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LEXICAL AND SEMANTIC DEVELOPMENT IN CHILDREN WITH COCHLEAR IMPLANTS

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“It is a journey and not a destination”
Warren Estabrooks

To my beloved sons Axel and Hampus

ABSTRACT

Introduction: Two important events have changed the prerequisites for children with severe-profound hearing impairment (HI). The first is the universal newborn hearing screening (UNHS) that enables early detection and identification of congenital or early-acquired HI. The second factor is the possibility of cochlear implant (CI) intervention at younger ages. There has been little previous interest in studying lexical-semantic ability in the new generation of children with CI who are implanted at a fairly young age, often bilaterally, in relation to the cognitive capacities that influence on use and knowledge of words.

Aim: The general aim was to explore lexical-semantic ability and development in a group of school-aged children with CI and in comparison with age-matched normal hearing (NH) children.

Method: The four studies examined different aspects of lexical-semantic ability in children aged 6-9 years. The cohort consisted of 34 children with CI and 39 age-matched children with NH (Study I-IV). In addition, two other clinical groups: children with language impairment (LI) (n=12) and children with autism spectrum disorder (ASD) (n=12) participated in Study II. A broad test battery was used, especially examining lexical-semantic ability but also non-verbal cognitive ability as well as phonological and learning ability. An error response analysis was conducted on a picture-naming test and a linguistic cluster analysis was performed on two different word fluency tasks with the purpose of examining strategies that the participants used when they retrieved words from their long-term-memory (LTM). Study IV had a partly longitudinal approach where a subgroup of children with CI (n=18) were examined more closely over time and in comparison with other age-matched groups at the age of 8-9 years. Statistical analyses were made primarily to examine group differences.

Results: Many children with CI reached age-equivalent lexical-semantic ability at the age of 8-9 years. Semantic knowledge and non-verbal cognitive ability predicted grammatical sentence understanding (GSU) in both groups (CI and NH). Children with CI used age-appropriate learning strategies and had similar cognitive capacities necessary for managing use and knowledge of words as NH controls. In addition, children with CI showed better outcome than children with LI or ASD. However, an atypical developmental pattern was found in Study II where children with CI had better expressive than receptive vocabulary compared to typically developed children with NH. Also, the variation of the outcome was overall greater in children with CI (Study I-IV). An age-related CI subgroup-pattern was found in Study I that led to the planning of a follow-up study, also examining retrospective, early spoken language development (Study IV). Children in the subgroup with higher group mean age at 1st CI had significantly poorer results on receptive vocabulary, phonemically based letter word fluency and GSU than NH controls. Despite of these results, the whole sample of children with CI had an age-appropriate level of expressive vocabulary and semantic feature knowledge. Semantic knowledge was demonstrated as an adequate ability to recognize semantic features and to use semantically relevant responses when lacking the lexical term while naming pictures (Study III). Children with a younger group

mean age at 1st CI had better early expressive language use, two years after CI-operation, than the other subgroup, and they were also able to catch up after school entry on receptive vocabulary and phonemically based letter word fluency ability (Study IV).

Conclusions: Children with CI and typical non-verbal cognitive ability did not have specific deficiencies in cognitive processing of lexical-semantic items, but some children had deficiencies with phonological and lexical-semantic knowledge. The results indicate that age at 1st CI is important as a starter engine of spoken language development through listening, but that other more linguistic-related factors and strategies also are important for development of lexical-semantic ability. There was a greater variation of the spoken language outcome in children with CI, and some atypical developmental patterns were found in the sample. Future studies of lexical-semantic ability in children with CI should therefore have a longitudinal approach and also explore the influence of environmental factors.

Keywords: children with cochlear implants, lexical-semantic ability, cognitive capacity, language development, non-verbal cognitive ability, age at CI intervention, language disorder, autism spectrum disorder

LIST OF PUBLICATIONS

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LIST OF ABBREVIATIONS

ASD	Autism spectrum disorder
AVT	Auditory verbal therapy
BNT	Boston naming test
CI	Cochlear implant
CIC	Cochlear implant clinic
CI-O	Children with CI and older ages at 1 st implantation
CI-Y	Children with CI and younger ages at 1 st implantation
cCMV	Congenital Cytomegalovirus
Cx26	Connexin26
dH HL	Decibel hearing level
dB SPL	Decibel sound pressure level
EI	Early intervention
gC	Crystallized intelligence
gF	Fluid intelligence
GSU	Grammatical sentence understanding
HA	Hearing aids
HI	Hearing impairment
HL	Hearing loss
Hz	Hertz
JLNS	Jervell and Lange-Nielsen syndrome
LENA	Language environmental analysis
LI	Language impairment
LTM	Long-term memory
NH	Normal hearing
PPVT-3	Peabody picture vocabulary test, 3 rd edition
PTA	Pure-tone average
PWM	Phonological working memory
SIR-2	Speech intelligibility rating scales, 2 nd edition
SLP	Speech-language pathologist
ToM	Theory-of-mind
WHO	World Health Organization

LIST OF DEFINITIONS (USED IN THE THESIS)

Breadth	Size of vocabulary
Cognitive capacity	Skills and strategies used for information processing that are built on underlying mental processing
Depth	Semantic knowledge including semantic feature knowledge, error response analysis, organisation of lexical-semantic networks
Figurative language	When something is said with the intention that something else should be understood e.g. metaphors, irony
Grammatical sentence understanding	The interpretation and receptive understanding of words in phrases and sentences
Lexical-semantic ability	Word knowledge and use of individual words in a person's vocabulary and in relation between words
Lexical-semantic representations	Word unit that incorporates both lexical form and semantic understanding
Mental lexicon	Storage and use of words and the organization of semantic, phonological and orthographic representations
Metaphor	Words that express figurative meaning like "it's a journey, not a destination"
Non-verbal cognitive ability	Describes a child's fluid intelligence; the ability of reasoning and solving new problems without using prior acquired knowledge but instead adopting abstract thinking
Picture naming	The ability of visually interpreting a picture and retrieving the label and corresponding concept from the long-term memory (LTM) simultaneously by selecting the right lexical-semantic and phonological representations and then mobilizing the lexical-semantic item on articulation level
Semantic knowledge	The actual or interpreted meaning of words or sentences
Storage of language	Retainment of memory traces concerning linguistic items on phoneme, morpheme, word and sentence level
Word finding	Storage and retrieval of word meaning
Word fluency ability	The process of retrieving specific words from LTM

1 INTRODUCTION

This thesis is an evaluation of intervention in a group of children who had little or no hearing ability when they were born. The intervention consisted of implantation of a cochlear implant. This is a hearing device that does not restore hearing to a normal level, but enables children to develop listening, spoken language communication as well as higher-level cognitive and language skills (Peterson, Pisoni & Miyamoto, 2010). The variation of outcome level is large within this small and heterogeneous population and the reasons for this variation will be described later (Duchesne, Sutton, & Bergeron, 2010; Geers, Strube, Tobey, & Moog, 2011; Peterson et al., 2010; Spencer, 2004).

The results of the four studies showed that many of the participating children with CI reached age-equivalent lexical-semantic ability results by the time they were 8-9 years. This was especially pronounced for those with younger ages at 1st CI implantation and relatively high levels of non-verbal cognitive ability, and a better semantic ability. In addition, some children with higher ages at CI intervention reached higher levels of lexical-semantic ability. This indicates that there are also other more linguistically related factors that influence vocabulary development in this heterogeneous population.

