross-modal recruitment of the auditory cortex, that is, visual or somatosensory modalities "take over" the function in secondary areas of the auditory cortex (Dahmen & King, 2007). Further, layer V cortical neurons are affected by early SNHL, possibly diminishing subsequent plasticity capacity (Dahmen & King, 2007).

Another aspect of cortical reorganisation has been observed as a function of training and

experimental manipulations. In Figure 2, Dahmen and King (2007) showed the effect of different experimental manipulations of the cortical tonotopic organisation in individuals with NH, so called distortions, see increased orange area in b) for frequencies of about 15 kHz. Effects have been reported after continuous pure-tone stimulation in infancy, pairing of tones in adults and frequency discrimination training.

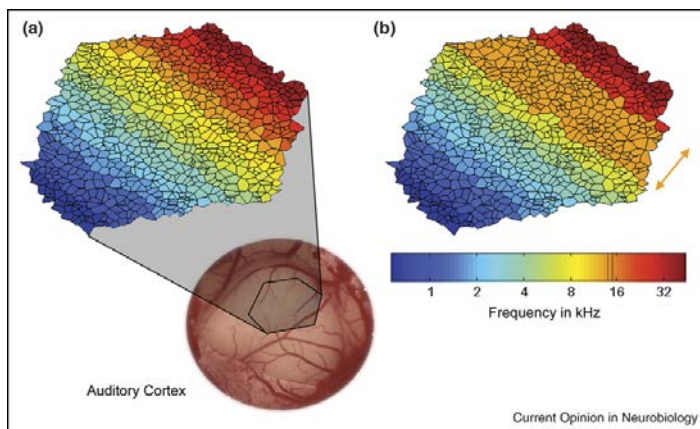


Figure 2. (a) Normal cortical tonotopic organisation for frequencies from 1 kHz to 32 kHz is observed. In (b) the orange area representing 15 kHz is expanded, thus overrepresented after training, (Dahmen & King, 2007). Reprinted with permission by the authors.

Behavioural observations

Infants with SNHL have been observed to discriminate between non-native speech sounds for a longer period of time during development than normally hearing infants do, which is a sign of a more immature initial state – and can be linked to slower language development (Kuhl, 2009). In a review by Jerger (2007) some general observations were made regarding the effects of SNHL on speech perception in children. These included for example problems in processing temporal sequences (detecting inter-stimuli-intervals/formant transitions, for example distinguishing between /ba/ and /da/) and voice onset time (distinguishing between the phonemes p-b, t-d, k-g). Further, Jerger (2007) reported linguistic consequences of speech perception difficulties, for example, in children's ability to acquire semantic and phonological knowledge. These difficulties increase with degree of HL. Eisenberg (2007) reported that in hard of hearing children (2-7 years of age) who used HA, vowels were more accurately perceived than consonant contrasts. Further, consonant voicing and frontal place (labial-

alveolar) were perceived more accurately than manner and rear place (alveolar-palatal). Generally, observations show that the longer the duration of deafness, the slower the onset of the perceptual milestones (not ignoring the 4 months of missed auditory experience in utero). Such early behavioural observations serve as a point of departure in the present thesis, where these aspects are further studied and discussed.

Speech perception may be related to theories of neurolinguistic development in children with normal hearing. According to Locke's theory of neurolinguistic development (1997) there are four individual phases the child passes during the first 36 months of life, which is the sensitive period for language acquisition. These phases overlap and each phase has its own commitment of neural resources (see Cardon et al. 2012). Locke stressed the importance of the second phase (onset at 5-7 months of age) when the child needs to collect and store utterances in a holistic way (compare lexical learning, Nettelbladt, 2007), basically subserved by mechanisms of social cognition, and situated in the right hemisphere (Locke, 1997). These utterances later serve as material for the onset of the analytical stage (20-37 months), when phonology, morphology and grammar develop. If the analytical stage is compromised this might appear as a tendency to treat the incoming speech signal in terms of syllables and consonant clusters (larger chunks), rather than individual phonemes (Briscoe, Bishop & Norbury, 2001). Thus, for example a child's phonological sensitivity may be limited to that the input contains frication and nasality, but the child is not aware as to the exact place in the speech signal, where these features occur. Lastly, I want to address the ambiguity of listening reactions in children having a SNHL (Cole & Flexer, 2011). Although a child might react to sounds in the environment, that is, the hearing allows audibility, it does not imply that the child hears well enough to detect word-sound distinctions or intelligible consonants, which are important aspects in developing speech. This phenomenon is particularly important in the assessment of hard of hearing children. That is, it is preferable that the test leader receives some kind of active response from the child (which more thoroughly reveals what the child actually hears) before the onset of a test session (Cole & Flexer, 2011).

In sum, early acquirement of speech perceptual fundamentals is important for two main reasons: first, it feeds the auditory cortex in both hemispheres during certain time-windowed phases, which sets the foundation for later neural plasticity and reorganisation. Second, these perceptual fundamentals form the building blocks of children's phonological, morphological, lexical and grammatical development.

Speech production

An important early developmental step in speech production for NH children is babbling, which appears in the following sequence: cooing (two months), canonical babbling (6-10 months) and jargon babbling at 10-12 months (Clark, 2009; Wermke, Leising & Stellzig-Eisenhauer, 2007). Jargon babbling refers to when the child varies the intonation contour of the babble sequences to the language around them, which is a consequence of the child's auditory perception being tuned to the stress pattern of the ambient language. During this phase, children also start to vary the syllables within a babble sequence, for example **bababa-mamama** (Clark, 2009). Vowels tend to have more variability than consonants during this stage. First words appear around the child's first birthday, and typically word production and babbling appear in parallel as late as age two or two-and-a half years. Considerable variability in typically developing children's early word forms is often found, and it seems that children construct their own phonology along slightly different paths (Clark, 2009). Generally, early words consist of phonologically unmarked or universal speech sounds, meaning that they are evident in a large number of languages in the world (Nettelblatt, 2007). Further, children tend to be selective as to what words they try to pronounce (regarding both sounds and shapes) which probably has its roots in earlier babbling (Clark, 2009). Until children master the articulatory programs necessary for early word production they substitute (e.g. voice initial voice-less sounds and devoiced final voiced sounds, or use a plosive instead of a fricative), assimilate (e.g., reduplicate the first syllable in a two-syllabic word, **packa- > pappa**, English "doll -> dod"), and omit (e.g., reduction of consonant clusters, omissions of final consonants and omissions of weak syllables; Clark, 2009; Nettelblatt, 2007) sounds in substantially consistent ways. For Swedish normal hearing children the consonant inventory before 4 years of age include voiceless plosives /p, t, k/ and nasals /m, n/, liquids /j/ and the fricatives /v, h/. Between 4 and 6 years of age voiced plosives are established, velar sounds /g, ŋ/, liquids /l/ and the fricatives /f, s/. Further fricatives /fi, ç/ and tremulants /r/ may need further time to be established, but usually are around 6 years of age. Phonemes that are specific for Swedish, and are less common in the worlds' languages, tend to be acquired later (see markedness by Hume, 2010). These characteristics seem to follow Jakobson's early theory (Jakobson, 1968) although a theory of phonematic contrasts claims that ease in production governs the children's early acquisition and the basic sound inventories of the world's languages (Clark, 2009; Nettelblatt, 2007). As for the vowel system, children acquire the cardinal vowels before 4 years of age and regarding suprasegmental development, stress and accent pattern production are established between 4 and 6 years of age (Nettelblatt, 2007), although

awareness of prosodic contrasts, and prosodic integration of sequences of items are usually accomplished as early as 18 months of age (Samuelsson, 2004).

The effects of SNHL on development of speech production

Early signs of severe HL in infants are qualitatively different babbling; for example delayed canonical and jargon babbling (Fagan & Pisoni, 2009). Further observations are limited consonant repertoires, and delayed spoken vocabulary development (Fagan & Pisoni, 2009). Generally, deaf children who receive cochlear implants early (before 3 years of age) show similar patterns of consonantal development as NH children (Spencer & Guo, 2012). In the thesis by Karltorp (2013, p 32) children who received their implants before 9 months of age showed no delay of spoken language development at all and their acquisition of speech intelligibility was faster than those implanted at a later age. Similar findings for children with CI who have been implanted before 2 years of age, have after 12 months of implant use been observed to develop speech production skills comparable to NH children (Mikolajczak, Streicher, Luers et al., 2013).

Blamey et al (2001) used conversational samples from nine children with CI (range 2:3 – 4:9 years of age three months pre implantation) to follow speech production development at nine points in time, the last at six years post-implant. At four years post-implant 90% of the syllables were intelligible to the listeners. For all children, consonants, clusters and words were more difficult to produce than vowels. At six years post-implant, consonant clusters reached a score of 40% correct. A more recent study (Adi-Bensaid & Ben-David, 2010) investigated word initial production of consonant clusters, consonant 1 and consonant 2 (CC, C1 C2) in six children 1:5-2:8 years of age, four months post implant and continuously until the children produced their first initial bioconsonantal cluster. All children received bilateral CI between 1:0-2:5 years of age. No information of their communication mode is given. The study showed that the children with CI followed the same developmental trajectory as the typically developing hearing children. The authors argued for a combinatory explanation of contiguity (deletion of C1 was the most common deletion pattern) and markedness (C2 was deleted in obstruent – liquid clusters) of the consonant deletions.

Wie (2005a) measured language development in the first 100 children in Denmark with CI (4.5 to 20 years of age with at least 2 years of implant use). At the end of the project, 61 of the children could produce enough speech to be tested with a speech test. Results in speech intelligibility rating showed that for these children, 48 per cent had unintelligible speech, and

twenty-two per cent had speech that could be understood by their family members. Further, 30% had intelligible speech for listeners who were experienced in the speech of children with HL. It should be noted that *all* children with CI were included in the follow up, that is, also children with scores below the normative range on non-verbal intelligence tests. In a more recent study, Wie (2005b) examined the expressive language development using naming and repetition tasks, together with parent questionnaires, in 21 children receiving bilateral CI between the ages 5 and 18 months. All children had an auditory-oral or auditory-verbal communication approach. Wie found that 57% of the children at 12-48 months post implantation had expressive scores within the normative range. Karltorp (2013) found that the age when 80 per cent of the consonants were correctly produced (pcc) correlated with time of surgery and that the increase was about 1.2 years per year, thus the developmental pace was slightly enhanced. Further, developmental bursts regarding speech production are more commonly seen in children implanted before 2.5 years of age (Connor, Craig, Raudenbush et al., 2006).

For children with HA with moderate SNHL more vowel substitutions, final consonant deletions and voicing problems, have been observed compared to NH children (Huttunen, 2001). Large-scaled studies of DHH using HA are quite rare and commonly they report expressive language scores only on a general level (Moeller et al., 2000). The study by Borg, Edquist, Reinholdson et al., (2007) serves as an exception. Borg et al. examined the language development in a cohort of Swedish children with HL 4-6 years of age, in sum 156 children (including unilateral, bilateral, conductive HL as well as SNHL). These children were compared to 97 NH children's scores. Increasing language delay was found for children with a HL > 60 dB HL. As for word production, multi-syllabic words were particularly difficult to produce for children with HL. Further, word stress production acuity was investigated. Both groups, children with HL and NH children, found multi-syllabic words with final stress more difficult to produce than words with initial stress. Children with HL performed at a significantly lower level than NH children regarding word initial stress (HL; 84%-61% NH; 97%), and word final stress (59%-31%: NH; 74%).

Consonant production in picture naming tasks has been compared between prelingually deaf children with CI and prelingually moderately-to-severely hearing impaired children using HA at nine years of age (Baudonck, Dhooge, D'Haeseleer et al., 2010). All children had typical non-verbal intelligence. Two-thirds of the children with CI were implanted < 5 years of age and the remaining part were older. The children with HA received their first HA < 2 years of

age. Consonant production contained considerable more phonetic and phonological errors in the children using HA who had a hearing threshold > 70 DB compared to the children with CI. Even in later implanted children consonantal production was advantageous.

In sum, perceptual constraints limit many aspects of speech production development in DHH children. Thus, parts of the speech signal that have a weaker intensity, that is, less acoustical energy are more difficult to incorporate in the language system, for example consonants and weak syllables in multisyllable words. These aspects will be discussed in relation to the empirical results in the present thesis. In children with a more severe HL using HA, speech production is more affected than in children with CI, concerning both phonetic and phonological errors. It is important to consider inclusion criteria in the studies, since speech production usually are more affected in DHH children with additional disabilities.

Output phonology and lexical access

There are two major theoretical contributions to our current understanding about the connections between output phonology, lexical acuity, and lexical access (Levelt, 2001; Ramus, 2001; Ramus & Szenkovits) and both acknowledge their intimate link. The relationship between output phonology, lexical and sublexical representations are shown in Figure 3 (Ramus & Szenkovits, 2008). In Ramus's model the addition of a sublexical representation-level has theoretical implications for children with SNHL and will be further discussed in the section about phonological representations. Levelt's theory of speech production states that each segment in the form-encoding system receives a code, for example horses -> /h, ɔ, r, s/ /ɪz/. These codes form the input for syllabification of the "mental syllabary" (see Frith, 1985 for a mental syllabary in reading), a store of highly practiced syllabic gestures, which in turn form the input for phonetic encoding. An item's syllabification is not stored in the mental lexicon but as Levelt puts it, "created on the fly", and is dependent on the context. Overt speech that targets the phonological word is a string of syllabic gestures, which is called its articulatory scores. The articulatory scores are the output of form encoding, and the final product of lexical access (Levelt, 2001).

Phonological representations

Claessen (2009) defined phonological representations "as the sound based codes associated with words, and the storage of this phonological information in long-term memory". An interesting question arises whether there are different stores for lexical and sub-lexical phonological information, as proposed by Ramus (2001) in the theory of lexical access, and further developed by Szenkovits and Ramus (2005) and Ramus and Szenkovits (2008). The

mental lexicon serves as the core of the model, which is divided into three parts; the semantic lexicon (meaning of words), the phonological lexicon (phonological forms, segmental and suprasegmental information), and the orthographic lexicon (orthographic forms, spelling of words). For the cognitive architecture of these mental representations, see Figure 3. According to the model the phonological lexicon is a *permanent store for word forms* only, whereas the sub-lexical representations is a *short-term storage for whatever speech you hear* that can be represented in phonological format, that is words, whole utterances, and parts of words (phoneme sequences, that is nonwords). The cognitive construct of sub-lexical representation has theoretical implications for children with CI. These children are commonly observed to have quite firm and specified phonological representations of real words, that is, the permanent store in long-term memory, LTM (Carter, Dillon & Pisoni, 2002; Dillon, Cleary, Pisoni et al., 2004; Wass, 2009), but have considerable difficulty with both lower-level and higher-level phonological processing of new words or nonwords, that, is the short-term store. This suggests that the short-term storage of sub-lexical representations is hampered in these children.