The origin of this project was clinical findings from children's follow-up visits at the cochlear implant clinic (CIC), Karolinska University Hospital. These showed that many children had significantly worse vocabulary ability, especially expressive vocabulary, compared with norm data and in comparison to their level of sentence understanding. This clinical finding was confusing and the seedbed for some specific research questions that resulted in the current PhD-project. The project plan included four specific studies aiming to explore various aspects of lexical-semantic ability on word level in children with CI of different ages, from infants to school-aged children. However, writing a thesis is a process and original plans seldom stay the same during the whole research journey. This thesis constitutes no exception. The initial findings from the first and second studies raised some new specific questions that led to the planning and conduction of a follow-up study of a subgroup of the cohort, with children with a lower group mean age at 1st CI intervention. It turned out that the 34 children with CI had a lot to contribute in the field of lexical-semantic ability both within the population and for children with NH and other clinical groups in the same ages.

The picture of an Egyptian sphinx on the cover of this book comes from one of the tests that were used in the project called the Boston Naming Test (BNT; Kaplan, Goodglass & Weintraub, 1983). A sphinx originally had the function of being the guard of the pyramids in Egypt. The word and concept of sphinx is said to symbolize strength, with a body of a lion and enigmatic richness, specifically meaning wisdom and intelligence, symbolized by the head of a human. These two specific symbols could also describe the meaning of the word *language*: rich, powerful and connected to cognitive abilities. Like the sphinx, language is also mysterious and not yet fully explained and the source of different theories regarding its true origin. In this work, the sphinx is not only the guard of the book. Neither is the sphinx only a metaphor for the concept of language.

The sphinx is also part of the answer of one of the main research question: how do children with less favourable conditions, in this case deaf or HI children with CI, develop lexical-semantic ability? One finding was that children with CI and better semantic knowledge, who for example could name low frequency words like the *sphinx* when they were 6-7 years, had better receptive vocabularies at the age of 8-9 years.

This thesis is a description of a unique sample of Swedish school-aged children with CI and their lexical-semantic ability, and how they compare with other groups of children. This has not been studied previously and will therefore potentially be an important contribution to the field of knowledge about spoken language development, especially vocabulary, in a new generation of children with CI meaning children who were implanted at fairly young ages, in most cases bilaterally and who are integrated in mainstream school settings.

2 BACKGROUND

2.1 LANGUAGE AND COGNITION

Use of language includes both socio-cognitive and linguistic-specific abilities (Bloom, 2002; Kuhl, 2010). Hearing ability is essential for development of both listening skills and spoken language (Houston and Bergeson, 2013). Language is universal to a certain degree (Berwick, Friderici, Chomsky, & Bolhuis, 2013) but also dependent on neuro- and socio-cultural aspects in how it is developed during childhood (Locke, 1997; Tomasello, Carpenter, Call, Behne & Moll, 2005; Vygotsky, 1978). The spoken language system incorporates domains like phonology, grammar, pragmatics and semantics (Bishop, 1997). Some of the universal characteristics within these language domains are easier to acquire for a young child than some of the more language-specific features that are learned later as a consequence of cognitive maturation (Berglund, Eriksson, & Johansson, 2001; Bloom and Markson, 1998; Noorgaard, Dale, Bleses, & Fenson, 2010). One could say that there is a mutual exchange between language and cognition (Bloom, 2002; Tomasello et al., 2005). Furthermore, within specific language and across different language borders there are clinical groups like children with language impairment (LI) or different levels of hearing impairment (HI) who have some more distinctive and group-specific language difficulties in common (Briscoe, Bishop & Norbury, 2001; Salameh, Håkansson & Nettelbladt, 2004). This thesis concerns one part of the language domains, namely the semantic knowledge and lexical-semantic ability in particular and also the possible influence of cognitive capacity. This is examined in a specific group of Swedish children with severe-profound hearing impairment (HI) and CI, who endured a period of auditory deprivation before implantation. *Lexical-semantic ability* refers, in this project to word knowledge and use of individual words in a person's vocabulary and relation between words.

Semantic knowledge is not only related to the linguistic domains, but also to most cognitive aspects (Baddeley, 2012; Jerger, Tye-Murray, Damian & Abdi, 2013; Tulving, 2002). The meaning of e.g. *sphinx* or a more commonly used word like *cow*, is not necessarily the same for all individuals (Tulving, 2002). The exact meaning of *sphinx* or *cow* also depends on in which context they occur, and in relation to type of sentence and/or communicative situation. If a person was asked to describe what a cow is in more detail, the answer probably would be "an animal" or one might even say "a farm animal" which demonstrates hierarchical word knowledge, by using superordinated words for the concept (Ahlsén, 2006). And if the same person is asked to describe the cow in even more detail, the answer could be associations like "an animal that gives us milk, that usually lives in a farm, have horns, hooves and often is brown and white". This tells us something more about this person's level of associations and feature knowledge of the concept and may therefore demonstrate *depth* and *breadth* in the *lexical-semantic network* around the word *cow* (McGregor, Oleson, Bahnsen & Duff, 2013). Word knowledge that is shared by other persons is part of the *semantic memory* (Tulving, 2002). Personally, if I was given the question I would probably simultaneously think about the cows that I have met during my summer holidays which would be part of my world knowledge in relation to the concept of cow, as it is

something that I relate to in a specific way (Tulving, 2002). And, if I would remember cows that I met at a specific year and place, that would refer to an *episodic memory* (Tulving, 2002). If a person from India would be asked the same question, the answer would probably be partly different, even if the animal and the concept are basically the same and have similar outer features, although expressed in another language. Cow is named “ko” in Swedish and “gaja” in Hindi, one of the official languages in India. However, not only the lexical term but also the *semantic meaning* of the word cow is different for a person in India, especially if the person lives in the countryside and is religious in comparison with a person with sparse knowledge of cows and without any religious connections of the concept (Tulving, 2002). A Swedish or Indian person might actually have more common knowledge about the word and concept of a sphinx compared with a cow. Probably neither of them would use the word sphinx frequently or have any personal connection to a sphinx, but still they would know or recognize the word and might have seen a sphinx in pictures or read about them in books. The cow and the sphinx can both be mythological concepts, depending on whom we ask about the meaning or explanation of the two concepts (Tulving, 2002). It would also probably be easier for both the Swedish and the Indian person to name cow before sphinx in a picture-naming task, due to stronger lexical-semantic representations of cow than sphinx (Jerger et al., 2013). This could be demonstrated as more detailed semantic knowledge about cows than for sphinxes, illustrated as larger and more robust lexical-semantic networks for cows, although with different individual semantic knowledge (Jerger et al., 2013, Tulving, 2002). Luria was a scientist who put forward theories in modern history about how the brain works as a functional system that can be divided into blocks and zones and how these interact (Luria, 1973). Since then, many researchers have been interested in the study of interaction of cognitive capacity and language and have examined brain activity in different ways, including in children with different clinical background such as children with hearing impairment (Kral and Sharma, 2012).

This brief introduction aimed to describe some of the dimensions of lexical-semantic ability in relation to aspects of semantic memory including episodic memory and semantic memory. Now follows a description of the *cognitive capacity* that is necessary for language development and lexical-semantic ability as well as a short description of atypical conditions that might have a negative influence on lexical-semantic development in childhood.

2.1.1 Cognitive capacity influencing on spoken language

There are different levels and dimensions of cognition and many of these are closely related to language processing and production including the lexical-semantic domain (Baddeley, 2012; Bloom, 2002; Hart and Risley, 1995). Cognitive capacity is essential both to word learning, current use and understanding of lexical terms and deeper semantic knowledge (Bloom and Markson, 1998; Baddeley, 2012; Nippold and Duthie, 2003). Focus in the present thesis is on the cognitive concepts of intelligence, working memory, long-term memory, theory-of-mind and executive functioning. These are described in some detail below.

Intelligence can be divided into *fluid intelligence* (gF) which can be described as the ability of reasoning and solving new problems without using prior acquired knowledge but instead adopting abstract thinking and e.g. performing activities like puzzles, and *crystallized intelligence* (gC), which in contrast to gF, is a measure of skills acquired through prior experience and knowledge connected to e.g. vocabulary (Nisbett et al., 2012). The two types of intelligences are both equally important for e.g. lexical-semantic ability and often there is a positive correlation between e.g. gF-ability and expressive vocabulary (Storms, Saerens & De Deyn, 2004). Fluid intelligence is developed during childhood and adolescence and declines in middle age, while gC may develop throughout adulthood and only decline gradually in old age (Nisbett et al., 2012). Intelligence is usually at the same fundamental level from birth and onwards, but develops gradually during childhood as a result of experiences of the world and language (Nisbett et al., 2012). However, gC intelligence can be positively influenced by higher level of language competence (Deary, Penke, & Johnson, 2010; Hart and Risley, 1995). Deary et al. (2010) concludes in their review that studies of intelligence benefit from having a developmental perspective from infancy to old age. This enables understanding of both the continuity of intelligence and individual differences due to changes during various phases during a lifetime.

Memory systems. Tulving (2002) has influenced the view that the human LTM system is separated into a *semantic* and an *episodic memory* system. The episodic memory incorporates personal memories, linked in time and place. Semantic memory includes aspects such as world knowledge and memory of language among other things. Another LTM system is the procedural memory. This develops gradually after a long period of practice and then becomes more or less unconscious like riding a bike, knowing the alphabet or reading.

Another theory of memory is the multi-component working memory model by Baddeley (2012). This is a memory system for temporary storage and processing of information over a short period of time. The model includes four different components, each responsible for different aspects of processing and storage of information. The central executive is responsible for planning, co-ordination and execution of cognitive operations. The central executive has a limited storage capacity. The phonological loop is responsible for short-term storage of phonological related stimuli and the visuo-spatial loop has a similar responsibility for visual and spatial material. The episodic buffer is responsible for binding of new incoming information and old information (e.g. language related information) stored in LTM.

The model has been used in several studies of children with NH and clinical groups, including children with HI, to explore the mechanisms of e.g. the role of the phonological loop in language developmental processing and aspects like word learning (Gilbertsson and Kamhi, 1995; Hansson, Forsberg, Löfqvist, Mäki-Torkko & Sahlén, 2004; Masoura and Gathercole, 2005; Willstedt-Svensson, Löfqvist, Almqvist & Sahlén, 2004).

Theory-of-Mind (ToM) is another important socio-cognition-related capacity that develops over time and that is crucial for social as well as linguistic development (in particular pragmatic skills) that interact closely with semantic knowledge (Bloom, 2002). Theory-of-mind is also referred to as *mentalization* and could be defined as the ability to use and understand another person's intended meaning of an utterance or sentence, including interpretation of prosodic patterns for expressing irony, emotions and underlying statements in communication with others (Fischer, Happé & Dunn, 2005; Sundqvist, Lyxell, Jönsson, & Heimann, 2014). Bloom and Markson (1998) argue that the underlying capacity for word learning in children, ToM, is demonstrated by their ability of intuitive learning of new words in interaction. Theory-of-mind influences the word learning process when children contemplate the thoughts of other people (Bloom, 2002).

Executive functioning (EF) refers to the ability to inhibit irrelevant information, to update the cognitive system with new, incoming information and to shift between different sources of information (Diamond, 2013). Executive functions are connected to frontal lobe capacity (Kavé, Kigel & Kochra, 2008). The higher-level aspects of EF are not fully matured until early adulthood (Diamond, 2013). These abilities e.g. enable individuals to pay *attention* to what they hear, what the expectations of them are in certain situations, to stick to rules and to stay focused in communication or while performing a verbal or non-verbal task, for example language assessment (Miyake and Friedman, 2012).

Phonological awareness refers to the ability of being aware and sensitive to the speech sound structure of spoken language (Melby-Lervåg, Lyster & Hulme, 2012). Phonological skills are important for detecting similarities and differences of speech sounds important for tasks like rhyming and manipulating language units which has been found to be important for literacy (Wass et al., 2008).

Word finding. There are different terms used by researchers that describe the availability of a person's mental lexicon. One term is word finding and another is word retrieval (Messer and Dockrell, 2006). Word finding includes both storage, which encompasses vocabulary size and organization, and retrieval of words, while word retrieval only describes the process of retrieving words from storage (Messer and Dockrell, 2004).

In summary, cognitive capacity and cognitive processes are closely connected to semantic knowledge and lexical-semantic ability (Baddeley, 2012; Bloom, 2002; Jerger et al., 2013; Wass et al., 2008). Therefore it is beneficial, and often necessary, to examine both when conducting studies of either domain.

2.1.2 Lexical-semantic development is a life-long project

There is a large individual variation regarding milestones in development of spoken language in children with NH and typical development (Bishop, 1997; Bloom and Markson, 1998). Language variation is typical also for adults and can for instance be demonstrated as different levels of literacy knowledge, vocabulary size and use of

figurative language (Kavé and Yafé, 2014). Education and older age is one important factor for this variation (Kavé, Knafo & Gilboa, 2010). To understand the impact of what an early acquired or congenital HI might impose on a child's linguistic and cognitive development, it is important to relate it to typical development in NH children. Therefore, a brief summary of lexical-semantic development with a lifetime perspective is presented below.

0-12 months: Spoken language development starts before a child is born, from the 20th week of pregnancy, when the cochlea is mature (Decasper, 1986). At birth, NH children can already recognize their mother's voice and even remember music that they have heard before birth (Granier-Deferre, Bassereau, Ribeiro, Jacquet & Decasper, 2011). Mampe et al. (2009) has also found that newborn infants have different kinds of screams, measured as language-specific prosodic patterns. From birth and until the child is around 6 months the child seems to be able to learn and categorize between any speech sounds and language (Kuhl, 2010). From this age on the child starts to reorganize the initial phonetic and phonological system and becomes more and more tuned to the linguistic patterns of the child's surrounding language, specifically the mother tongue of their caregivers (Lacerda, 2005). In parallel to speech reorganisation, the young child gradually starts to use the voice with more intent, imitates vocalisations and learns to use speech sounds in a more communicative way (Kuhl, 2010). Then, at around 8-10 months of age, an important milestone in language development starts to occur, when the child begins producing *canonical babbling* (Oller, Eilers, Neal & Schwartz, 1999, Moeller et al., 2007a). The fundamental building blocks of spoken language are gradually established; the phonetic system (Kuhl, 2010; Lacerda, 2005), preverbal skills including communication patterns (Bloom, 2002) and perceptual identification of suprasegmental information that is important for language understanding (Kuhl, 2010) and word learning (Moeller et al., 2007b). Natural gestures and body language are also part of the spoken language development from early ages (Miller and Gros-Louis, 2013) and through life (So, Yi-Feng, Yap, Kheng, & Yap, 2013).

To summarize, children learn the most important basis of spoken language during their first year of life (Kuhl, 2010; Lacerda, 2005; Oller et al., 1999;). And this is done through social and oral interaction (Bloom, 2002; Miller and Gros-Louis, 2013).