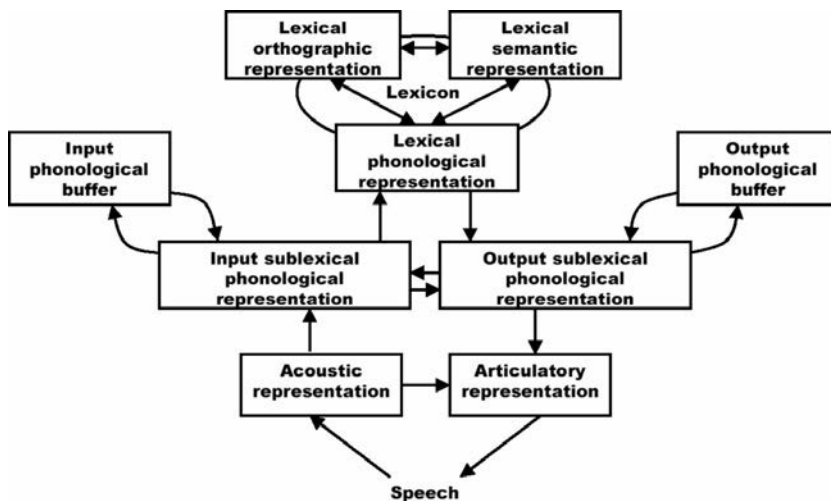


Figure 3. The information-processing model of lexical access including separate input and output sublexical phonological representations (Ramus & Szenkovits, 2008). With permission by the authors.

How well a child recognises the phonological properties of a particular word changes through development and is dependent on when they start reading (Bentin & Leshem, 1993). In the early stages, a holistic articulatory gesture is associated with the meaning of a word (Claessen, 2009). As vocabulary grows the stored code is more segmented, as a consequence of lexical restructuring. At the age of five children show sensitivity to syllabic structure, and at approximately seven or eight years of age, the representations contain phoneme-sized units (Claessen et al., 2009; Liberman, Schankweiler & Fischer, 1974). In children with a SNHL using HA or CI there are several challenges in developing distinct phonological representations. First, fluctuating and impoverished speech perception through childhood makes the encoding into the phonological lexicon difficult. This may be considered an effect of the HL itself *in combination* with restrictions of the technical devices that do not deliver rich enough temporal and spectral information of the speech signal (Nitttrouer et al., 2012). Second, small vocabularies might affect the neighbourhood density (ND) of words in the phonological lexicon (that is the number of words that can be generated by replacing a single phoneme in a target word in a single position, see Chen, Vaid, Boas et al., 2011). As a consequence of reduced ND, the perception of a word's phonemes is decreased. As argued by Zamuner (2009), early perceptual sensitivity aids lexical acquisition, supporting continuity across speech perception and lexical acquisition, (that is, the PRIMIR theory; Processing Rich Information from Multidimensional Interactional Representations).

Phonological working memory

Phonological working memory or the phonological loop is one part of Baddeley's component model of working memory (Baddeley, Gathercole & Papagno, 1998; Baddeley, 2012). It refers to the mechanisms involved in retention of speech based (or speech like) material over a short period of time, typically 1-2 seconds (Henry, 2012). Specifically, it involves a phonological store, which holds information in phonological form, and an articulatory rehearsal process, which refreshes/maintains decaying phonological forms in the store. Experimental studies show that articulation rate, word-length and phonological similarity (particularly when tasks are presented in the auditory mode) influence children's performance through development (Henry, 2012). Typically, children develop the articulatory rehearsal process gradually which usually happens around six or seven years of age (Eysenck & Keane, 2005, p 199; Henry, 2012). Of particular interest to SLPs is 'what happens' during the 1-2 seconds in the phonological store and how these events may act to explain individual differences in clinical populations. Here, the sub-lexical representation level (Ramus &

Szenkovits, 2008) may shed further light to explain the difficulties in phonological WM that children with CI exhibit. As Ramus states (e-mail communication, 2012), the sub-lexical representation level may act as a short-term store of whatever speech the child hears. Thus, language units of different size are contained here. The more “hooks” this speech material may connect to in the lexical representational level in LTM, the easier the child will have in repeating and consequently learning new words. Similar lines of thought are discussed by Tamburelli and Jones (2013) and Wass (2009).

Lexical access

Several factors interact with children’s lexical access ability. In the following some of these will be considered. Some are developmental factors, for example, age of acquisition (AoA), some can be related to the characteristics of the ambient language (phonetic salience, word frequency, WF and ND), and additionally, some are factors within the individual child (language learning capacity, hearing ability and WMC).

The AoA effect refers to the finding that the younger a child is when acquiring a word, the faster the child processes it. The AoA has been observed in different lexical tasks, for example in picture naming, word and speeded word naming, and in lexical decision-making (Juhász, 2005). The phonetic context relates to the effect of the phonological characteristics of the ambient language. Here, degree of salience of the phonetic features and the segment’s position within a word, seem to govern the ease and the speed that words are recalled (Claessen et al., 2009; Snowling, 1994; Zamuner, 2009). Goodman, Dale and Li (2008) reported WF effects in children for open class word production. Thus, children learned words earlier that appeared more frequently in parent’s input. Additionally, Stokes (2010) has contributed to our understanding of the ND and WF effect in toddlers’ lexical production. Low-vocabulary children have been observed to score significantly higher on ND and significantly lower on WF than high-vocabulary children did, that is, slow lexical learners may be extracting statistical properties of the input language in a different manner compared to fast lexical learners.

The ease, with which a word’s phonological properties are processed, has been found to interact with WMC and hearing sensitivity (Classen, 2013; Janse & Newman, 2012). Janse and Newman (2012) found that NH young adults with poorer WMC were supported in their nonword identification by similar sounding items. For individuals with reduced hearing sensitivity (older adults), an effect of ND was found in nonword identification, that is, long-

term linguistic knowledge (larger vocabularies) supported degraded auditory representations. Wass (2009) found that when children with CI were familiar with the words to be accessed and when they received semantic cues, their performance was improved. Wass further argued that when the quality of the phonological representation and the auditory input signal was high enough, children with CI might have relatively efficient processes of lexical access for both speech recognition and output phonology (Wass, 2009). Further, Wass found that some of the children with CI performed on a par with NH children both in accuracy and speed, acknowledging the heterogeneity in this population. Moeller et al.'s review of children with a mild-severe HL (2007) found slower lexical access in these children, suggesting subtle effects of HL on lexical retrieval. In sum, phonological processing and lexical access have a reciprocal relationship through development. This reciprocity will be further discussed in the present thesis.

Visuo-spatial working memory

The visuo-spatial sketchpad is one of the two “slave-systems” in Baddeley’s four-component model of working memory (Baddeley, 2012), which is used for temporary storage and manipulation of spatial and visual information (Eysenck & Keane, 2005). Further division of visuo-spatial working memory is made into *the visual cache* that stores information about visual form and colour, and the *inner scribe*, which is essentially an analogue to the articulatory rehearsal process in the phonological loop (Eysenck & Keane, 2005). Visuo-spatial WM is important in reading development, particularly in fluent reading at later reading acquisition stages (Tobia & Marzocchi, 2014) and in arithmetic’s (Barnes, Raghubar, English et al., 2014). In the present thesis, visual and spatial aspects of visuo-spatial WM are not treated as separate skills.

The development of visuo-spatial WM is highly dependent on an increased amount of grey and white brain matter in the fronto-parietal brain network (Myatchin & Lagae, 2013). Myatchin and Lagae (2013) studied visuo-spatial WM in 6-14 year old typically developing children using neurophysiological and behavioural measures. Behaviourally, maturation was evident in terms of faster response times and increased accuracy scores. Stable levels for easy visual WM tasks (one-back matching) were evident at 11 years of age, and for more demanding visual WM tasks at 14 years of age (two-back matching). In older children, higher engagement in the right hemisphere, that is, lateralisation indicated a more mature fronto-parietal network in visual WM tasks.

Research literature on the development of visuo-spatial WM in DHH children using HA or CI is sparse. Wass found levels in children with CI on visuo-spatial WM tasks comparable to those in NH children (Wass, 2009). Fagan and Pisoni (2007) assessed visuo-spatial skills in children with CI, 6-14 years old and found low average score performance on a group level. Approximately one third of the children showed some difficulty. Visual skills have also been addressed in relation to effects of reduced auditory stimulation during childhood. Fagan and Pisoni (2009) argued that children with CI might weight visual cues more heavily in speech perception tasks, that is, showing reduced bimodal fusion in processing incongruent audio-visual stimuli. Willis, Goldbart and Stansfeld (2014) investigated six children with CI exhibiting difficulties in acquiring spoken language (implanted < 3 years of age) who used spoken language as their primary mode of communication. Their study showed significantly higher scores on visuo-spatial WM-tasks for the children with CI compared to the NH children, used as a comparison group. Willis et al. (2014) suggested that those children's visual skills could be utilized in therapeutic and educational settings to improve language learning. Children with mild-moderate-severe SNHL using HA have been found to have reduced visuo-spatial working memory as assessed by the Corsi-Bloch test compared to NH children (Stiles, McGregor & Bentler, 2012).

Complex working memory

Complex working memory refers to the simultaneous storage and processing of information. When defining complex working memory, there is a need to acknowledge another one of the components in Baddeleys component model of working memory, that is, the central executive. The central executive is a domain-general component responsible for processing of information in dual tasks. The central executive is partly incorporated in the capacity theory of comprehension (Just & Carpenter, 1992). Just and Carpenter (1992) propose that cognitive capacity constrains comprehension, and more in some individuals than in others. During listening comprehension as in the Sentence completion and recall task (SCR-task), used to test complex WM in the present thesis, several different processes occur in parallel. These are, the listener must be able to quickly retrieve some representation of earlier words/phrases in a sentence, and relate them to the word they are to produce, thus use thinking in analogues (to complete the sentence). At the same time, computations like logical reasoning and comparison are performed. Then the child needs to direct his/her attention to the final part of each sentence, that is, the word they themselves have produced, to be able to recall them. Thus, they need to *activate* processing, storage and retrieval. Just and Carpenter's

theory (1992) expressed capacity as the maximum amount of activation available in WM, to support either of the two functions. The central executive that is responsible for language comprehension, controls these processes. In reading studies that have been examining complex WM (or general WM) it is important to consider what type of tasks that have been used, since they might require none or different amount of phonological or semantic processing. In the study by Wass (2009), children with CI were been found to have better complex WM (assessed by the SCR-task), as compared to their phonological WMC. Cleary, Pisoni and Kirk (2000) investigated WM spans as predictors for spoken word recognition and receptive vocabulary in children with CI. A contribution from WM was only seen in the span tasks that incorporated an auditory processing component, thus not specifically linked to a general purpose of WM. In the study by Stiles et al. (2012) children with mild-moderate-severe SNHL using HA showed overall resistant WM systems. Sub-analyses showed a different interaction between Corsi-spans and executive functioning in children with SNHL, that is, children with small spans showed reduced executive functioning and vice versa. This pattern was not observed in children with NH. For both groups WMC was significantly correlated to vocabulary size, that is, better working memory was associated with larger vocabularies.

In sum, complex WM for children using CI might be slightly impaired, but not as much as phonological working memory. Interactions between WMC and other domains, for example vocabulary show its contributory role in language development.

READING ACQUISITION

Reading – a language-based skill

Reading is primarily a linguistic skill that can be traced back at very early stages of language development (Catts, Fey & Proctor-Williams, 2000; Foorman, Anthony, Seals et al., 2002; Lundberg, 2002, 2009; Lyytinen, Erskine, Tolvanen et al., 2006; Nauclér & Magnusson, 1998; Scarborough, 1990). In the following paragraph, I start by acknowledging important language precursors to reading acquisition. I continue by presenting Frith's developmental stages of reading acquisition observed in typical development (1985), although originated several years ago, Frith's theory still has strong influence on current theories of reading development. I end the section by addressing some important differences between spoken and written language acquisition and explaining characteristics in children at risk of developmental dyslexia, which, at the surface, resembles the difficulties that DHH

demonstrate. As will be evident to the reader, I use the term phonological sensitivity, as opposed to phonological awareness. I chose this term since it embraces broader aspects of phonological processing than the strict ability to manipulate the constituent parts of words and sentences.

Essentially, sensitivity to the phonological structure of language sets the foundation for decoding, whereas lexical, grammatical and syntactic knowledge form the basis for reading comprehension (Hulme & Snowling, 2014; Nauclér & Magnusson, 1990; Lundberg, 2002, 2009; Reutzel, Camperell & Smith, 2002). As will be outlined, the development of phonological sensitivity (Stanovich, 2000) starts very early in life and continue to refine as an effect of children acquiring literacy (Lyytinen, Ronimus, Alanko et al., 2007). Thus, increased phonological sensitivity is to be expected as children experience the salient architecture of printed words (Lundberg, 2002). Longitudinal studies, among others, the Jyväskylä longitudinal study of dyslexia (Lyytinen et al., 2006), have shown early differences in the preattentive level of auditory processing in infants at risk for dyslexia and controls (Leppanen, Richardson, Pihko et al., 2002). Consequently, early auditory processing may be seen as one of the earliest precursors of reading acquisition since it feeds into native language speech perception skills, that in turn are interrelated with the development of phonological processing and vocabulary (Fagan & Pisoni, 2009; McBride-Chang, 1996; Zhang & McBride-Chang, 2013). Children are believed to start with holistic representations of adult words that gradually get more enriched and reach more precise detail from the contrasting sound segments (Clark, 2009). The same developmental trajectory is believed to be true for children's early utterances, that is, children are first able to interact in conversation with their environment with the use of *formulaic* utterances and “holistic phrases not subjected to grammatical analyses” (Locke, 1997). Similarly, in reading acquisition children start out with an holistic approach towards written language (Lomax & McGee, 1987), for example, recognising traffic signs or logos (graphic awareness), and gradually obtain more fine-grained knowledge about print, such as being able to recognize the initial letter sound in their own name (phonemic awareness) and being able to sound/spell out words of objects in their own drawings (phoneme-grapheme correspondence).

Locke's theory of neurolinguistic development (1997) has been described in the section on speech perception and is further elaborated here, due to its close links to children's analytical development. When approaching the second birthday, children reach the analytical third

phase believed to be time-locked between the age of 20-37 months. During the analytical stage n cortical areas in the left hemisphere that are predisposed for grammatical and phonological mechanisms, reorganize. Grammatical development and analysis revealed at the morphological level in spoken language, depend on a successful earlier second phase (accelerating from 5 months up to 20 months). Here, the storage of utterances/lexical material (by listening to the speech of others) take place. Further, when the grammatical mechanisms set in, this works as a springboard for the child to achieve a much larger lexicon. At this time point in time children are increasingly more capable to analyze the words in the speech signal with respect to their including parts. The rationale of the theory is that later lexical, grammatical and reading acquisition will be affected if there is something preventing optimal conditions particularly in the second phase. This may either occur as a consequence of dispropriate stimulation or be caused by internal factors. As studies on DHH children indicate, *early language experience irrespective of modality input* is the main important factor to successful language and reading acquisition (Geers, 2003; Leybaert, 1998; Leybaert & D'Hondt, 2003).