13-24 months: When a young child with NH has expressed their first spoken words, at around 8-15 months of age, an intensive word learning process has started, even if the foundation for this development was made long before the child started to use own words (Kuhl, 2010). However, the first words are learned slowly (Bloom and Markson, 1998). The child has to accomplish two things: recognize familiar strings of sound in the speech signal and attach the meaning of the sounds to an object. At around 18 months, when the child knows about 50 words, the child starts to combine words in small sentences and now understands more than she or he can express (Bloom, 2002). In the coming years, the vocabulary usually grows quickly and almost miraculously, with little effort from the child (Bloom and Markson, 1998). However, studies show group-differences of vocabulary outcome within the normal population of children at this age, where some children might use around 250 words and others only 10 (Fenson, Dale, Reznick, Bates, Thal & Pethick, 1994). The child is considered to be highly

active in this learning process and at the same time benefits from a supporting environment (Kuhl, 2010; Fernald, Marchman & Weisleder, 2013). Children have different learning strategies, but most children start to use nouns and focus on names for persons, animals, toys and food before they start to use action words like verbs (Clark, 1993). The age between 18 and 24 months is usually called the *word spurt period* because of the rapid learning of new words (Blom and Markson, 1998).

2-5 years: At this developmental phase phonology continue to develop and the child also starts to focus more on syntactic and grammatical learning, including morphological markers. Later, at the time when a child is around three years old, the child learns around 4 words per day (Fenson et al., 1994). Still, there are some sounds that might be more difficult to pronounce like the r-sound and s-sound in Swedish and the combination of consonant sounds i.e. st-ocking. The influence of phonology and grammar is evident in this phase of vocabulary development (Bloom and Markson, 1998). Moreover, from this point the child also starts to discover and become more phonologically aware and interested in rhyming (Whitehurst and Lonigan, 1998).

6-7 years: At this age a child learns around 7 words per day (Fenson et al., 1994). Just before school entry some children start to learn to read and are now aware of that words consist of letters that correspond to speech sounds and in parallel they also start to reorganize their vocabulary (Cronin, 2001; Nelson, 1977). Old, already established words and concepts get deeper meanings as the child starts to relate these words more in hierarchical order and in relation to more detailed world knowledge of an already known object (Nelson, 1977). For example, this can entail knowledge about that an elephant being not only an animal with a trunk but also one that has tusks and who lives in Africa or India. One finding from a recent study of NH children 6-9 years, by Monzalvo and Dehaene-Lambertz (2013), showed that only one year of reading instruction was sufficient to increase activation in brain regions involved in phonological representations and sentence integration.

8-18 years: At school entry and onwards a child learns around 12,1 words per day and at the time when the child leaves high school he/she understands around 60 000 words (Bloom and Markson, 1998). Literacy is an important source for this *second word-spurt*. Abstract words and low frequency words that seldom are used are generally learned later than more concrete and high frequency used words. The expansion of vocabulary requires reorganisation into categories of words and this is now done both in semantic and phonemic categories. In parallel, and as a result of learning many new words during school years, children continue to get more associations of concepts and gradually develop the connections between words and concepts in their lexical-semantic network. Inference ability and syntactic awareness are important for school age children to understand definitions of unfamiliar words (Marinellie, 2010). Lastly, figurative language and complex metaphors are usually not learned and understood until later, in higher school years (Nippold and Duthie, 2007). Socio-cultural aspects influence on word learning as well as influence from learning other languages in school.

Adulthood: Vocabulary learning is a life-long commitment and includes both world knowledge and processing skills (Kavé, Knafo, Gilboa, 2010). The difference for

children and adults is the reduced ease of learning new words; children may learn a new word after only one exposure (Bloom, 2002). At the same time children have more difficulties in the sense that they have to use bottom-up strategies in word learning and language understanding, while older children and adults have the possibility of using top-down processes, meaning that they can fill in gaps and interpret sentences and words by using already established linguistic knowledge. This means that they are not as vulnerable to worse listening environments as NH children, as they can more easily understand despite not hearing the whole acoustic message. Lexical-semantic acquisition involves both world knowledge and processing skills. However, all individuals, irrespective of age and hearing ability benefit from better acoustics (Cole and Flexer, 2007).

To summarize, children in early school years (6-9 years) are in an intensive period in their lexical-semantic development. They have gone through an intensive initial word-learning phase in their early childhood and are now at a stage where they are becoming more focused on the deeper meaning of words leading to a reorganisation of their vocabulary. Additionally, after school entry they increase their breadth and depth of vocabulary significantly. Reading and knowledge learning contributes to the extension of vocabulary. This is done at the same time as when they are developing higher level phonological and literacy skills in conjunction with socio-pragmatic competencies, both important for their continuous word learning processes throughout life.

2.2 LEXICAL-SEMANTIC KNOWLEDGE AND ORGANISATION

Semantics has been studied in many different disciplines like linguistics, neuropsychology, philosophy and lately also computer science. The view of the true nature of semantics has been debated since the time Plato lived and is yet not uniform. In this thesis, semantic is primarily studied by exploring the linguistic and neuropsychology perspective with a focus on word meaning and word use, on a lexical and semantic level.

2.2.1 Lexical-semantic knowledge

One definition of lexical knowledge is that it is the ability to recognize lexical units and to connect certain strings of speech sounds and supra-segmental patterns, related to lexical units, to a specific meaning and further to relate these lexical-semantic units hierarchically to super- and subordinated words and categories (Nelson, 1977). In the word-learning process, phonological representations of a new word are linked to a meaning and then categorized and placed in the mental lexicon storage (Jerger et al., 2013). The same lexical unit can be connected to different categories and may have a different meaning for individuals or subgroups. The meaning of a word is also affected by the syntactic, prosodic or situational context. One example of this phenomenon is the previously mentioned word *cow*, which means different things for a child that has never seen a cow or for a child that lives on a farm. The lexical term can be recognized and repeated, but the semantic knowledge differs (Tulving, 2002). Another example is the sentence “The sphinx drank all the milk” which is correct in terms of lexical and syntactic rules, but from a semantic perspective sounds strange and unlikely, unless it is refers to a sphinx in a fairy tale, where everything is possible, or perhaps is expressed

with a humorous tone of voice. In summary, lexical-semantic knowledge is complex, and the same lexical term can mean different things depending on factors like individual semantic knowledge (Tulving, 2002), lexical and grammatical rules or different socio-linguistic manners when talking about an object.

2.2.2 Semantic storage

Some theories of the semantic memory system and how e.g. semantic knowledge is stored have been mentioned already (Tulving, 2002; Baddeley, 2012). According to another, more linguistic theory, the *Hierarchical network model of semantic memory*, semantic memory is stored and organized hierarchically, basically divided in three different levels (Collins and Quillian, 1969). The most general level is the super-ordinated category e.g. “animal” or “food”. The second level category contains more specific information of the general super-ordinated category e.g. “farm animal”, “vegetable”. The third level is the most specific exemplar of a subcategory concept, like “carrot” for vegetable or “cow” for farm animal. This hierarchy system is believed to be built-up from early ages as a consequence of an individual’s language experiences, general cognitive maturation and reorganization of the vocabulary (Nelson, 1977; Cronin, 2001). One finding that confirms this adoption is that children usually do not associate a word like cow with the word mammal until they reach a certain age. Depending on their age they would instead associate the word cow with words like “milk” or auditory descriptions like “a brown and white animal that gives milk and says moo”. Research indicates that the reorganisation of words starts around 6-9 years of age, which means that children go from using more syntagmatic associations to more paradigmatic associations (Nelson, 1977). One example of a shift could be to name a sphinx with “statue” instead of “pyramid”.

Other theories concern the organization of lexical-semantic representations of concepts in categories, meaning that they are composed of different pieces of information, called *semantic features*. Early developed association fields in spoken language development are words related to categories like food, body parts, clothes, family members, daily routine activities, animals and toys (Clark, 1993; Fenson et al., 1994). One way of defining concepts according to their semantic features is to divide them into two categories: (1) the first category-level containing features that are shared by many members of a concept like mammals and therefore will not distinguish one member from another (2) the second category-level containing more specific features that will distinguish a member from another (cow-cat-lion) (see Figure 1).