Frith's (1985) theory of reading acquisition which was published in an influential paper on developmental dyslexia, has helped our understanding of the phase-like manner typically developing children learn to read. Although tested and modified throughout the years (Stuart, 1988) the theory's basic claims that reading acquisition occurs in three phases identified by three strategies, still hold today. These are the *logographic*, *alphabetic* and *orthographic* phases, the strategies of which now will be explained. The *logographic* strategies refer to the instant recognition of a known word. Letter order is largely ignored and phonological factors are entirely secondary. Guessing based on contextual factors are common in this stage. The *alphabetic* strategies are analytical. Here the child uses knowledge in phoneme-grapheme correspondence to decode words. Letter order and phonological factors play a crucial role. Alphabetic skills enable the child to decode unfamiliar words and nonword. The *orthographic* strategies develop last, they involve instant recognition of words into orthographic units that correspond to a limited set of morphemes. The morphemes are mentally represented by abstract letter-by-letter strings and may be used in analogy with a syllabary (compare Levelt's theory of speech production, 2001) to create an almost unlimited number of words. The orthographic strategies are characterized by being analytical and non-visual (as opposed to the logographic) and by operating on larger units and being non-phonological (as opposed to the phonological). In sum, the analogy between language and reading acquisition is, that both

abilities start out on larger units and gradually, through development, enable the processing of smaller units, in a more refined and detailed manner. In skilled reading, language (that is, phonological, morphological, semantic-syntactic and pragmatic competence) and visual processing (that is, the formation of abstract letter and orthographic representations) are used flexibly to derive meaning from print (Bavelier, Green & Seidenberg, 2013; Bitan, Manor, Morocz & Karni, 2005; Bjaalid, Høien & Lundberg, 1997; Byrne, Coventry, Olson et al., 2009; Castles et al., 2009; Rastle, 2007). Thus, the reader must both be able to do analytical, detailed translational work to decode unfamiliar words, as well as have a large enough orthographic lexicon that makes immediate, instant recognition of words possible (see dual-route models of reading, Stanovich, 2000).

Written language – connecting visual input with language

The basic principle of the alphabetical written languages is that each letter in the alphabet has a sound value. The point is that written – as well as spoken – language has a certain set of graphemes/phonemes which in isolation usually do not hold any meaning (although there are exceptions, in Swedish for example ‘i’ = in), but put together can create an unlimited number of combinations with meaning (Melin, 2004). In learning to read, it is therefore of the utmost importance that children learn the letters of their language (Caravolas, Lervåg, Defior et al., 2013; Hulme, Bowyer-Crane, Carroll et al., 2012), grasp the alphabetical principle at an early age, understand how to blend letters into syllable and words, which with enough practice and tutoring make them become automatized fluent readers (Lundberg, 2002). It is at the letter and word level that phonological and lexical skills merge with the visual components of language, which is acknowledged in, for instance neuroimaging studies (Parviainen, Helenius, Poskiparta et al., 2006) as well as in dual-route models of reading (Stanovich, 2000).

Children at risk for developmental dyslexia

One aspect that differs between spoken and written language acquisition is that the majority of children develop reading through instruction (Richardson & Lyytinen, 2014). Approximately 20% of beginning readers struggle to develop reading despite adequate opportunities, and without having any severe sensory or cognitive deficits (Richardson & Lyytinen, 2014). When children grow older and continue to show difficulties with reading, a proportion of them, between 5-10% (Siegel, 2006), will be diagnosed with developmental reading disorder, dyslexia. The most common and valid explanation of dyslexia is the

phonological deficit hypothesis (Rack, Snowling & Olson, 1992). The phonological deficit hypothesis implies that the majority of children with dyslexia have difficulties with phonological coding, which in turn is dependent on phoneme sensitivity, letter-sound mapping, name encoding and verbal memory (Ramus, 2001, Vellutino, Scanlon & Tanzman, 1998). The most efficient remedial methods for children with dyslexia are those that have a phonics approach (Galuschka, Ise, Krick et al., 2014; Hatcher, Hulme & Snowling, 2004; Richardson, 1984; Snowling & Hulme, 2012). Typically, phonics approaches contain three main ingredients; phoneme-grapheme correspondence training, blending of phonemes to syllables and words, and segmenting words to their constituent phonemes (Phonicsplay, 2008).

Reading acquisition in DHH children

The heterogeneity among the DHH children across a variety of domains complicates the picture of how reading development is accomplished in this population (Powell & Wilson, 2011). Studies show that subsets of the DHH children seem to close the gap to NH children, especially those who have benefitted from early auditory stimulation and particularly in the younger school years (Easterbrooks & Beal-Alvarez, 2012; Geers, 2003). Still, other findings show that DHH children seem not to reach reading levels on a par with their hearing peers regardless degree of HL and early use of CI (Marschark, Sarchet, Convertino et al., 2012). A challenge for DHH using HA or CI is that the HL itself prevents them to perceive the phonemes clearly which would have negative consequences for reaching fine-grained levels of phonological processing skills (Nittrouer et al., 2012) and consequently, for developing efficient phonological decoding strategies. Wass et al. (2009) and Park, Lombardino and Ritter (2013) speculate that this qualitatively different phonological sensitivity may lead to changed reading strategies, in favour of visually based decoding.

In sum, at the surface DHH children share similar phonological weaknesses as children with developmental dyslexia. This motivated the use of an intervention program that had a phonics approach, that is, to supply DHH children with training ingredients previously found to be effective in children with dyslexia (Richardson & Lyytinen, 2014).

COMPUTER-ASSISTED INTERVENTION METHODS

In this section I begin by addressing some general issues regarding cognitive and linguistic intervention. Table 1 gives short presentations of Swedish computer programs for language, reading and cognitive intervention. What these programs have in common is that they have been developed to support specific clinical populations (for example children with speech impairment and children with attention difficulties) and have been evaluated scientifically and/or clinically. Finally, I present our choice of computer program, Graphogame, and some arguments as to why we selected this intervention program for the DHH children in the present study.

Cognitive and linguistic intervention: general issues

Transfer

Performing the same cognitive or linguistic task repeatedly leads to improved performance on that particular task in most cases (test-retest effects). The important question is the extent to which training with a particular task can be generalized to non-trained tasks. The level of transfer can be graded from: (a) transfer within the same domain (e.g., WM) but to other stimuli and a different response mode; (b) transfer to other cognitive constructs (e.g., from WM to non-verbal reasoning); and (c) transfer to everyday behaviour (Klingberg, 2010).

Persistence

Short-term effects are often noticed in intervention studies; thus, we may observe increased improvement immediately post intervention. However, considering the amount of effort and time that parents, children and therapists put into engaging in training, the ultimate goal is to demonstrate long-term effects. Thus, follow-up tests should be included to evaluate whether the extra training is worth the effort.

Table 1 presents eight Swedish computer-programs for different groups of children in need of intervention regarding speech, phonology, reading, and cognition. The programs have been developed within research groups as well as in clinics, and the training has been carried out either in school or in a clinical setting.

Table 1. Swedish computer programs for speech, language, reading and cognition

Area	Program	Aim	Evaluation
Speech	<p>ARTUR, the ARTiculation TUtoR (Engwall et al., 2006)</p> <p>Box of Tricks (Öster, 2006)</p>	<p>Serves as a speech training aid. Uses three-dimensional animations of the face and mouth to give feedback on the difference between the user's deviating pronunciation and a correct pronunciation.</p> <p>Supports children between 4-10 years of age with speech- and hearing impairment. Displays articulation features graphically so-called speech pictures, intended to help the children become more aware of his/hers own articulation. Uses pictures to support spoken words and has a pre-recorded training vocabulary. The exercises work together with recognition through a phoneme-based comparison of the child's production with the production of a reference speaker. A typical training period lasts three months.</p> <p>Supports phonological awareness. Enables sound-based training on the phoneme, word segment and word level (Fälth, 2013). Picture-based exercises for rhyme, addition, position and segmentation. A typical training period consists of 8-10 hours of training during a 6-month period (Fälth, 2013).</p>	<p>Interviews with small samples of language-impaired children who have practiced with the program. Has not been assessed regarding intervention effects.</p> <p>The Swedish version has not been clinically evaluated (Öster, 2006, p 175) but the two other European versions have, with varied outcomes regarding speech intelligibility improvement.</p>
Phonology	<p>COMPHOT (Ferreira, Gustafson & Rönneberg, 2003)</p>	<p>Supports children with phonological disorders in the ages 3-8 years. Inspired by the Nuffield approach for children with dyspraxia (Williams, 2009). Uses a picture alphabet to support expressive phonology and phonological awareness. It delivers listening and production exercises at the phoneme, syllable, word and phrase level. The child's own recordings are used to enable auditory feedback on the child's pronunciation. A typical training period includes 5-6 times of practice/week 20-30 minutes per session for six weeks.</p>	<p>Observed to have positive effects on reading ability and phonological awareness in children with reading difficulties (Gustafson et al., 2007).</p>
Reading	<p>Talkwise (Pratvis; von Mentzer, 2008)</p> <p>Omega IS (Heimann et al., 2004)</p>	<p>Supports children with phonological disorders in the ages 3-8 years. Inspired by the Nuffield approach for children with dyspraxia (Williams, 2009). Uses a picture alphabet to support expressive phonology and phonological awareness. It delivers listening and production exercises at the phoneme, syllable, word and phrase level. The child's own recordings are used to enable auditory feedback on the child's pronunciation. A typical training period includes 5-6 times of practice/week 20-30 minutes per session for six weeks.</p> <p>Initially supported the communicative ability in children with autism. Targets children in the early school years to create sentences by clicking on text-buttons with words. Provides auditory feedback from words/sentences. Animations illustrate the meaning of a sentence. Delivers top-down training in a three-way-interaction between the child, the teacher and the computer. Typical training periods include 7-8 hours of training, 15 minutes per session during half a year in the children's educational setting.</p>	<p>Talkwise is still under development (Svensson, 2011). Has been clinically evaluated in children with phonological disorders (NH and CI). Positive effects were seen on expressive phonology and speech intelligibility in the majority of the children.</p> <p>Has been shown to improve reading skills on short and long term in school children with reading difficulties (Heimann et al., 2004; Fälth, Svensson & Tjus, 2011; Fälth et al., 2013).</p>

		<p>Studies who have used the program delivered the training in parallel with special instructions at the special pedagogue (Fälth et al., 2013). Has been adapted for deaf children by adding video-clips with sign language (Omega IS-D), and is currently in use in a research project for deaf children at Linköping University (Holmer et al. ongoing).</p> <p>Delivers training with letters, morphemes, words and text in conjunction with auditory feedback for children in the early school years. Typical training periods include 7-8 hours of training, 15 minutes per session during half a year in the children's educational setting.</p>	<p>Gustafson, Ferreira & Rönnberg (2007) reported statistically significant effects on children's orthographic skills after working with the program. Thus, transfer a) has been observed.</p>
	<p>DOT (Ferreira, Gustafson & Rönnberg, 2003; Gustafson, Ferreira & Rönnberg, 2007).</p>	<p>Uses flashcard exposure to support reading fluency later school years (11-17 of age). A word/nonword is spoken out loud using synthesized speech followed by a fixation dot on the middle of the screen. The words' morphemes or syllables are flashcard exposed on the screen, with the beginning of the word placed where the fixation dot was. Finally, a marker is displayed on the screen Here, the student writes/spells the word. A typical training period consists of two-to three training sessions a ' 15-20 minutes per week during three months.</p>	<p>Children with reading difficulties have practiced with the program. These children's reading progress was higher compared to the reading progress in an average student during the same period of time.</p>
	<p>Read-write (Johansson, 2010)</p>	<p>Aims to improve WM capacity in children with ADHD. The children in the original study practiced with the program 15-20 minutes on a daily basis for 4-5 weeks. Training sessions typically included visuo-spatial tasks, backwards digit-span tasks, letter-span tasks and choice reaction time tasks.</p>	<p>Training effects were observed on the trained tasks as well as on non-verbal complex reasoning and non-trained visuo-spatial tasks. Robomemo has been used in several different clinical populations (Söderqvist et al., 2012, Thorell et al., 2009). However, a recent published meta-analysis (Melby-Lervåg and Hulme, 2013) that included 23 studies of WM training concluded that WM training programs (including Robomemo) appeared only to produce short-term, specific training effects that did not generalize.</p>
Cognition	<p>Robomemo (Klingberg, Forssberg & Westerberg, 2002)</p>		

Graphogame intervention and the present study

Graphogame is an internationally established program aimed to support phonemic differentiation and phoneme-grapheme connections in children with dyslexia (Lyytinen, Erskine, Kujala et al., 2009; Richardson & Lyytinen, 2014). Graphogame is developed within the Jyväskylä Longitudinal Study of Dyslexia, JLD (2012) in cooperation with the Agora Research Institute at Jyväskylä University. The JLD-studies target different areas, for example early auditory-phonetic and linguistic skills, auditory and speech perception, and cognitive, language and phonological skills. Several studies have investigated the effects of Graphogame (Brem, Bach, Kucian et al., 2010; Lyytinen, Ronimus, Alanko et al., 2007; Lyytinen, et al., 2009; Kyle, Kujala, Richardson et al., 2013; Saine, Lerkkanen, Ahonen et al., 2011). The rationale behind the choice of program was that the originators acknowledge speech perception difficulties as one of the key issues in dyslexia (Lyytinen et al., 2009). Consequently, much effort has been put into the delivery of an optimal sound quality during practising. Moreover, focus in the Graphogame training is largely on phonemic differentiation, which is positive for DHH participants who have difficulties perceiving the phonemes clearly due to the HL and due to limitations of their technical device (Bouton, Serniclaes, Bertoni, et al., 2012) Graphogame intervention has proved effective for phonological skills, thus, transfer from phoneme-grapheme correspondence training and decoding exercises to phonological sensitivity has been observed (Kyle et al. 2013). Additionally, neural-level effect on written language processing has been observed (Guttorm, Alho-Näveri, Richardson et al. 2011), that is, posterior areas of the brain that specialize in visual processing, including written language, have been significantly activated while purely training with letter–sound correspondences. Further, electroencephalography (EEG) recordings have revealed that learners’ brains could differentiate two types of visual symbols very briefly after visual presentation of either written words or symbol strings. Additionally, Graphogame intervention has had persistent effects on several reading accuracy measures at one-year follow-ups from grade 2 to grade 3, as compared to regular reading intervention and mainstream reading groups (Saine et al., 2011). Further, Graphogame is delivered by means of the Internet, which facilitated training not only in the DHH children, who were spread over a relatively large geographical area in the present study, but also in children in general. Finally, Graphogame was the only available program that combined a phonics approach to reading in combination with exercises for phonological sensitivity.

To conclude, several computerized program have been used for children with reading and language related problems in Sweden. However, only a few of them have been used with DHH children. Graphogame was the computer program that to the greatest extent met the needs of the DHH participants.

GENERAL AIMS

The aim of the present thesis was to implement an intervention method in DHH children to support phonological processing skills and reading, a method that up till now has been exclusively directed to children with NH. The aim was further to study the effects of the intervention in DHH children compared to a reference group of NH children.

Specific aims

The following more specific research questions were addressed in the studies:

1. What characteristics do we find in deaf and hard of hearing children's phonological processing skills/reading ability compared to children with normal hearing?
2. What are the effects of a computer-assisted reading intervention with a phonics approach on phonological processing skills/reading ability in deaf and hard of hearing children using cochlear implants, hearing aids, or a combination of both, and in a reference group of children with normal hearing?
3. What cognitive factors and demographic variables are associated with phonological change/reading improvement in the groups?
4. How does segmental and suprasegmental characteristics in nonword repetition in children with bilateral cochlear implants and in children with normal hearing connect to nonword decoding in reading?

METHOD

Methodological challenges

In the following section I will address some methodological issues that are important to take into account when planning and conducting studies in DHH children with CI or HA. I will do this in relation to the methodological challenges faced in conducting empirical research studies on a small, heterogeneous clinical group, such as DHH children, and particularly when performing intervention studies in this group of children. For more general aspects regarding methodological issues that are connected to the characteristics of the population the reader is referred to Wass (2009) or Löfkvist (2014).

During the last decade an increasing number of theses have been published on communication and language ability in DHH children, where the majority of the children have used CI or HA (Asker-Árnason, 2011; Holmström, 2013; Ibertsson, 2009; Sandgren, 2013; Löfkvist, 2014; Wass, 2009; Öster, 2006). However, compared to other clinical populations, for example children with reading difficulties or ADHD, the number and consequently also the knowledge, is still quite sparse. When the level of knowledge in an area is low, in combination with heterogeneity of the population, research approaches based on more descriptive research questions may be the most fruitful way to go in the initial stages. Following this, hypothesis-testing designs may be employed.

Tests and procedures

One important methodological issue, and a consequence of this relatively unexplored research area, has been the lack of standardized and comprehensive tests and procedures that have been possible to apply in DHH children. This has driven the development of new tests or use of relatively new analytic procedures. This has also been the case for research within child language at large in Sweden, although, that area has a longer tradition. One example is the SIPS, *the Sound Information Processing System*, (Wass, 2009). The SIPS is a computerized test platform that assesses different aspects of cognition, for example, working memory. The SIPS was developed by Wass (2009) and is assessed in a total number of 146 children, whereof 28 children with CI. The SIPS is now widely used among researchers in child language in Sweden. Similar patterns of performance in different clinical groups have been observed repeatedly, which indicates that the test constructs are reliable. For example, the Nonword repetition test has been used among children with language impairment as well as

among deaf and hard of hearing children (Kalnak, Peyrard-Janvid, Forsberg et al., 2014; Pfändtner & Wallfelt, 2013; Wass, 2009).

Further examples of new procedures that have been used to study DHH children and teenagers is for example the key-stroke logging program, ScriptLog, (Asker-Árnason, 2011; Ibertsson, 2009), the referential communication task (Ibertsson, 2009), and mobile eye-tracking (Sandgren, 2013).

In the present thesis the SIPS (Wass, 2009) served the purpose to assess different aspects of working memory, phonological processing skills, and lexical access. Via five tasks (seven measures) of phonological processing skills, a phonological composite score was constructed. Five measures were from the SIPS and two (per cent words and per cent consonants correctly produced) were from a test for output phonology (Hellquist, 1995). The rationale behind the composite score was theoretically motivated and generally might be viewed as assessing different aspects of children's phonological representations (Ramus & Szenkovits, 2008), for example, phoneme discrimination, speech production, and decision-making about the phonological structure in nonwords, that is, higher-level phonological sensitivity. Also statistically the phonological composite score was motivated. Pearson's correlation coefficient showed moderate to strong correlations between normally distributed units (Nonword Repetition, pcc, Nonword Discrimination, and Phoneme Identification; $r = .50 - r = .74, p < .01$ for all correlations) as well as moderate to strong correlations between units violating assumptions of normality, as tested by Kendall's Tau-b's correlation coefficient (Phonological representations, Nonword Repetition, pnwc, Phoneme-test, pwc and pcc; $r = .51 - .91, p < .01$ for all correlations). The variables of the composite showed high internal consistency (Chronbach's alfa = .86, average $r = .49$ mean $r = .49$), suggesting they measured a similar construct. With the use of the phonological composite score, overall group-comparison with NH children's performance was possible. The phonological composite score served as the dependent variable in Studies I and II.

Further tests have been used in slightly new ways in the present thesis. The SCR-task (Wass, 2009) was used to test complex working memory as it is originally aimed for, but was extended to form a measure of lexical access and semantic organisation. Since the SCR-task uses pre-recorded sentences it taps children's speech recognition skills. In the SCR-task children's verbal responses, their completions, were analyzed. Lexical access as measured with the SCR-task was used in paper II and III.

In the nonword-decoding task (TOWRE; Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne, 2009) additional levels of analyses were entered. These were, per cent phonemes correctly decoded and four categories of children's decoding errors: phoneme insertions, phoneme deletions, phoneme substitutions and lexicalisations. These new levels of analysis were used in paper IV.

Heterogeneity

One interesting and demanding aspect in conducting research among DHH children, who use CI or HA, is the large variation in cognitive and linguistic performance. The ambition in the present intervention study was to include as many DHH children as possible, and to offer the intervention despite different background variables. Thus, possibly, the characteristics of the present sample reflect the characteristics in DHH children at large in the "real world". In the present sample all children had a SNHL, they used their hearing devices continuously on a daily basis, they used spoken Swedish in their educational setting, they performed within the normal range on a test for non-verbal intelligence, and they were all 5, 6 or 7 years of age. Thus, five variables were in common for the participants. But, the following variables differed between the children; age at diagnosis, age at implant/hearing aid, kind of technical hearing device, cause and degree of hearing loss, communication mode used at home, and reading level pre intervention. These aspects are worth considering, because they may have affected how the children were able to perform the training and also how they were able to appreciate the intervention. For example, a child with a deaf parent might have had problems benefitting from the program to the same extent as a child with normally hearing parents, since it is difficult for a deaf person to give the same support as a normally hearing person in that setting. Other examples related to children's communication mode might also have affected the performance. For example, sign interpreters were present during testing sessions to translate the test instructions for children who used sign language in their homes. With the use of sign interpreters, test sessions were prolonged, which may have challenged these children's endurance. Aspects related to reading ability need also to be considered. For example, a reading child might have used their orthographic skills to compensate for a degraded auditory percept, and consequently might have disregarded the auditory signal aimed to support phonological processing. How these aspects affect the results are unknown to us as researcher. Therefore, we need to be careful in drawing general conclusions about the effects of the intervention, and we need to consider these aspects in future studies. Additionally, large heterogeneity implies that overall group results should be treated with great caution.

Speech perception

Speech perception scores were only available in a small number of the children due to difficulty obtaining these scores from the Audiological clinics. However, considering the ambiguity of the hearing loss (Cole & Flexer, 2011) all children's audition was carefully examined before starting the test session by first asking the parents concerning the status of their technical hearing device. Second, checking of children's hearing and speech recognition was conducted by auditorily presenting a sentence from the external loudspeaker for them to repeat. The volume was adjusted according to the child's answer, that is, when a child expressed that he/she found it hard to hear, the volume was increased to ensure a comfortable audible level for each individual child. Further, up till present days there is a lack of normative data on speech recognition tests in DHH children, which leaves us uncertain to tell in detail how these children actually discriminate between speech sounds in words. However, an ongoing project is currently aiming to close this gap by developing a Swedish word discrimination test involving discrimination of minimal word pairs where discrimination, identification and production are targeted (Nakeva von Mentzer, Hällgren, Hua et al., ongoing).

Intervention setting

When performing the training, the inventors of Graphogame highly recommend children to use headphones to enable clear and rich auditory information. In the present study, the DHH children could not do so because of possible interaction with their technical hearing devices. Optimally, in future studies, specially designed headphones could be a solution to obtain as good speech signal as possible. But, as our goal was to offer as ecologically valid a study as possible, this was not a reasonable way to proceed. Thus, children were advised to do their training in the same manner as they normally used a computer, and all children decided to practice with external loudspeakers.

In sum, different aspects have challenged the conduction of an empirical high-quality research study. The goal however, has been to treat the data with caution, and to see the results as pointing in certain directions rather than making generalisations to other samples than the one studied. Further, interesting results have been obtained which may in the future, form an inspiration to sharper hypothesis testing. For example, what does it mean that letter naming skills drives phonological change? What are the mechanisms behind such a result? In future

studies optimally randomized control designs should be used so that maturation and test-retest effects are more carefully controlled for. In the present thesis, it was not an option to put children on a waiting list or deny children training. The ambition was to offer the intervention to as many DHH children as possible and study the effects on this specific group.

Participants

A total of 48 children, 5, 6 and 7 years of age participated in the studies. There were 32 DHH children (20 girls, 12 boys) using CI (n=11) or HA (n=15), or both in combination (n=6), and 16 children with NH (5 girls, 11 boys). Nineteen of the children had a severe/profound HL with a Pure Tone Average (PTA) at 70 dB Hearing Level or more unaided. Eleven had a moderate HL and two had a mild HL (PTA 34). All children participated in Studies I-III. Children with bilateral CI and individually age-matched children with NH participated in Study IV.

The inclusion criteria for DHH were that they should have a mild, moderate to severe, or profound bilateral SNHL and be full time users of CI and/or HA. They should perform within the normative range on nonverbal intelligence measures. No other disability that could affect their speech, language or cognitive development should be present. They should speak Swedish at preschool or school, but could use another language at home. Deaf and hard of hearing children were recruited from the Audiological clinic at Karolinska University hospital in Stockholm and from the Audiological clinic at Lund University hospital. Just below forty families accepted the invitation and were given written and spoken information about the study. One child with bilateral CI withdrew after the first testing due to difficulties for the parents to travel to the EEG-laboratory at Stockholm University. Additionally, three families with a child with bilateral CI, one child with bimodal hearing, and two children with HA had approved to participate, but withdrew since they could not be offered the intervention until next term.

Children with NH of the same age constituted the reference group. The inclusion criterion for the reference group was normal hearing ascertained at the regular hearing screening at 4 years of age and reported by their parents in a written consent form. They should perform within the normative range on nonverbal intelligence measures. They should speak Swedish in their educational setting and have no disability that could affect their speech and language development. Children with NH were recruited from preschools and schools in and outside

the city of Stockholm. Two children with NH were excluded due to too little computer-assisted training.

Group comparisons

Group comparisons were made according to children's hearing status (DHH and NH), according to children's technical hearing device (CI, n = 17 and HA, n = 15; bilateral CI, n = 11 bilateral HA, n = 15 and bimodal hearing, n = 6), according to age (5-year olds, n = 20, 6-year olds, n = 15 and 7-year olds, n = 13), due to a median split according to the children's initial PhPS (skilled and less skilled DHH children; n = 16, n = 16). Additionally, reading children were analysed specifically in Study III (reading children, n = 31; NH, n = 12, DHH, n = 19).

Aetiological and hearing background

The aetiology of HL was hereditary in 15 of the children (one child had Jervell-Lange-Nielsen syndrome) and unknown in 14. Cytomegalovirus (CMV) (1 child) and toxicological exposure (1 child) were the causes of the known non-hereditary HI. One child had been regarded at risk for HL at the neonatal screening procedure. The mean age at diagnosis was 1 year and 7 months, with a variation from 0 weeks to 5 years. Half of the children were diagnosed before or at one year of age. Seven children were diagnosed > 2 years of age, thus had a progressive HI. One of these children was born deaf on one ear and developed a progressive HL on the other ear. The mean age of receiving HA was 2 years and 8 months (n = 21, bilateral HA and CI/HA) and the mean age of receiving CI (n = 11) was 1 year and 8 months. All children with CI were, as is routine in Sweden, fitted with bilateral conventional HA after the diagnosis of HI. Children with bilateral CI had used their CI for at least 32 months (M = 60, SD = 16.3). Mean age at diagnosis was 10 months (range 1-19). Mean age at implant was 20 months (range 8-39). Seven of the children with bilateral CI were prelingually deaf (diagnosed before 12 months).

Support by SLPs

Approximately 75% of the children went for controls/received speech-language therapy. None of the participating children received therapy at the time of intervention.

Communication mode and educational setting

Four of the DHH children had another spoken language besides Swedish, two children used sign language at home as their main mode of communication and used spoken Swedish at school. Three children used sign to support their spoken language. One child had another language background and was exposed to the Swedish language at one year of age. Twenty-

four children were mainstreamed and eight children attended special classes for children with HL. No child was receiving speech therapy during the study except for regular controls at the Audiological clinic.

In the reference group of children with NH there was one child who spoke another language besides Swedish.

Table 2. Study design

Assessment	Baseline 1 (B1)	4 weeks	Pre intervention (B2)	4 weeks	Post intervention (P1)
Cognition and language	Phonological processing skills Lexical access Letter knowledge		Same as at B1 Complex WM Visual WM Non-verbal intelligence Reading	Computer-assisted reading intervention with a phonics approach 10 min/ day	Same as at B2
Neurophysiological measures			Event Related Potentials: Mismatch negativity (MMN) and N400		Same as at B2

General procedure

The data collection for the four studies was made between February 2010 and June 2011. The computer-assisted training was performed during three periods: during spring 2010, autumn 2010 and spring 2011.

Test procedure

Forty-two of the children who were residing in the Stockholm region were tested by the first author in all studies, an SLP at all three test points, in total 126 test sessions. Six of the children who were residing in the Lund region were tested by two other SLPs (one SLP tested five children and the other SLP tested one child), in total 18 test sessions. All SLP's had extensive experience of testing children with HL. Similar test procedures were ascertained through a mutual test administration checking before the onset of the study. At B1 the children were given three testing options: at home, at school or at the clinic. B2 and PI were administered in a sound proof room at the Linguistic Department at Stockholm University, where the majority of the children came with their parents for EEG-recordings. Six DHH children were tested at the Humanities Laboratory and Department of Logopedics and Phoniatics at Lund University.

Baseline 1 included eight tests. One break was given after the child had completed four tests. The B1 test session lasted approximately 50 min. B2 and PI included 14 tests. Eight of these tests were the same as in B1. One pause was given when the child had completed eight tests and additional pauses were given when the child requested so. Since the children also came to have EEG-recordings at B2 and PI, EEG recordings shifted with the tests for language and cognition. Typically two children came for testing in the morning. One child started out with the EEG-recordings and came for the cognitive tests afterwards, and vice versa. For the design of the study see Table 2.