Yet, another way of describing semantic features is to base them on typical representations of a concept or word. Rosch (1975) introduced the so-called *Prototype theory*, proposing that categories of words are organized around some specific prototypical words, meaning the most typical exemplar of a category, which could be defined as the most robust lexical-semantic representation on a concept. These words are usually learned first by children and are named more quickly and easily than less typical exemplars (Sigurd and Håkansson, 2007) (see Figure 2).

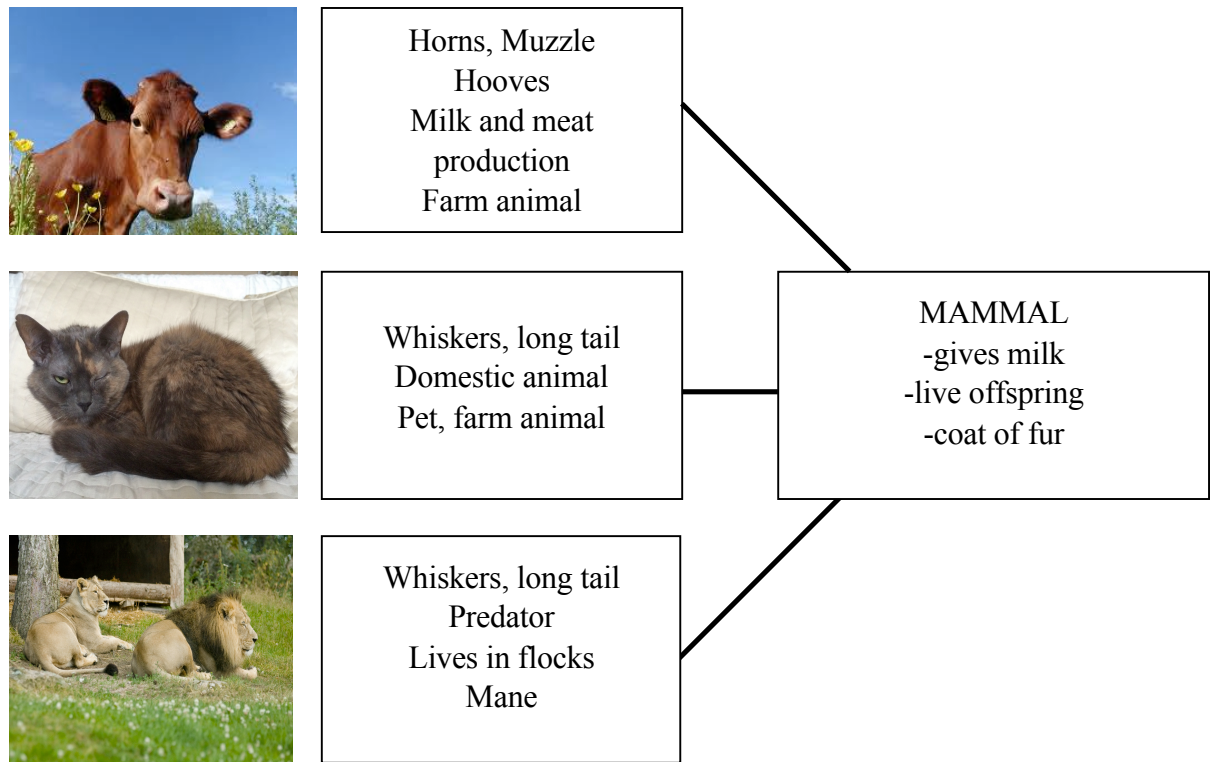


Figure 1. Illustrating *semantic feature knowledge* of cow, cat and lion, with two category levels.

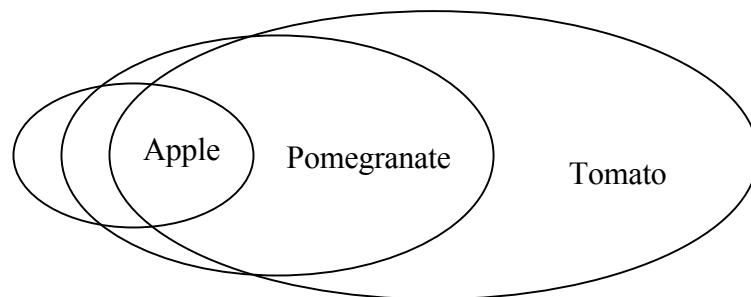


Figure 2. *Prototype* example for the word FRUIT with apple representing a more typical exemplar and tomato representing the most atypical exemplar of fruit.

Semantic distinctive features have also been used to describe linguistic categories, as illustrated in Figure 3. In this way, necessary and sufficient conditions for belonging to certain categories are defined (Ahlsén, 2006). Adults have no difficulties in defining categories in distinctive semantic features and children with typical development seemingly learn this by themselves, with age.

	Living	Non-living	Male	Female
Cow	+	-	-	+
Sphinx	-	+	(+)	(+)
Girl	+	-	-	+

Figure 1. *Semantic distinctive features* of three words (cow, sphinx and girl).

Finally, another clinically based theory aiming to understand the nature of how semantic feature knowledge is processed and learned have been presented by Patterson, Nestor & Rogers, 2007. Patterson et al. (2007) argue that every single task, regardless of how it is perceived, engages and activates all senses, movement, shape and function. The idea of a broader perception and total experience, including the sensorimotor activation, requires the semantic memory to be constructed in a particular way. The evidence for this assumption was the finding of patients with semantic dementia and injuries in the anterior temporal lobe, with specific symptoms like restored episodic memory but deficient semantic memory. The authors claimed that linguistic information that is activated passes a common *hub* in the brain, situated in the anterior-temporal lobe. *Semantic feature knowledge* could thus be described as conceptual knowledge built on a joint sensory, motor and linguistic system, with the so-called distributed *Distributed-plus-hub* view, see Figure 3. According to Patterson and colleagues (2007) the hub is where all the information concerning a concept has to pass while e.g. an individual recalls a word or interprets a word instead of the more traditional belief that certain linguistic domain are situated only in specific areas in the brain. One example for this could be that when a person thinks of a word like a tomato, the colour, shape, taste and word are activated simultaneously. If the word were cow, the specific movements of cows would also be activated simultaneously (Patterson et al., 2007). With a more traditional model, the word tomato or cow would be activated, separately, in specific areas, and not jointly.

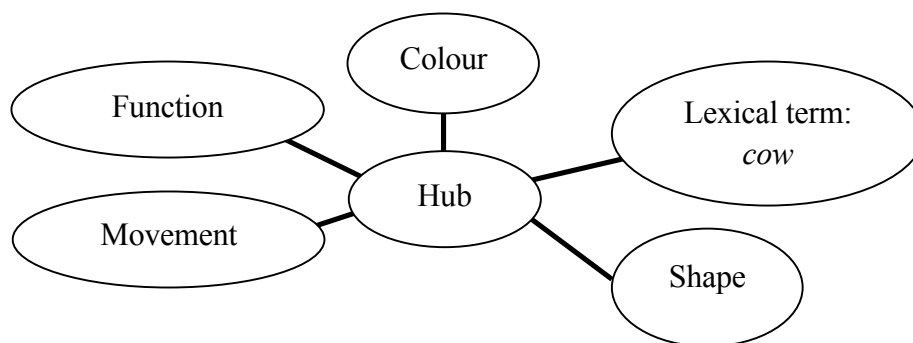


Figure 3. Schematic model of *semantic feature knowledge* activation according to the Distributed-plus-hub view by Patterson et al., 2007.

To summarize, different theories discuss the nature of lexical-semantic representations from different perspectives but seem to agree on that there is a hierarchical order, developed through childhood and that features of lexical-semantic concepts build on already acquired knowledge with a broad and deep semantic perspective including knowledge of the world and perceptual experiences. Lexical-semantic ability is sometimes divided in two categories; breadth and depth. These two terms distinguish between size, organisation and semantic knowledge and can also easily be implemented into some of the common assessment tools that are usually used both clinically and in research for examining vocabulary development (Storms Saernes & De Deyn, 2004; Tallberg et al., 2008). Additionally, other more qualitative analyses for estimation of linguistic clustering or error response analysis in picture-naming tasks, may both give information both of size (breadth) and semantic knowledge including lexical-semantic networks (depth) (Tallberg et al. 2011). These two categories are therefore applicable when describing typical development, methods and some of the results of the project.

2.2.3 Breadth in lexical-semantic ability

Receptive vocabulary includes words that are understood when we hear or read them in a context and encompasses a larger numbers of words than expressive vocabulary that can be described as words we use when we talk or write. Receptive word knowledge is learned earlier than expressive vocabulary use and highly relies recognition of phonological and lexical-semantic representations (Stoel-Gammon, 2011). Children usually therefore understand much more words than they can use themselves. In early childhood situational context understanding as compared to language understanding is a well-known phenomenon. Additionally, sometimes the recognition does not have to be understood correctly because the context helps the child understand the meaning of an utterance. For expressive vocabulary, this is not possible in the same way. It is in many senses a harder and more complicated task to put a name to a picture correctly or to express exactly what you want to say. This requires that both lexical-semantic and phonological representations are emerging or established, having knowledge of the concept as well as efficient word retrieval ability (Storms et al., 2004). Additionally, the child preferable has to be able to mobilize correct articulation, in a short period of time, and cannot guess and use exclusion strategies like when pointing on a picture in a closed-set setting such as a test of receptive vocabulary. Picture naming can also be called word finding (Messer and Dockrell, 2006), which includes both knowledge and processing of lexical-semantic units. It is normal for all persons to occasionally have difficulties of finding the right word – and this is more common in children (Messer and Dockrell, 2006). Furthermore, children who are in a developmental phase might not yet have all the words they need to explain what they want to say. Word finding problems in typically developed and NH children can often be explained by factors like age of word acquisition, word frequency and lexical neighbourhood (Newman and German, 2005). Lexical neighbourhood means e.g. words that only differ by one segment like “sea” and “she”. Better word learning ability and as a consequence larger vocabularies in NH children has been associated with higher level of dense word learning, like the ability to perceive and learn words that sounds alike, like she and sea. Slow learning in children who are late talkers resulting in smaller vocabularies has been

explained by their different way of extract lexical meaning of words (Stokes, 2010). One sign of a possible late talker is a slow or late onset of expressive vocabulary (Stokes, 2010). Kavé, Knafo & Gilboa (2010) have studied word retrieval in a lifetime perspective. They found that difficulties with word-retrieval in NH and typically developed children for most part can be explained by a less developed and thereby smaller mental lexicon, while the reason for adults instead is less efficient cognitive capacity, that is the ability of retrieving words from the LTM becomes less efficient.

2.2.4 Depth in lexical-semantic ability

Depth of vocabulary refers to how well a child knows a word (McGregor et al., 2013). With a well-functioning depth of vocabulary a child can easier understand and find an exact definition and thereby also be more flexible and nuanced while communicating with others (McGregor et al., 2013). Another aspect of depth is the associations and connections in between words. Many connections between words and well-defined lexical-semantic representations influence positively on development of depth (Schoonen and Verhallen, 2008).

Depth in lexical-semantic ability can be measured by using different methods, including a semantic feature knowledge test, like the Repeat and point-test by Hodges, Martinos, Wollams, Patterson & Adlam (2008). Semantically developed test materials for children are sparse. One way to examine the depth in vocabulary is to use an error response analysis while conducting a picture-naming test. The responses give information of lexical-semantic organisation and especially associations in between words (Storms et al., 2004; Brusewitz and Tallberg, 2008).

Another way of studying the lexical-semantic network is to conduct word fluency tasks (Kavé, Kigel & Kochva, 2008; Sauzéon, Lestage, Raboutet, N'Kaoua & Claverie, 2003; Tallberg, Ichachova, Jones-Tinghag & Östberg, 2008). The individual is asked to generate words within a specific category during a period of 60 seconds. Semantic word fluency, often the category animal task, is especially thought to give information about the depth in semantic knowledge (Riva, Nichelli & Devoti, 2000). A phonemic-based word fluency task may give information both about vocabulary, size and executive functioning (Baldo, Shimamura, Delis, Kramer & Kaplan, 2001). One way of gaining semantic information from a word fluency task is to study the pattern of words that are retrieved, if they are clustered in any way, i.e. are connected, either semantically or phonemically (Schwartz, Baldo, Graves & Brugger, 2003).

Lexical-semantic creativity in typical child development

Children use different strategies to compensate for lexical gaps in their vocabulary. One common way is the incidence of over- and sometimes under-extension, such as using the word daddy for all men or using the word car only as a label for the family car not for cars in general (Jerger and Damian, 2005). According to Clark (1993), children do seem to create these generalizations of words based on their form, movement, size and way they sounds. Another common way of compensating for lexical gaps in vocabulary is to create new words when needed (Brusewitz and Tallberg, 2008). Swedish children often combine two separate lexical units and to some extent also add a morpheme to a lexical unit (Clark, 1993). Children use what they have and know in a creative way to

convey meaning. Other mentionable developing process in typical language development is the *U-shape pattern* (Stoel-Gammon, 2011). This means that a child might use the right word-form first like the word “gick” in Swedish (went) and then begins to produce the word wrongly “gådde” according to morphological rules and then again starts to use the right word-form again. This is an example of how the child learns language actively and starts to pay more attention to linguistic units and rules (Stoel-Gammon, 2011).

2.2.5 Lexical-semantic development in clinical groups

Some children have specific difficulties in developing, using and understanding spoken language compared with typically developed children. There are many different clinical groups that have such deficiencies, like children with developmental disorders (Vandereet, Maes, Lembrechts, & Zink, 2010), language impairment (LI) (Briscoe et al., 2001), pragmatic language impairment (PLI) (Ketelaars, Alphonsus Hermans, Cuperus, Jansonius & Verhouver, 2011), autism spectrum disorder (ASD) (Miniscalco, Hagberg, Kadesjö, Westerlund & Gillberg, 2007), Down’s syndrome (Berglund, Eriksson & Johansson, 2001), preterm children (Stolt, Korja, Matomäki, Lapinleimu, Hantaja, & Lehtonen, 2014), children with acquired brain injuries (Zetterquist & Jennische, 2010), blindness (Nejati and Asadi, 2010), dyslexia (Hulme and Snowling, 2013), Attention-Deficit Hyperactivity Disorder (ADHD) (Bruce, Thernlund & Nettelbladt, 2006), children with cleft lip and palate (Klintö, Salameh, Olsson, Flynn, Svensson, & Lohmander, 2014) and children with repeated otitis media (Winkel, 2006).

Additionally, groups like children who are adopted late or come from bilingual families with parents who do not speak the majority language have other language conditions in childhood than children who are born and raised monolingual (Delcenserie and Genesee, 2013) or simultaneously learning two or more languages (Masoura and Gathercole, 2005). These circumstances do not cause problems with language development in general, but could delay for instance vocabulary development (Hemsley, Holm & Dodd, 2013). Hemsley et al. (2013) found in their study of sequentially bilingual children that lexical representations of their second language (L2) was easier to acquire if their conceptual representation to the word already had been established in their first acquired language (L1). Words with greater conceptual distance between L1 and L2 were harder to acquire (Hemsley et al., 2013). These results indicate the importance of promoting that parents use their mother tongue in communication with their child (Salameh et al., 2004).