Each child had the test order on an individual paper for him/her to follow, thus served as visual support. After each task was completed, the child made a cross beside it. Instructions were presented orally for all children. When a child needed more detailed instructions, the examiner gave additional explanations. A sign language interpreter was used in two cases for children who used sign language at home. With the use of a sign interpreter the duration of the test session was slightly prolonged. The computerized tests used were selected from the Sound Information Processing System, that is, SIPS (Wass, 2009). All of the tests selected

from SIPS were auditorily presented through two external loudspeakers (Logitech S-100). These were placed on each side of a portable laptop computer with 38 cm screen (1024 x 768 pixels). Before testing, the volume of presentation was adjusted to a comfortable level for each individual child. Additionally, the examiner asked the parent or the child whether the technical hearing device was working properly. To assure that the child could hear the speech stimuli, they were asked to repeat a short initial part of the Sentence Repetition Test from the SIPS (Wass, 2009). The volume was adjusted to fit each child's request of proper hearing. The children's oral responses in the tests were recorded on the computer through the microphone of a Sennheiser headset, using Audacity recording software version 1.2.6 for later transcriptions and/or analysis.

Assessment methods, analyses and scoring

All tests used in the studies are presented in Table 3, and described in more detail below.

Phonological processing skills

The phonological composite score is described first, followed by each constituent measure.

A *phonological composite score* was calculated by a unit weighted-procedure, that is, each unit was calculated in per cent accurate, and then summarized to a global score. Seven measures from five tasks of phonological processing skills presented below constituted the phonological composite score. Measures were: 1. Nonword repetition (per cent nonwords correct; pnwc), 2. Nonword repetition (per cent consonants correct; pcc), 3. Phoneme test (per cent words correct; pwc), 4. Phoneme test (pcc), 5. Phonological Representations, 6. Nonword Discrimination, and 7. Phoneme Identification.

A *phonological change-score* was created for Studies I and II. The phonological change-score between the two time periods was calculated by subtracting the phonological composite score at B1 from the score at B2, followed by subtracting the score at B2 from the score at PI.

A *Nonword Repetition test* was used to assess phonological working memory (SIPS; Wass, 2009). In this task, the children were asked to repeat individual 3–4 syllable nonwords. Children's performance was audio recorded. In Studies I and II children's performance was scored in two different ways: 1) binary scoring: per cent nonwords correct (pnwc). Here, the child received a score of 1 if no alteration of the phonological structure was made; ex. /drallabelli/ -> /brallabelli/ was scored zero, and 2) per cent consonants (pcc) correctly reproduced.

Table 3. Cognitive tests at B1, B2 and at PI

Area		Test	Quantification ³
Non-verbal intelligence		Raven's colored matrices ¹ (Raven, 1995)	Percentiles, raw scores
Phonological processing skills		Nonword repetition, NWR (SIPS; Wass et al., 2008)	Per cent nonwords out of 24 (pnwo) ² Per cent consonants out of 120 (pcc) ²
		Phoneme test (Hellqvist, 1995)	Per cent consonants correct out of 207 (pcc) ² Per cent words correct out of 72 (pwo) ²
		Phonological representations, (SIPS, Wass et al., 2008)	Per cent responses correct ² (max = 18)
		Nonword discrimination, accuracy and latency (SIPS, Wass et al., 2008)	Per cent correctly discriminated pairs of nonwords ² (max = 8) Mean response latency (ms)
		Phoneme Identification, accuracy and latency, (SIPS, Wass et al., 2008)	Per cent correctly identified phonemes ² (max = 12) Mean response latency (ms)
Lexical access		Naming test (Hellqvist, 1995)	Per cent independently named pictures
		Sentence completion and recall ¹ (SIPS; Wass et al., 2008)	- Total number correctly completed sentences (max=18) -Semantically acceptable answers (max=18) -Semantically deviant answers (max=18) -Other
Complex working memory		Sentence completion and recall ¹ (SIPS; Wass et al., 2008)	Total number of correctly recalled words (max=18)
Visual working memory		Visual Matrix ¹ (SIPS, Wass et al., 2008)	Per cent correctly recalled/reproduced patterns (max=8)
Phoneme-grapheme correspondence		Lower-case letter names – pointing (Clay, 1975) Lower-case letter sounds – pointing (Clay, 1975) Lower-case letter sounds- naming (Frylmark, 1995)	Per cent correct responses (max = 26) Per cent correct responses (max = 26) Per cent correct responses (max = 24)
Reading ability	Phonological decoding Orthographic decoding	Test of word and nonword reading efficiency ¹ (TOWRE; Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne et al., 2009)	Number of correctly decoded words. (max=208) Number of correctly decoded nonwords in 2x45 s. (max=126) Number of totally decoded words and nonwords (max = 334)
	Passage comprehension	Woodcock reading mastery test-Revised ¹ (Woodcock 1987; Swedish version by Byrne et al. 2009)	Number of semantically correct answers. (max=68)

Notes: SIPS=Sound Information Processing System, ¹= not administered B1, ²= included in the phonological composite score, ³ different quantifications have been used in the studies, see Method section

In study IV additional segmental and suprasegmental analyses were performed. Segmental analyses were per cent phonemes (ppc) and per cent vowels (pvc) correctly repeated. Suprasegmental analyses included five measures: 1) proportion of syllable omissions in pre- versus post-stressed positions, 2) number of insertions in legal and illegal consonant clusters, 3) number of correctly repeated legal and illegal consonant clusters, 4) syllable length, and 5) primary stress correct (per cent correct out of 24 for both measures). Additionally, children's cluster repetition was categorized in four categories; number of consonant omissions (e.g., /spj/ -> [sp]), consonant substitutions (e.g., /str/ -> [spr]), vowel epenthesis (e.g., /sml/ -> [sməl]), and consonant additions (e.g., /nt/ -> [nts]).

The Naming/Phoneme test, described below was used to assess output phonology (Hellquist, 1995). Children's performance was scored both binary (by calculating children's responses) as per cent words (pwc) correctly produced and as pcc produced. Children's performance was audio recorded.

A Phonological representation task (SIPS; Wass, 2009) was employed to assess how children identified mispronunciations of real words. Thus, it taps into the child's phonological representations of words in long-term memory, complex WMC, and sensitivity to phonemic structure. First, the child was asked to name a picture. Then five different versions of the word were auditorily presented—one at a time. One version was correct and the others differed in one phoneme. The child was asked to decide whether the word was correct or not by responding “yes” or “no” after each stimulus. The score was the total number of correctly recognized items. 1 point was given for a correct identification of the right pronunciation, 0.25 p for versions that differed in one phoneme. Maximum score was 18.

A Nonword Discrimination task was used to assess discrimination of phonemes within nonwords (SIPS; Wass, 2009). In this test, the child was asked to decide whether two spoken nonwords were identical. Responses were given by pressing a key on the computer. The nonwords were presented in 16 pairs and each target nonword was presented in two conditions, once paired with an identical nonword and once with a nonword differing by a single phoneme (e.g., patinadrup–patinadrup, patinadrup–patinavrup). The child had to make correct decisions in both conditions to receive a score. The maximum score was 8.

A Phoneme Identification task was used to assess the ability to identify a phoneme within a nonword (SIPS; Wass, 2009). A phoneme was presented to the child followed by a nonword.

The task was to decide whether the phoneme was present or not in the nonword by pressing a key on the computer. The maximum score was 12.

Lexical access

The Naming/Phoneme test (Hellquist, 1995) was used to assess lexical access in Study 1. A picture with everyday objects was presented to the child. The child was asked to name orally the picture that the test leader pointed to. Children's performance was audio recorded. The score was the total number of named pictures. Semantic (e.g., "a car that drives people who are ill. . .-ambulance") or phonological (e.g., "s. . .-star") cues were given when the child was unable to name the picture. Independently named pictures were scored 1 p per item. Semantic and/or phonological cues gave 0.5 points reduction in scores per item. Maximum score was 72.

The Sentence Completion and Recall task (SCR), from the SIPS (Wass, 2009) was used in two ways: first, to assess complex working memory in Studies II and III as described below, and as a measure of lexical access in Study II. Children's spoken answers were categorized in four categories: 1) Expected, 2) Semantically acceptable (within the same category; supra-, side- or sub-ordinated), 3) Semantically deviant (not within the same semantic category), and 4) Others, (no answer, repetition).

Complex working memory

The SCR task (Wass, 2009) was used to assess complex working memory in Studies II and III, that is, the capacity to simultaneously store and process information over a short period of time. The task was to listen to series of sentences with the last word missing and to fill in and memorize the missing words, e.g., "Crocodiles are green. Tomatoes are", and thereafter to repeat the words that were previously filled in. The series included two, three and four sentences.

Visual working memory

The Visual Matrix test from the SIPS (Wass, 2009) was used to assess visual working memory in Studies II and III. A pattern of filled-in cells in a five by five matrix was displayed on the computer screen for two seconds. Thereafter, the task was to replicate the pattern of filled in cells in an empty matrix. The level of difficulty increased from 1 to 8 filled-in cells. The children received scores for the highest level of difficulty at which they correctly reproduced two out of three test patterns. Maximum score was 8.

Reading ability

Phoneme–grapheme correspondence

Two tasks were used to measure recognition of lower case letters from names or sounds (Clay, 1975). In these tasks, letter sounds and letter names, the child was presented cards with four letters in a row. The child was instructed to point to one out of four letters as the test leader read the sound or the name of the letter aloud. The maximum score was 26.

The third task, letter naming, was used to measure naming of lower case letters (Frylmark, 1995). The child was presented with a chart of letters in six rows. The task was to name each letter as the test leader pointed. The maximum score was 24. Children’s performance was audio recorded. The letter knowledge tasks were used in all four studies.

The Test of Word Reading Efficiency (TOWRE; Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne et al., 2009) was used to assess decoding of real words and decoding of nonwords. Both measures were used in Study III. Nonword decoding was used in Study IV. The child was required to read aloud as many words/nonwords as possible in 45s. He/she was also asked to read as correctly as possible. This procedure was repeated twice with two separate lists of words/nonwords. Children’s performance was audio recorded.

Word and nonword decoding was scored in two ways in Study III;

- 1) Reading accuracy. The children received credits for every word/nonword read correctly (maximum score 208/126). The total decoding score for both words and nonwords was 334.
- 2) Decoding errors. The children’s decoding errors were calculated as per cent incorrectly decoded words/nonwords of the total sum of read words/nonwords (correct and incorrect).

The nonword-decoding task in TOWRE was further analysed in Study IV according to the following:

1. Binary – Per cent nonwords correctly decoded (pnwc). Vowel quality was scored according to the following for nonwords with CV - or CVC - structure: ex. “ba” received a score of 1 when pronounced [ba] or [ba], “bat” received a score of 1 when pronounced [bat] or [bat]. For nonwords where the vowel was followed by a double consonant, for example, “kratti”, only [krati] not [krati] received a score of 1 following the orthographic rules in Swedish.
2. Per cent phonemes correctly decoded (ppc).

An additional two measures were included, in order to capture children's decoding strategies more accurately, that is, per cent nonword trials (pnwt) and proportion of nonword decoding errors (pnwe) in relation to trials (see nr 3 and 5 below). In pnwt correctly and incorrectly read nonwords were included. The rationale for this was that the analysis gives an estimate of how much of the nonword-decoding task the child completed in 45 seconds and, therefore, made it possible to better differentiate between children reaching similar ppc scores. For example, child nr 2 (CI) who reached a ppc of 13, made attempts to read out 42 nonwords in 45 sec. Out of these 27 were incorrect (64%). The age-matched NH peer also reached a pcc of 13 but only attempted to read out 37 nonwords and out of these only 18 were incorrect (49%). Further, we analyzed the influence of the phonological complexity of orthographically transparent nonwords on children's nonword decoding ability, that is, nonwords with no clusters and nonwords with clusters.

3. Per cent nonword decoding trials (correct and incorrect: pnwt). The rationale for this analysis was to capture children's decoding strategies more accurately. Both correctly and incorrectly read nonwords were included. The analysis gives an estimate of how much of the nonword-decoding task the child completed in 45 seconds and made it possible to better differentiate between children reaching similar ppc scores. For example, in Study IV child nr 2 (CI) who reached a ppc of 13, made attempts to read out 42 nonwords in 45 sec. Out of these 27 were incorrect (64%). The age matched NH peer also reached a ppc of 13 but only attempted to read out 37 nonwords and out of these only 18 were incorrect (49%).

4. Per cent orthographically transparent nonwords correctly decoded, nonwords with no clusters, and nonwords with clusters. This analysis was performed in order to analyse the influence of phonological complexity of orthographically transparent nonwords on children's nonword decoding ability.

5. Per cent nonword decoding errors out of nonword decoding trials; (pnwe).

6. Additionally, nonword decoding errors were categorized in four categories; phoneme insertions, phoneme deletions, phoneme substitutions, and lexicalization, that is, making a real word of the target nonword

The Woodcock Passage Comprehension Test (Woodcock 1987; Swedish version by Byrne et al., 2009) was used to assess reading comprehension in Study III. This test uses a cloze procedure to assess the child's ability to understand passages of connected text. The children's

semantically accepted answers were scored. The maximum score was 68.

A Reading Composite Score was calculated as the summary in per cent of the total decoding-score of TOWRE and the passage comprehension-score of Woodcock. This was completed pre and post intervention in Study III.

Reading change was calculated as the difference in three respective scores (word decoding, nonword decoding, and passage comprehension) between post and pre intervention in Study III.

Statistics

Descriptive statistics were first conducted in all studies.

In Study I reported data of PhPS and letter knowledge are reported from all test points, baseline 1, baseline 2, and post intervention (B1, B2 and P1). Parametric tests were used to explore within- and between-group differences. Separate analyses for gender were carried out, but no gender differences were found. One-way ANOVA was conducted at B1 and at B2 to reveal between-group differences. Between-subject factor at B1 and B2 was children's technical aid (3 groups; 1.CI, 2.HA and, 3.NH). Tukey's honestly significant test was used for multiple comparisons. The second between-subject factor was the time period for DHH children's phonological change (2 groups; 1.B1 to B2 (N = 14), 2.B2 to PI (N = 18)), thus the group of DHH children (N = 32) was divided according to the time period when their phonological composite score showed a positive change. A mixed design ANOVA was used to analyze within and between subject effects related to time period B1 to B2, B2 to PI, and B1 to PI for the phonological composite score, that is, to compare effects in participants' PhPS throughout the study, as well as between group effects (2 groups; 1.B1 to B2 (N = 14), 2.B2 to PI (N = 18)). Pearson's correlation was calculated to examine the effect of children's initial phonological composite score, on the phonological composite change-score between B1 and B2, as well as between B2 and PI. Pearson's correlation was calculated between DHH children's background variables and variables related to the intervention. A repeated measure ANOVA was used to analyze the constituent measures of the phonological composite scores in the two groups of DHH children separately (B1 to B2 vs. B2 to PI).