There are some theories that suggest that difficulties for children in clinical groups have domain-specific reasons and other theories propose that language disorders are explained by more general domain deficits as a result of an impairment in perception, like HI or language processing including memory (Gathercole, Pickering, Ambridge, & Wearing, 2004). Karmiloff-Smith (1998) represents the neuroconstructive approach, claiming that much of the variance of language development in typical and atypical cases can be explained by development itself, which therefore is the key of understanding developmental disorders. The neuroconstructivism theory focuses on

how the developing brain is linked to genes and brain growth in conjunction with environmental factors like language stimulation from caregivers.

Children with language impairment (LI) are a heterogeneous group (Sheng and McGregor, 2002; Bishop, 1997) just like children with CI (Peterson et al., 2010). This clinical group have specific difficulties in developing spoken language, despite NH and normal intelligence (Briscoe, Bishop & Norbury, 2007). The variation in language outcome is large but usually manifested as difficulties to process phonology and grammar as well as to improve vocabulary (Hansson and Nettelbladt, 2002). Typically, children with LI have word-finding problems and less developed semantic knowledge (Sheng and McGregor, 2010). Another heterogeneous group of children is the group of children with autism spectrum disorder (ASD) having known difficulties in the development of language, but for other reasons than children with LI (Bishop, 1997). Children with ASD often have deficits in understanding aspects related to cognitive aspects like ToM, resulting in less functioning socio-cognitive functioning and some also have a developmental disability (Fischer et al., 2005). Some children with ASD might have normal vocabulary sizes, but difficulties using their words in communication (Miniscalco, Rudling, Råstam, Gillberg & Johnles, 2014).

Children with mild-moderate HI also have an affected language development (Moeller, McCleary, Putman, Tyler, Krings, Hoover & Stelmachowicz, 2009). Even if they may catch up regarding language understanding, many may have persistent difficulties with grammar and phonology (Moeller et al., 2009). Lieu (2013) found that children with single-sided deafness had spoken language difficulties. Children with fluctuating hearing caused by otitis media are also at risk for having prolonged language delay (Winkel, 2006). One important difference between children with CI and children with mild-moderate HL, besides different hearing technologies, is that some children with HA not uses their devices full time while children with CI uses then to a higher degree (Anmyr et al., 2012). This may have a negative impact of spoken language results in the group of children with HA. Tomblin, Oleson, Ambrose, Walker & Moeller (2014) showed in their study of children with mild-moderate HL, fitted with HA, that it is not only important to fit HA early, but also to make sure that the HA provide an optimal level of audibility and to consider that also children with mild HI may have deficiencies in language development. In some cases there are children who have coexisting difficulties i.e. combined SLI and HI (Keilmann, Kluesener, Freude & Schramm, 2011).

2.3 CHILDREN WITH COCHLEAR IMPLANTS (CI)

Children with CI are heterogeneous in many aspects mainly due to age- and hearing-related factors but also to cognitive, linguistic and aetiological issues and environmental factors (Peterson et al., 2010; Geers et al., 2011; Spencer, 2004). Children with CI have experienced an initial period in life with auditory deprivation or a severe loss of hearing (Kral and Sharma, 2012) and after the CI implantation they continuously have different auditory experiences compared with normal hearing (NH) peers (Peterson et al., 2010). Auditory input with a CI is impoverished compared with natural acoustic hearing. A child with normal hearing (NH) has thousands of hair cells tuned to different frequencies, while only up to 22 electrodes from a CI stimulate the

auditory nerve for a child with CI (Houston & Bergeson, 2013). This means that listeners with a CI have more difficulties with frequency resolutions compared with NH individuals (Loizou, 2006) and also have a less dynamic range than for acoustic hearing (Zeng, 2004). This affects listening skills and spoken language development (Houston and Bergeson, 2013). Another challenge for children with CI is to hear spoken language in noisy environments, because they are at risk of missing out on salient speech features (Asp et al., 2012; Litovsky et al., 2012).

The variability of language outcomes is large within the population (Niparko, Tobey, Thal, Eisenberg, Wang, Quittner, & Fink, 2010). Longitudinal results in the group of children who were implanted fairly late and before the 20th century show difficulties in language and higher-level cognitive-linguistic functions like reading (Geers, 2010). However, recent longitudinal data show a different pattern in the “new generation” of children who were implanted after 2000, with a large number of children who have developed age-equivalent receptive vocabulary and language understanding (Karlton et al., submitted; Nicholas and Geers, 2013). During the relatively short period in history with cochlear implantation for children, since around 1987 worldwide and since 1990 in Sweden, several “generation shifts” have occurred within the small population. Some of these generation shifts can be explained by three major changes; (1) the event of new-born hearing screening leading to (2) earlier identification of the child’s HI in turn leading to (3) earlier ages CI implantation (Yoshinaga-Itano, Sedey, Coulter & Mehl, 1998). Furthermore, the technical development of the CI devices has been significant during the last decade (Geers, 2006). In addition, nowadays most children in Sweden have bilateral CI, which have proved to have a positive influence on aspects like listening in noise and sound localisation (Asp et al., 2012) and language understanding (Boons et al., 2012).

However, a CI surgery in isolation is not enough for a child with severe-profound HI to develop spoken language abilities. CI intervention also includes early intervention (EI) for language development (Joint committee of infant hearing, 2013). The CI operation should be accompanied with listening and language stimulation (Yoshinaga-Itano, 2013) preferably from caregivers who interact with the child in meaningful conversations (Quittner et al., 2013). Besides the changes with earlier ages at implantation, the educational situation has also changed, especially in Scandinavia, with more focus on auditory-verbal (AV) approaches than previously (Percy-Smith et al., 2012). Still, besides these changes over the years including implementation of an AV-based EI approach, there are subgroups within the population that do not seem to reach their optimal potential. One reason for that seems to be environmental factors like socioeconomic status (SES) (Ching, Geers, Szagun and Stumper, 2012). Furthermore, there are children within the population who have additional disabilities requiring special care and even more tailor-made intervention approaches, often with a focus on other more visual-based communication modes, rather than spoken language.

Because of the great heterogeneity in the small population of children with CI it is necessary to describe some of the factors that might have an impact and influence on lexical-semantic ability in the sample of children with CI. Focus will be on the Swedish situation and the factors are related to age, hearing and aetiology and also environmental aspects.

2.3.1 Hearing impairment (HI)

Children with CI do not have restored hearing with CI, due to the fact that they were born deaf or with a severe-profound HI or acquired deafness at an early age. Hearing impairment is a very common sensory deficit that affects around 360 million people around the world (WHO, 2014). Approximately 32 million of them are children aged 0-14 years. Around 0.5-5 of every 1,000 infants worldwide is born with or develops HI in early childhood (WHO, 2014). The majority of children with HI live in developing countries. In Sweden around 3 of every 1,000 newborn infants are identified with a permanent HI.

The HI can affect one or both ears and causes both communicative and social difficulties. For a child who is in the phase of developing language, any kind and degree of HI has a negative impact on the spoken language acquisition (Joint committee on infant hearing, 2013, Moeller et al., 2009). Many children with HI worldwide would benefit from using HA. However, current production of HA meets less than 10 % of the global need (for both adults and children) according to WHO (2014). For some children with severe-profound HI, the HA are not enough to facilitate a spoken language development through listening (Alford, Arnos, Fox, Lin, Palmer, Pandya, Rehm, Robin, Scott, & Yoshinaga-Itano, 2014). Instead they would benefit from a CI intervention or sign language approach or a combination of both alternatives (Joint committee on infant hearing, 2013). The focus in the thesis is to describe the outcome and characteristics of children with severe-profound HI with CI, as they are the ones who represent the sample in the empirical studies.