In Study II reported data are from B2 and PI. All cognitive measures but reading were included. Two different group comparisons of children's performances were made by

independent samples t-tests for normally distributed data and the Mann-Whitney U-test for non-parametric data pre intervention. First, with children's hearing as between-subject factor (1.NH, 2.D/HH), and, secondly, with the initial level of the phonological composite score for DHH as a function of a median split (1.phonologically skilled DHH, 2.phonologically less skilled D/HH). Pearson's correlation coefficient (in case of skewed data Kendall's Tau-b) was calculated between the phonological composite score pre intervention and cognitive tasks (lexical access, complex and visual WM, phonological latency scores, and letter knowledge). This was conducted in three groups; 1.NH, 2.D/HH, and 3.phonologically less skilled DHH children. Following this, a correlation analysis between all cognitive tasks, and the phonological change score was performed. This was done in three groups; 1) NH (N = 16), 2) DHH (N = 32), and 3) phonologically less skilled D/HH children (n = 16). To examine the total contribution of the factors, the significant correlates were put into a multiple linear regression analysis (backward method).

In Study III reported data are from B2 and PI. A Mann-Whitney U test was used for group and age comparisons at both test points. A paired samples t-test was conducted to analyse improvement in reading accuracy (words, nonwords and passage comprehension) from B2 to PI in NH children. Wilcoxon Signed Rank Test was chosen as the nonparametric alternative for DHH children. Mixed design ANOVA was used to analyse differences in decoding errors (per cent words and nonwords decoded incorrectly) with time as within-group factor (from pre to post intervention) and children's hearing status as between-group factor (2 groups; NH and DHH). Only children who were judged as readers at pre intervention (NH: n = 12, DHH: n = 19) were included that is children who read at least one word/nonword correct on TOWRE at pre intervention were included. Children with knowledge in phoneme-grapheme correspondence but who did not blend sounds into words were excluded. Following this, correlations (Kendall's Tau-b; NH: N =16; DHH: N = 32) were calculated between reading change-scores (see Method section) and all measures obtained pre intervention as well as between reading change-scores and demographic variables.

In Study IV reported data are from PI. Considering the small sample size all data was analysed using non-parametric statistics. Group comparisons were made using the Mann Whitney U-test. Correlational analyses were conducted using Spearman's correlation coefficient. P-values < .05 were considered significant.

Intervention program and setting

The computer-assisted intervention was accomplished by means of an originally Finnish-Swedish version of Graphogame (www.graphogame.com) which was translated into standard Swedish. Recordings were made at the Humanities Laboratory at Lund University with a female voice speaking standard Swedish. The program focuses on the correspondence between phonemes and graphemes and delivers training in different backgrounds and formats. It begins by presenting falling balls with letters or letter sequences on the screen. The task for the child is to click on the right ball among others that matches the target sound, before it reaches the bottom of the screen. Graphogame follows the bottom-up or phonics approach (McArthur, Eve, Jones et al., 2012; Trezek & Malmgren, 2005) by first introducing the spoken phonemes with their corresponding graphemes, then mono-syllabic words (CV, VC) and, finally, more complex words (CCV, VCV, VCC, CVCV). The program enables individual intervention since it adapts itself to each child's level of performance. An algorithm in the program presents approximately 20% of the items from the pool of new connections between phonemes and graphemes, yet to be learned, in a way to benefit the player's learning. Progression through the game is controlled so that around 80% will be correct. The program demonstrates how to blend isolated sounds into syllables and words and, thus, offers basic exercises for spelling (Lyytinen et al., 2007b). Graphogame has a child friendly design to keep children's motivation high, for example they may choose their own favourite game character, a princess, an animal, or a knight, and after each level of difficulty they are rewarded with tokens presented in another background, for example a castle or a garden. One special game is the ghost-and-ladder game where children make the ghost climb up the ladder when giving correct responses (Kyle, et al., 2013).

The Swedish version of Graphogame includes 56 levels, categorized in three themes according to the phonological and orthographical complexity of the words. It starts with isolated capital letters and their corresponding sounds, then introduces the lower case letters, advances to one-syllable words with CV (consonant vowel) structure (theme 1), proceeds to VC, CVC, VCC and CVCC structures (theme 2), and finally delivers training for up to seven letter words (theme 3). The words at theme 3 contain initial consonant clusters as well as words with the first examples of larger grapho-phonemic units, namely the bigraphs: "ng" /ŋ/, "sj" /ʃ/ and tj /tʃ/.

All participating children were asked to practice ten minutes per day for four weeks with the game. They were told to practice in a way that corresponded as closely as possible to their way of normally using a computer. If the DHH children listened through external loudspeakers or through a hearing loop in the normal case, they were instructed to continue to do so when they were practicing. Nonetheless, all children listened through external loudspeakers when practicing. In case the DHH child experienced difficulties to discriminate between voiceless plosives (that is, p-t), the parents were advised to show the difference between the sounds by explicitly articulating that is, showing their mouth movements to the child. Thus, if a DHH child had phonemic knowledge but experienced difficulties to perceive the difference between phonemes should not prevent them from continuing to the next level (Adams Jager, 2003).

The treatment integrity of the training program was accomplished by means of personal and written information, web-sms, e-mail correspondence and phone calls from the first author (Gresham, MacMillan, Beebe-Frankenberger et al., 2000). The majority of the testings at baseline 1 was made in the children's homes. This enabled the first author to give parents personal advice regarding an optimal intervention setting, for example, that the child could do the training in a silent room where it was possible to shut the door. In those cases where the families did not follow the training schedule, they were informed to compensate missing days by increasing the daily practice with additional training. Dates and time of day when training took place, total amount of training time (h: min), reached level in the game (max. 56), and per cent tasks correct were registered automatically for each child (Lyytinen et al., 2007). Mean time of daily practice was 7 minutes and total mean time of practice was 202 minutes (SD = 52, 85 - 334). Group comparisons (NH, DHH) of the mean score of total amount of playing, reached levels in the game, or per cent mean correct during practice showed no significant difference.

Ethical considerations

Written parental informed consent was obtained for all the participants. All participants were informed in spoken and in written form that participation in the study was voluntary and that they could withdraw at any time without stating the cause. In cases of fatigue during testing some tests were called off for some children. This was mostly evident in children who had not begun to read during the study. The Regional Committee for Medical Research Ethics in Stockholm, Sweden dnr_2009/905-31/2) approved the study.

SUMMARY OF THE STUDIES

Study I.

Computer-assisted training of phoneme-grapheme correspondence for children with hearing impairment: Effects on phonological processing skills.

Aims

The aims of study 1 were to examine DHH children's phonological processing skills (PhPS) in relation to children with NH at B1 and B2 with a computer-assisted intervention program that focused on phoneme-grapheme correspondence training. Further, overall and specific effects of computer-assisted phoneme-grapheme correspondence training on PhPS were investigated.

Method

A total of 48 children, 5, 6 and 7 years of age participated in this study. There were 32 deaf or hard of hearing (DHH) children using cochlear implants (CI) or hearing aids (HA), or both in combination, and 16 children with NH. The study had a quasi-experimental design with three test occasions separated in time by four weeks; baseline 1 (B1) and 2 (B2) pre intervention, and the third occasion, post intervention (PI). Group comparisons at both test points pre intervention were made between children with CI ($n=17$), children with HA ($n=15$) and children with NH ($n=16$). Further, children's initial phonological composite score (B1) served as a variable to compare phonological change at B2 and at PI, that is, a correlation analysis between phonological change and children's initial score (B1) was performed. Children performed tasks measuring different aspects of phonological processing, lexical access, and letter knowledge. All children practiced ten minutes per day at home with the computer-assisted program supported by their parents.

Results and discussion

Study 1 showed that children with NH outperformed DHH children on the majority of phonological processing tasks, except for phoneme identification.

At B1 the NH children outperformed both groups of DHH children (CI and HA) on the following measures; the phonological composite score $F(2, 45) = 22.59, p < .001$, the Nonword repetition test (pnwc) $F(2, 45) = 82.27, p < .001$, (pcc) $F(2, 45) = 33.78, p < .001$, and the task on phonological representation, $F(2, 45) = 6.26, p < .05$. For the Phoneme test,

(pwc and pcc) the significant difference was between NH children and children with CI, (pwc) $F(2, 45) = 3.63, p < .05$, (pcc) $F(2, 45) = 3.14, p = .05$. Additionally, children with CI performed at a significantly lower level than the other groups on the Nonword discrimination task $F(2, 45) = 13.74, p < .01$. These results show that children with CI had difficulties with phonological representations and nonword discrimination, as well as with output phonology.

At B2 all significant group differences from B1 remained and one was added; lexical access ($p = .06$ at B1, $p = .02$ at B2), $F(2, 45) = 4.11, p < .05$. The significant difference was between children with NH and children with CI. Thus, although the task of lexical access was relatively easy, as was indicated by scores close to the ceiling, it still differentiated children with CI as having relatively weaker naming skills. There was no significant difference between the groups on letter knowledge. It should be noted, however, that children with HA showed an overall lower performance level when comparing means, than the other two groups at all points in time.

The intervention showed most positive effects on phoneme–grapheme correspondence as measured by the letter knowledge tasks, with moderate to strong effect-sizes from B1 to PI. One of these, knowledge of letter sounds, showed a significant improvement from B2 to PI in all children (CI, HA, NH), $F(2, 67) = 19.4, p < .01, \eta^2 p = .30$. So did the Phoneme test pwc $F(2,60) = 3.86, p < .05, \eta^2 p = .11$, and pcc $F(2, 60) = 6.16, p < .01, \eta^2 p = .17$. The phonological composite score, however, showed a significant improvement for all children, evident at B2 as well as at PI, indicating a continuous development throughout the study.

To analyze the effect of children's initial level of PhPS (B1) a two-step correlation analysis was conducted in all children; first, between the phonological change-score from B2 to PI and the initial phonological composite score, and second, between the phonological change-score from B1-B2 and the initial phonological composite score. Only the first two-step correlation analysis was significant, ($r = -.42, p < .01$). When analyzing the separate groups (CI and HA), the same negative correlation was only obtained in children with CI ($r = -.63, p < .01$). Thus, children who obtained an initial low level on the phonological composite score, and specifically children with CI, showed specific benefit of the intervention. When analysing the constituent parts of the phonological composite score strongest effect sizes were observed in the nonword repetition task used to assess phonological working memory, evident at B2 as well as at PI.

In sum, the intervention proved effective regarding phoneme-grapheme correspondence accuracy and output phonology in picture naming, for all children. For some DHH children phonological processing skills were boosted relatively more by phoneme-grapheme correspondence training. This reflects the reciprocal relationship between phonological change and exposure to and manipulations of letters.

Study II.

Predictors of phonological change in deaf and hard of hearing children who use cochlear implants or hearing aids

Aims

The aim of Study II was to examine cognitive abilities, specifically working memory (WM), lexical access and letter knowledge, in relation to PhPS at pre intervention, and to phonological change at post intervention.

Methods

Details of the participants were identical to those reported in Study 1. Analyzed assessment points were B2 and PI. Group divisions were made according to 1) children's hearing status, that is, DHH children (N=32) and NH children (N=16) and 2) DHH children's level of PhPS; phonologically skilled (n=16), and phonologically less skilled (n=16). The phonological composite score and the phonological change score served as dependent variables. Cognitive variables, that is, complex WM, lexical access, PhPS and letter knowledge, were compared between DHH and NH children at B2. Further, associations between cognitive variables and PhPS were examined at B2 as well as between complex WM, lexical access and PhWM (NWR, pnwc) in each group respectively. Finally, cognitive variables were entered as predictors of phonological change at PI. Lexical access was assessed by means of children's verbal responses (sentence completions) in the SCR-task.

Results and discussion

Significantly higher performance was observed in NH children compared to DHH children on half of the cognitive measures. These were three out of four categories of the lexical access task (expected answers, semantically acceptable answers and other answers), three out of five

tasks of PhPS (the phonological composite score, phonological representations, and nonword repetition), and letter knowledge for sounds. Comparable levels were observed on lexical access-deviant answers, the two phonological latency scores, complex and visual working memory and two tasks of letter knowledge. For group comparison regarding working memory performance at B2 and PI, see Figure 4.

When group comparisons were made in relation to DHH children's initial PhPS results showed that phonologically skilled DHH children outperformed phonologically less skilled DHH children on all cognitive variables except four; these were, three aspects of lexical access (semantically acceptable, semantically deviant and others) and phoneme identification latency. Age (80 vs. 72 months) did not differ significantly between the groups. These results demonstrate the high degree of heterogeneity within DHH children's PhPS, that is phonologically skilled and less skilled DHH children show highly disparate performance. Additionally, phonologically less skilled DHH children displayed a more limited WMC.

Less efficient lexical access is most probably due to, a) reduced auditory stimulation during critical developmental periods, that is duration of deafness/unaided hearing, and b) a result of that the DHH child having developed his/her language with a distorted auditory signal. This probably leaves the DHH child with a poorer lexical network (fewer lexical representations and weaker links between them) in long-term memory.

A significant correlation between complex WM and the phonological composite score at pre intervention was only observed in DHH children, but not when we analyzed phonologically less skilled children separately. One interpretation is that when dealing with phonological processing tasks, complex WM may contribute differently in the group of DHH children. Some of them may be able to use their executive abilities to shift attention between different aspects of the incoming speech signal as they were in the SCR-task. It is therefore important to further examine WMC in children with HA and CI since it is a robust predictor of many different aspects of language functioning. In children with NH age was the variable with strongest correlations with the phonological composite score.

When we examined factors that predicted phonological change there were different patterns within the groups. In children with NH only two variables were associated with phonological change: weak lexical access (that is, negative correlation) and letter knowledge for sounds (that is, positive correlation). The latter finding may be interpreted as support for

neurocognitive studies, which have shown that orthographic knowledge shapes the phonological representations involved in spoken language processing.

Weak initial performance on a task for phonological representations, which captures both lower level and higher level auditory processing, was the only significant predictor of phonological change in DHH children. Letter naming (positive correlation) was associated with phonological change in DHH children with weak initial PhPS. DHH children with a later identified HL and who were later implanted, showed greater phonological change.

In sum, the Phonological representation task served as a sensitive and broad measure to identify DHH children in need of intervention. For children with weak initial PhPS letter-naming skills acted as driving force for phonological change.

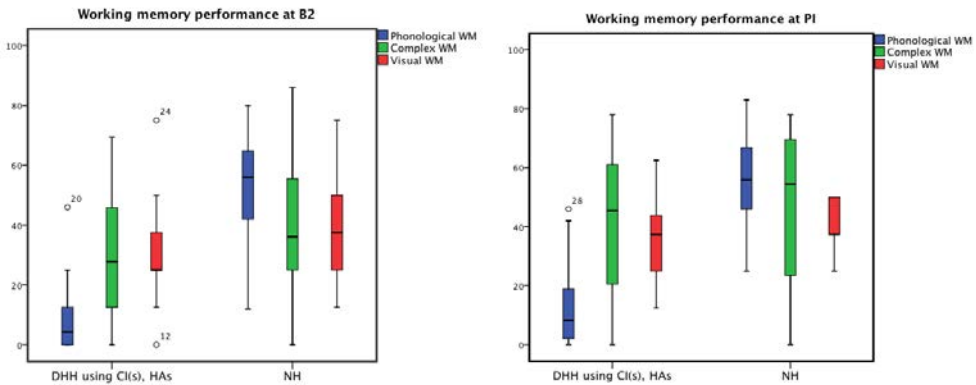


Figure 4. Phonological WM (the Nonword repetition test, per cent nonwords correct), complex WM (the Sentence completion and recall task) and visual WM (the Visual Matrix task) in deaf and hard of hearing children using CI or HA, and NH children at B2 and at PI intervention (PI)

Study III.

Computer-assisted reading intervention with a phonics approach for children using cochlear implants or hearing aids

Aims

The first purpose of study III was to compare NH and DHH children's reading ability at pre and post intervention. The second purpose was to investigate effects of the intervention. Third, cognitive and demographic factors were analyzed in relation to reading improvement.

Methods

Details of the participants were identical to those reported in Study 1 and II. Group divisions were made according to 1) children's hearing status, that is, DHH children (N=32) and NH children (N=16), and 2) children's age; 5, 6 and 7 years. Dependent variables were decoding accuracy and decoding errors of words and nonwords (TOWRE; Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne et al., 2009) and passage comprehension (Woodcock Reading Mastery Test, Revised, 1987; Swedish version by Byrne et al., 2009). Cognitive and demographic variables were used to analyse associations with reading improvement. These were the phonological composite score, lexical access as measured by the SCR-task, letter knowledge, complex and visual working memory, nonverbal intelligence, age at CI/HA, gender, and age at B2.

Results and discussion

There was no statistically significant difference for reading ability at the group level in relation to children's hearing status, although NH children showed overall higher reading scores at both test points (B2, PI). However, age comparisons revealed a statistically significant higher reading ability in the NH 7-year olds compared to the DHH 7-year olds. Post intervention significantly higher scores were evident in word decoding accuracy and passage comprehension. There was also a significant reduction in nonword decoding errors in both NH and DHH children. These results support the notion that offering a computer-assisted intervention program delivered at home is an alternative way to support not only NH children with reading difficulties but also DHH children to develop phonological decoding proficiency.

Reading improvement was associated with complex working memory and phonological

processing skills in NH children. This suggests a combined influence of domain general variables and phonological processing skills in reading development for the NH children. Correspondent associations were observed with visual working memory and letter knowledge in the DHH children. These results suggest that DHH children's beginning reading may be influenced by visual strategies that might explain the reading delay in the older children. Thus, in seven-year old children graphemes must be fused with phonological information to enable independent blending of graphemes to syllables and words, as in efficient phonological decoding.

Study IV.

Segmental and suprasegmental properties in nonword repetition – An explorative study of the associations with nonword decoding in children with normal hearing and children with bilateral cochlear implants

Aims

The first aim of study IV was to scrutinize segmental and suprasegmental aspects of nonword repetition in two groups of children individually matched for age, including children with NH and children with CI. The second aim was to analyze the associations between nonword repetition and nonword decoding in both groups.

Method

Participants were eleven children with bilateral CI (nine girls), 5:0-7:11 years ($M = 6.5$ yrs.), and eleven normal-hearing (NH) children (five girls), individually age-matched to the children with CI. Children's performance at PI was analyzed. Measures were the Nonword repetition test (SIPS: Wass, 2009) and nonword decoding from TOWRE (Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne et al., 2009). The Nonword repetition test was scored and analyzed in three ways; 1. Binary - per cent nonwords correct; pnwc, 2. Segmentally - per cent consonants correct; pcc, per cent vowels correct, pvc, and per cent phonemes correct, ppc, and 3. Suprasegmentally - proportion of syllable omissions in pre versus post-stressed positions, number of insertions in legal and illegal clusters, per cent correctly repeated legal and illegal clusters, syllable length and correct primary stress. Additionally, children's cluster repetition was categorized in four categories; number of

consonant omissions (e.g., /spj/ -> [sp]), consonant substitutions (e.g., /str/ -> [spr]), vowel epenthesis (e.g., /sml/ -> [sməl]), and consonant additions (e.g., /nt/ -> [nts]).

Nonword decoding was analyzed and scored according to the following: 1. Binary - per cent nonwords correctly decoded (pnwc), 2. Per cent phonemes correctly decoded (ppc), 3. Per cent nonword-decoding trials (correct and incorrect: pnwt). 4. Per cent orthographically transparent nonwords correctly decoded; nonwords with no clusters, and nonwords with clusters, 5. Per cent nonword-decoding errors out of nonword-decoding trials (pnwe). 6. Additionally, nonword-decoding errors were categorized in four categories, thus analyzed qualitatively; phoneme insertions, phoneme deletions, phoneme substitutions, and lexicalization, that is making a real word of the target nonword. A composite score was computed based on performance in three letter tasks; recognition (matching phonemes and letter names to graphemes) and naming of lower case letters).

Results and discussion

Normally hearing children outperformed children with CI on all aspects of the nonword repetition task. The largest difference between the groups was found for totally correctly repeated nonwords, binary scoring. Here, NH children repeated 54% of the nonwords correctly compared to the children with CI who only reproduced 5% correctly. These results imply that repeating completely novel words after a single auditory-only presentation, without making any alterations of the phonological structure, is extremely challenging for children with CI. Suprasegmental analysis of syllable length in the nonwords revealed that children with CI made far more syllable omissions than did the NH children, and predominantly in pre-stressed positions. Suprasegmental analyses of stress patterns must be anchored in the characteristics of the ambient language. Word stress in English, as well as in Swedish, have a trochaic bias (strong-weak pattern) and, in early language acquisition, weak syllables are often omitted in pre-stressed positions. This finding suggests a trochaic bias, not only in language acquisition, but also in nonword repetition in Swedish children with CI, in line with studies on English-speaking children with CI. The last prosodic aspect of nonword repetition that was investigated revealed that children with CIs made significantly more consonant omissions and consonant substitutions when repeating legal clusters in the nonwords as compared to the NH children. Here, the children with NH showed ceiling effects, i.e., hardly any omissions or substitutions occurred. Thus, for NH children 6.5 years of age with typical language development, repeating legal consonant clusters with two or three consonants in

nonwords seems not particularly demanding. Among children with CIs however, reproducing legal consonant clusters in nonwords was challenging for almost all of them, even though exceptions were found. For example, children nr 3 and 6 repeated 75% vs. 100% of the legal clusters correctly.

No significant difference was found for nonword decoding accuracy between the groups. Additionally, no significant difference was found in the decoding of transparent nonwords with or without clusters. An error analysis of the children's nonword decoding showed a statistically significant higher percentage of nonword decoding errors in children with CI. The additional qualitative analyses showed that children with CI made significantly more phoneme deletions in decoding than NH children.

Children with NH showed positive and significant associations between nonword decoding (pnwc and ppc) and the majority of aspects of nonword repetition. In children with CI nonword decoding (pnwc) was significantly and negatively correlated *only* with one aspect of nonword repetition, namely, with consonant omissions in nonwords with legal clusters. Thus, being able to repeat fine-grained aspects of the speech signal, i.e. consonant clusters that follow the phonotactic rules of the ambient language were associated with a higher nonword decoding proficiency. Using fine-grained units have for long been acknowledged as particularly important in the reading of unfamiliar items, as nonwords (Stanovich, 2000).

Age was significantly correlated to nonword decoding in children with NH but no demographic variable came out significant in children with CIs. However, letter knowledge did. This acknowledges the importance of phonological processing skills and letter knowledge in decoding, not only in children with NH (Hulme et al., 2012) but also in DHH children using CIs.

GENERAL DISCUSSION

Summary of the findings

In this thesis, phonological processing skills, lexical access, working memory and reading ability is examined in DHH children 5, 6, and 7 years of age using CI(s) or HA compared to an age-matched reference group of NH children. All children took part in a computer-assisted intervention program with a phonics approach with 10 min of daily practice during four weeks. The training was accomplished by means of the Internet and support was given by a SLP via web-sms, telephone calls and e-mails to achieve treatment integrity. Overall findings will now be discussed followed by a section on clinical implications. Finally, some suggestions for future research will be mentioned.

The results from Studies I and II replicated previous findings of weak PhPS and lexical access in DHH children. Levels comparable with NH children were observed on complex and visual WM. These results are expected and similar findings have been reported in previous research within this field. For research regarding phonological processing skills with findings in line with the present thesis, the reader may consult Briscoe et al. (2001), Geers et al. (2003), Geers, Tobey, Moog et al., (2008), Ibertsson, Willstedt-Svensson, Radeborg et al. (2008), Lee, Yim and Sim (2012), and Wass (2009). For findings regarding lexical access see Lee, Yim and Sim (2012), Löfkvist (2014), and Schwartz et al. (2013). The slightly different results regarding lexical access as compared to the thesis of Löfkvist (2014) will be discussed in the section on lexical access below.

Furthermore, all children improved their accuracy in phoneme–grapheme correspondence and output phonology as a function of the computer-assisted intervention. For the whole group of children, and specifically for children with CI, a lower initial phonological composite score was associated with a larger phonological change between baseline 2 and post intervention. For DHH children with weak initial PhPS, letter knowledge served as a mediating factor for phonological change. Deaf and hard of hearing children with a later identified HL and who were later implanted, showed greater phonological change.

In Study III comparable levels in reading ability were found for both groups (DHH and NH) in the 5 and 6-year old children, but a higher proficiency in the NH 7-year olds. The results at post intervention indicate that the intervention was effective for word decoding, for nonword-decoding proficiency, and passage comprehension in both NH and DHH. The results may

imply that DHH children's reading improvement was influenced by visual strategies whereas in the NH children by PhPS and complex WM. In a recent study by Park and Lombardino (2013) children with a mild to moderate SNHL with weak PhPS displayed a similar reading pattern as the DHH children in the present study. Thus, according to the authors, these children may have compensated weak PhPS by relying on visually or partially developed orthographic skills in word reading.

Study IV confirmed the findings from earlier research on nonword repetition (NWR) in children with CI. Thus, repeating completely novel words after a single auditory-only presentation without making any alterations of the phonological structure is extremely challenging for these children. For research on nonword repetition in children with CI with findings in line with the present thesis the reader may consult Carter, Dillon and Pisoni (2002), Dillon et al. (2004), and Ibertsson et al. (2008). Several new insights were also gained. First, results suggest a trochaic bias, not only in language learning in typically developing children (Carter & Gerken, 2003), but also in NWR in Swedish children with CI, in line with the findings in English-speaking children with CI. Second, different error patterns in nonword decoding were observed in children with NH and children with CI. Phoneme deletions occurred almost exclusively in children with CI. Further, a strategy of lexicalizing nonwords was more frequently observed in the children with CI compared to the NH children.

What is new?

In the present thesis for the first time, results from a computer-assisted reading intervention with a phonics approach delivered by means of the Internet in DHH children's homes, are presented. With this new method, the children did not need to travel to the clinic for individual face-to face intervention, but could practice at home, with support by their parents and a SLP. Additionally, for the first time a phonics-based reading intervention, which for long has been proved effective for children with dyslexia, has been accomplished among Swedish DHH children. This is an important step, possible to realise due to technical advancement during the last fifty years and increasing knowledge regarding the importance to recognize DHH children's residual hearing and the need of phonological intervention. One important finding is the demonstration that letter knowledge may serve as a mediating factor for phonological change in children with weak PhPS. This may have implications for other clinical groups, for example, children with language impairment. Indications of different

strategies and error patterns in phonological processing and reading also present novel and fertile contributions, to further research and clinical work, as well as technological development.

Phonological processing skills

Generally DHH children had weaker PhPS than NH children at all test points. However, there was a large variation among the DHH children. Approximately 20% (six children) performed within -1 SD of NH children. These were children using HA (one child had bimodal hearing), whereof five children had a progressive hearing loss. Their mean age of diagnosis was approximately four years. Five children who used CI (three children had bimodal hearing) had the weakest phonological processing skills. Out of these children, one was congenitally deaf. All five children were diagnosed before two years of age but received their implants relatively late (mean age at implant was 34 months, range 15-55 months). Three of these children used sign language as the main communication mode at home or used sign to support spoken language. Additionally, one of these children had another language background and was not exposed to Swedish until one year of age. Rotteveel et al. (2008) investigated the influence of onset and duration of deafness, and mode of communication on speech perception skills in congenitally, prelingually and post-lingually deaf children using CI in the Netherlands. The post-lingually deaf children had the best speech perception skills. Further, duration of deafness played a major role in speech perception ability in congenitally deaf children, but communication mode was a major factor in prelingually and post-lingually acquired deafness together. Thus, children who were placed in a spoken language educational setting acquired auditory perception skills at a faster rate than children in total communication school-settings. It could be argued that children who had weaker speech perception skills should probably be placed in educational setting with sign language more often. But, as Rotteveel et al. reasoned, the educational system for DHH children in the Netherlands formerly had fewer options for oral communication programmes. Together, the results from the present thesis seem to support the findings from a large number of studies that auditory plasticity is highest during sensitive periods early in life, and that auditory stimulation alters and shapes the auditory and phonological network positively (Kral, 2013; Rotteveel, Snik, Vermeulen et al., 2008; Sharma, Dorman, Spahr et al., 2002; Sharma, Dorman et al., 2002b; Sharma, Dorman & Spahr, 2002a).

Lexical access

The DHH had weaker lexical access skills compared to the NH children, and particularly children with CI. This was evident in the picture-naming task (Hellquist, 1995), as well as in the sentence completion task (SCR, Wass, 2009). However, DHH children's performance was relatively better and more homogeneous in the picture-naming task. Schwartz et al. (2013) stress that for children with CI lexical access in speech recognition is more challenging than in speech production. In the SCR-task pre-recorded sentences that children need to process auditorially within a certain time limit are used. Thus, the SCR-task challenges children's speech recognition skills.

Picture naming is visually prompted. A picture prompts children's word retrieval and children need to access semantic knowledge, which is believed to show typical development in children with CI. Thus, here the picture prompts children's word retrieval. In the SCR-task on the other hand, lexical access is not visually but semantically, grammatically and phonologically prompted.

In the SCR-task DHH children produced significantly less expected answers, less semantically acceptable answers, and more 'other' answers than the NH children did. Even some children, particularly children with CI with a longer duration of deafness, solely produced 'other' answers, that is, did not give any answer at all or repeated the final word of the first sentence. Recently, Löfkvist (2014) found comparable performance in picture naming in children with CI and age matched NH children, aged 6-9 years. Similar results have also been reported for children with CI in the study by Wechsler-Kashi, Schwartz & Cleary (2014). Löfkvist (2014) found that children with CI more often gave semantically acceptable responses than omitted or semantically irrelevant answers. Especially, the group of younger implanted children had good lexical-semantic outcome (Löfkvist, 2014). The reason for the apparently dissimilar results obtained in the present thesis for lexical access compared to those of Löfkvist (2014) and Wechsler-Kashi et al (2014) is largely explained by the differences in task demand. Thus, in their studies children's speech recognition skills were not challenged since they use picture tasks tapping word association and lexical organisation.

Working memory capacity

Comparable performance was observed in complex and visual WM in DHH children and children with NH in the present study. As is shown in Figure 4 (p. 71) there was a large variation in complex WM performance in both groups. Thus, results on a group level must be

interpreted with caution. However, the findings show that although the SCR-task challenges speech recognition (that is, it is domain sensitive) the majority of the DHH children managed to use their executive functions to shift attention between sentence content (semantic, grammatical and phonological processing), and retrieval (storage). Bayliss et al. (2003) analysed the residuals in complex WM tasks in adults and children with NH and suggested that beyond the processing and storage operations, coordination between them, that is, processing speed is essential. Further, Bayliss et al. showed that across tasks (verbal and visuospatial), the domain specific storage capacity was an important determinant of complex span performance. These researchers concluded that in all, three factors explained the variance in performance: a general processing factor, a verbal storage factor and a visuospatial storage factor. In the case of the SCR-task the verbal storage factor can be defined as the children's lexical access skills, that is their mental lexicon in LTM. Indeed, additional correlation analysis of the associations between complex WM, lexical access-expected answers and NWR (pnwc) showed a reversed pattern in DHH children and NH children. For DHH children only the correlation between complex WM and lexical access was significant ($r = .52, p < .01$). For children with NH on the other hand, only the correlation between complex WM and phonological WM was significant ($r = .55, p < .05$). Thus, the interpretation of this is that DHH children may have relied relatively more on storage operations whereas NH children may have relied relatively more on phonological processing when performing the SCR-task. The present findings are promising since complex WMC is essential in children's listening comprehension (Just & Carpenter, 1992; McInnes, Humphries, Hogg-Johnson et al., 2003; Moser, Fridriksson & Healy, 2007; Pimperton & Nation, 2014) as well as in reading (McVay & Kane, 2012) and in maths (Cowan & Powell, 2014). The findings also show the importance of undertaking vocabulary work in DHH children since larger vocabularies are associated with better WMC (Stiles et al., 2012). Nonetheless, it is important to note that for DHH with weak PhPS overall WM difficulties were evident. Although these children's group mean age was 8 months lower no significant age difference was observed compared to skilled children, so these difficulties may not solely be explained by age. Accordingly, phonologically less skilled children will most likely need additional support in several learning situations where use of complex linguistic skills, such as literacy, is required.

Literacy

An observed trend in the data was that children with HA showed an overall lower performance on tasks of letter knowledge at all test points. Additional analysis of decoding ability and reading comprehension revealed a similar pattern. As group sizes are small these results should be interpreted with due caution. Nonetheless, they may reveal slightly different parental/educational approaches towards children using CI as compared to children using HA (Asker-Árnason, 2011). Higher performance in children with CI could reflect an increased awareness among parents and educators of the importance to support language and literacy in this group. One way to bridge this apparent difference may be parent education. Indeed this has been fruitful in recent projects, for example, in Germany where parents of children with HL received communicative responsiveness training and were taught how to use dialogic book reading to support language and literacy (Reichmuth, Embacher, Matulat et al., 2013).

There was no significant difference between DHH and NH children's reading ability as measured by decoding and passage comprehension on a group level, but significantly lower reading scores were evident in DHH 7-year olds when age-group comparisons were made. There was also a tendency towards more nonword decoding errors in the DHH children. The subsequent correlation analyses showed significant associations between decoding improvement and visual WM as well as with letter knowledge in the DHH children. For children with NH correspondent significant associations were found with PhPS and complex WM. Thus, the DHH children apparently relied more on visual strategies. In beginning reading PhPS and letter knowledge are crucial factors, combined with vocabulary and syntactic competence that is children need several skills to become proficient readers (Bryant, 1998; Lundberg, 2009; Lyytinen et al., 2006). One could argue then that although the DHH children had undergone the intervention program with a phonics approach, they did not completely use the strategies taught in the program. A recent follow up study of the participating children two years after taking part in the intervention, seem to contradict that notion (Pfándtner & Wallfeldt, 2013). Preliminary results showed comparable group means for both decoding and reading comprehension in the DHH and NH children. A subsequent correlation analysis between reading and different aspects of WM revealed significant associations between all aspects of reading and complex WM, as well as between all aspects of reading and nonword repetition in children with CI. A similar pattern was found in children with NH but not in children with HA who had no significant association between reading and WM. Thus, for participating children who took part in a two-year follow-up post intervention,

decoding and passage comprehension levels were comparable to those of NH, and similar cognitive strategies were observed.

Reading intervention is considered effective if the effect sizes are greater than 0.13-0.23 (see Torgesen et al., 2001). Effect sizes greater than this were found on phoneme-grapheme correspondence (letter sounds, $\eta\rho^2 = .30$), word-decoding accuracy ($\eta\rho^2 = .40$) in all children, and for nonword-decoding accuracy ($\eta\rho^2 = .41$) in NH children, and as a reduction of nonword decoding errors ($\eta\rho^2 = .26$) for DHH children. The Phonological representation task ($\eta\rho^2 = .32$) and the Nonword repetition task ($\eta\rho^2 = .43$) also reached the recommended effect-size level in children with weak initial PhPS. The latter may be observed as transfer effects since there were no exercises in the intervention program that trained these skills particularly. Transfer effects were also observed in nonword decoding.

Clinical implications

Although, mainly a trend in the data, children with HA seemed to have weaker reading development, observable at all levels assessed; letter knowledge, decoding and reading comprehension. These results point to the importance for SLPs to assess the language and reading development in these children continuously and to give directed advice to parents and teachers of techniques to facilitate language and reading.

The Phonological representation task was easy to use and served as a sensitive and broad measure to identify children who benefitted the most phonologically from the computer-assisted intervention. These findings need to reach clinicians who work with DHH children or children with language impairment, since the task may assist in the identification of children in need of phonological intervention.

Letter knowledge skills served as a mediating factor for phonological change in children with weak PhPS. This may imply that other clinical groups, who are at risk of language and reading difficulties, for example children with language impairment and children at risk of developing dyslexia, could benefit from the reading intervention in a similar way. SLPs and teachers need to adopt and evaluate this approach in their clinical intervention guidelines much earlier than is customary today, particularly due to reading ability being one of the most important factors for academic success.

The findings relating to nonword repetition and associations to nonword decoding in children with CIs gave several clinical implications. To support DHH children's phonological decoding strategies phonological training should aim to improve their cluster production skills and syllable structure of words.

CONCLUSIONS

- Children who are deaf and hard of hearing, using cochlear implants or hearing aids constitute a heterogeneous group. Several factors, such as, age at diagnosis, duration of unaided hearing, and degree of hearing loss, contribute to the variation.
- Early intervention is the most crucial factor, which serves as a foundation for later, successful cognitive development.
- Overall, the results from the present thesis support the notion, that offering a computer-assisted intervention program delivered at home, is an alternative way to support deaf and hard of hearing children's phonological development and decoding proficiency.
- Specifically, children with a longer duration of unaided hearing and a more severe hearing loss benefitted comparatively more from the intervention.
- The results from the present thesis may be seen as a contribution to fulfill the theme of UNESCO for 2014: "Equal Right, Equal Opportunity: Education and Disability". Particularly, by acknowledging reading ability as one of the most important tools in the education of deaf and hard of children.

FUTURE DIRECTIONS

Children with hearing loss using hearing aids need to be followed carefully through childhood, particularly regarding reading acquisition. Optimally, regular assessment of letter knowledge skills and phonological sensitivity should be performed at the clinic.

The findings regarding nonword decoding in children with cochlear implants point to the need of deeper analyses of these children's reading strategies. One possible way to proceed is to further investigate how larger language units, such as the morpheme, come into play orthographically.

Further, it would be interesting to study how and in what order orthographic entities are formed, used and automatized.

SWEDISH SUMMARY (SVENSK SAMMANFATTNING)

Bakgrund

Barn med hörselskador som använder cochleaimplantat (CI) eller hörapparat (HA) utgör en heterogen grupp avseende språk- och läsförmåga. Olikheterna kan på många sätt förklaras av barnens skiftande bakgrundshistoria som grad och orsak till hörselskadan och tidpunkt för diagnos. Likaså skiljer sig barnen åt avseende kommunikationssätt. Övervägande delen av familjerna använder det talade språket som första språk men många barn behöver stödtecken, och en liten andel har döva eller hörselskadade föräldrar där teckenspråket är första språk.

Hörselskadade barn utvecklar talat språk utifrån en förvrängd och grumlig talsignal. Likaså har så gott som samtliga haft kortare eller längre perioder av dövhet eller oförstärkt hörsel. Dessa faktorer i kombination med att de tekniska hjälpmedlen inte kan återställa skadade hårceller eller återskapa normal hörsel, får konsekvenser på såväl tidig som senare språkinlärning och kognitiv utveckling. Många av de hörselskadade barnen kommer således behöva språklig intervention under sin uppväxt och skoltid, något som kompliceras av att familjerna dels är utspridda över ett stort geografiskt område, liksom att barnen ingår i olika pedagogiska miljöer.

Huvudsyftet med denna avhandling har varit att undersöka om det är möjligt att genomföra datorbaserad lästräning i hemmet för hörselskadade barn med stöd av logoped och föräldrar. Anledningen till att vi valde ett lästräningsprogram och inte ett mer övergripande talspråksträningsprogram var att vi dels ville koncentrera träningen kring något som alla barn behöver lära sig, att läsa, dels för att stärka uppfattningen av språkets minsta byggstenar, fonemen. Interventionsprogrammet som vi har använt erbjuder barnvänliga övningar som syftar till att stärka kopplingen fonem-grafem i olika spelmiljöer. Svåraste nivån erbjuder träning av sjubokstavsord. Orden i programmet är huvudsakligen ljudenligt stavade för att på ett tydligt sätt åskådliggöra sambandet mellan tal och skrift. Barnen lyssnar på bokstavsljud, korta och längre ord och ska sedan klicka på motsvarande bokstavssymbol, skrivna ord på skärmen. Likaså erbjuder programmet enklare övningar för stavning.

Delsyften i avhandlingsarbetet har varit att studera effekter av träning på fonologisk bearbetningsförmåga och kognitiva faktorer som är associerade till denna. Ytterligare delsyften har varit att undersöka läsförmåga, d.v.s. avkodning och läsförståelse och hur

kognitionen spelar in för läsförändring som en funktion av träning. Ett slutligt delsyfte var att detaljerat undersöka hur barn med bilaterala CI repeterar respektive avkodar nonord i jämförelse med åldersmatchade normalhörande barn.

Deltagarna i avhandlingsarbetet var totalt 48 barn. Trettiofyra barn använde CI och/eller HA. Sexton barn med normal hörsel (NH) utgjorde referensgrupp. Samtliga 48 barn undersöktes i Studie I-III och 22 barn undersöktes i Studie IV. Studien hade en kvasi-experimentell design med tre testtillfällen. Varje testtillfälle ägde rum med fyra veckors mellanrum och omfattade prövning av fonologiska färdigheter och bokstavskänedom. Testerna för fonologiska färdigheter sammanlades till ett fonologiskt kompositmått för att möjliggöra gruppjämförelser och övergripande kunna studera effekter av träning. Testtillfälle två och tre omfattade även prövning av komplext och visuellt arbetsminne, ickeverbal kognitiv förmåga och läsning. Alla deltagare övade i snitt 7 min per dag under fyra veckors med lästräningsprogrammet.

Studie I visade att barn med hörselskada uppvisade stor variation avseende fonologisk förmåga. Endast 20 procent presterade inom normalvariationen för normalhörande barn. Detta var barn med HA, undantaget ett barn med bimodalt hörande (CI på ett öra och HA på det andra). Gruppjämförelser vid första och sista testtillfället visade att barn med CI uppvisade stora svårigheter med fonologiskt arbetsminne, medan barn med HA hade svagare bokstavskänedom. Interventionen visade generell positiv effekt på samtliga barns bokstavskänedom, och en signifikant förbättring av fonologisk bearbetningsförmåga hos barn som hade ett svagt fonologiskt utgångsläge. Detta var i övervägande del barn med CI.

Studie II pekade på att barn med hörselskada och barn med normal hörsel använde delvis olika kognitiva strategier när de utförde uppgifter för fonologisk bearbetning. Hos barn med hörselskada samvarierade komplext arbetsminne med det fonologiska kompositmättet medan motsvarande korrelation hos normalhörande barn observerades med ålder. När vi studerade sambanden mellan fonologisk förändring och kognitiva faktorer i grupperna utmärkte sig måttet för fonologiska representationer hos de hörselskadade barnen. Resultaten visade att barn med svag prestation på måttet på fonologiska representationer före intervention förbättrades förhållandevis mer fonologiskt.

Studie III visade att samtliga fem- och sexåriga barn hade en jämförbar läsförmåga både före och efter intervention. Vid sju års ålder hade barn med NH en bättre läsning än barn med hörselskada. Läsförändring hos barn med NH var associerad med fonologisk

bearbetningsförmåga och komplext arbetsminne, medan den hos barn med hörselskada var associerad med visuellt arbetsminne och bokstavsbenämning. Resultaten skulle kunna tyda på att den tidiga läsutvecklingen hos barn med hörselskada är mer beroende av visuell förmåga och ortografiska strategier. Detta kan vara orsaken till den långsammare läsutvecklingen hos de äldre barnen med hörselskada.

Studie IV visade att barn med CI hade stora svårigheter med nonordsrepetition i förhållande till barn med NH. Emellertid presterade de något högre än tidigare liknande studier har visat. Eventuellt kan detta härledas till att barnens nonordsrepetition i föreliggande studie prövades efter träning. Barnen med CI gjorde många fler stavelseutlämningar än barn med NH, och främst av stavelsen före den betonade i ord med sen betoning (s.k. jambiskt betoningsmönster, ex. svag-STARKT). Detta resultat pekar på att svenskans metrik (rytm- och betoningsmönster) påverkar inte bara hur normalhörande svenska barn tillägnar sig modersmålets prosodi, utan även hur svenska barn med CI repeterar nonord. Fler och kvalitativt annorlunda nonordsläsningsfel hos barn med CI pekar på att de använder fonologiska avkodningsstrategier utifrån den fonologi de behärskar bäst.

Resultaten från de fyra studierna pekar sammantaget på stor heterogenitet hos gruppen hörselskadade barn men att detta inte hindrar genomförandet av fonologisk lästräning i hemmet. Effekter av träning observerades främst hos barn med svag fonologi vilka oftare hade haft en längre duration av dövhet. Resultaten visar på delvis annorlunda kognitiva strategier hos gruppen hörselskadade barn både när de utför uppgifter för fonologisk bearbetning och läsning. Det är viktigt att logoped, pedagoger och föräldrar får ökad kunskap om detta så riktade insatser ges till barnen i unga åldrar.

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Cecilia Nakeva von Mentzer

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