The degree of HI is measured as the best ear pure-tone average (PTA) across 500, 1,000, 2,000 and 4,000 Hertz (Hz) according to the guidelines of WHO (2014). The HI is measured by which frequencies a person hears and at what hearing level (HL) measured in decibel (dB) speech sounds are perceived. A HI can be referred as Mild: 26-40 dB HL; Moderate: 41-60 dB HL; Severe: 61-80 dB HL or Profound: >81 dB HL (Alford et al., 2014). Hearing impairment is also classified as either congenital or acquired, depending on when the HI occurs in life (Alford et al., 2014). In Sweden there are around 60 children every year with congenital or pre-lingual acquired severe-profound HI who are in question for CI intervention. Today (May 2014) there are nationally around 900 children who have received one or two CI, since 1990.

2.3.2 Audiological management

The best possible spoken language development requires individualized and high fidelity early intervention programs for children who are severe-profoundly HI and their families (Yoshinaga-Itano, 2013). Therefore, it is crucial with an early identification and diagnosis of HI in infants and toddlers, preferably before 3 months of age (Morton and Nance, 2006; Joint Committee on Infant Hearing et al., 2013). This process should in turn lead to the prompt fitting and use of appropriate state-of-the-art hearing technology, as soon as possible after the initial audiological assessment, and no later than after 6 months of age (Joint Committee on Infant Hearing et al., 2013).

Identification of hearing impairment (HI)

Today, it is possible to screen for HI at the time when babies are born at the maternity hospital (Morton and Nance, 2006). The universal newborn hearing screening (UNHS) has been established in Sweden since 2006, but this is still not the case in the whole world. The UNHS makes it possible to identify children with severe-profound HI after a couple of weeks. Still, there are some Swedish children who are not identified early but that come later for their first audiological assessment. In some cases it is explained by that they are immigrants from countries without UNHS. There are also some children who have better hearing when they are born, and therefore pass the initial screening test or who have a mild HI that progress and who later develop a severe-profound HI (Alford et al, 2014).

Initial hearing aid (HA) fitting period

Cochlear implantation often starts with a HA trial. Initial HA fitting period is important as it enable children who will later have a CI to start developing auditory pathways but also for audiological purposes, to investigate functional hearing (Kral and Sharma, 2012). When a baby with HI starts to use HA it might be challenging for the caregivers to support full-time listening with HA, as they might not see any signs of auditory responses in child with HA. At this point, it is especially important for caregivers to receive guidance and support from professionals who knows how and why it is important with full-time HA use (Moeller et al., 2009). The period with HA has been getting shorter for babies in Sweden, as the ages at 1st CI implantation have decreased (Karlton et al., submitted). However, there will always be some children who start with a longer period of HA use before CI intervention (Dettman et al., 2004). For children with additional needs, for example due to mental retardation, it could even be wise to prolong the period of HA trial and wait before deciding for or against a CI, because of delayed maturation of critical auditory areas in the brain. For children who are deaf due to meningitis, it is on the contrary important not to wait, but to implant immediately because of the risk for ossification of the cochlea. This can have a negative influence in surgery and the insertion of the electrode and later language outcome.

CI intervention

A CI is a hearing device that consists of external parts, a microphone with a speech processor and a transmitter, which are connected to the implanted part, which in turn contains a receiver connected to an electrode. The electrode is inserted into the cochlea and transmits electrical impulses through the cochlea and auditory nerve, which are perceived as sounds in the auditory cortex of the brain (Yoon, 2011). If HA is not enough amplification for developing spoken language through acoustic hearing, a CI is an option for children with severe-profound HI (Nikolopoulos and Vlastrarakos, 2010). It is not a cure for deafness, but in normal circumstances the electrical stimulation with CI leads to cognitive and linguistic learning (Nikolopoulos and Vlastrarakos 2010). The formal criteria for receiving a CI have changed during the years, since the first child was implanted. Initially only adults could have CI, but then children with acquired HI and later on also children with congenital HI were included as presumptive candidates. Initially, children with additional disabilities ASD were excluded as CI candidates (Nikolopoulos and Vlastrarakos, 2010). Today, all individuals who have a functioning auditory nerve and who hear worse than 70-80 dB HL bilaterally without HA, on an

average level of 250, 1,000, 2,000 and 4,000 Hz, are potential candidates. Another criteria are that the hearing threshold with HA in sound field is more than 50 dB HL for 2 and 4 kHz. The technical development in the area has been tremendous during the last decade and is one of the main keys, together with earlier age at implantation, to having more children who achieve age-equivalent levels of spoken language.

What does CI intervention mean?

A CI intervention is not merely an operation or two, but includes preparations and intervention post-implantation from a multidisciplinary team. The most important participants in the teamwork are the parents. Professionals working at day-care centres, preschools, schools where children spend a lot of their time and other family members like grandparents are also important persons in the intervention process. Before the CI operation a thorough investigation is done, primarily for investigating the hearing ability but also preverbal communication and general status of health. A computer tomography (CT) scan and/or magnetic resonance imaging (MRI) is conducted pre-implantation to check that the child has a functioning auditory nerve and for possible complicating malformations in the cochlea that might influence the surgery and potentially later outcome with CI. After the CI surgery, the actual auditory and oral stimulation process follows, which includes different kind of intervention from several professionals in close partnership with the child's caregivers, primarily at the local habilitation centre.

The interdisciplinary CIC team works in parallel and jointly with the local team to support the families and, more specifically, to facilitate the child's adaption of the CI. The CIC team at Karolinska University Hospital consists of administrative staff including a coordinator, SLPs, audiologists, engineers, social worker and medical doctors. If a child is implanted with a CI, the role of the SLP is to guide and support families in the learning process of supporting listening skills and spoken language of the child and to cooperate in different ways with the home-based SLPs and/or teachers of the deaf. The contact with the child and family is especially intense during the first year post-implantation. After the one-year follow-up visit, the child with CI returns to the CIC every half-year until she/he has had the implant for four years. Then the family comes back once a year for regular follow-up visits. Children are followed longitudinally from the time of surgery until they are around 18 years in this way and after that they are followed at a sparser interval, but regularly throughout life.

Influence of age at 1st CI implantation

There is now clear evidence children with HI who comply with the criteria for CI benefit from early implantation (Colletti, Mandalá, Zoccante, Shannon & Colletti, 2011; Nicholas and Geers, 2013; Wie, 2010). The rapid development of younger ages of 1st implantation has changed the landscape of possibilities for deaf infants and children with pre-lingual severe-profound HI. Most studies imply that children should be implanted before 24 months of age to gain optimal language development (Hayes, Geers, Treiman and Moog, 2009; Tobey et al., 2013). Furthermore, some recent studies show that there are significant differences between children implanted around 12 months and at around nine months or younger (Nicholas and Geers, 2013; Karltorp et al., submitted; Leigh, Dettman, Dowell & Briggs, 2013). There is a possibility of a certain bias, also discussed by Nicholas and Geers, 2013. Many of the children that are

operated on early may belong to a particular group of children with well-informed parents with higher SES, who seek the best possible opportunities both regarding earlier CI intervention and EI options. This makes these groups non-representative for the whole population of children with CI but still, the results shows the potential of all children with HI who need CI to reach better outcome with earlier ages at implantation. In addition to the discussion of age at implantation, recent longitudinal studies have shown that age at 1st implantation cannot solely explain a positive outcome (Szagun and Stumper, 2012) and that some children who are implanted at later ages can catch up over time (Tobey et al., 2013). However, the variation in outcome is large and the mean scores of general language ability for children implanted after 2.5 years were significantly lower than for children implanted at earlier ages (Tobey et al., 2013). Tobey and colleagues (2013) suggest that it would be beneficial to study individual differences within both of the age groups as one way to try to understand why some younger implanted children fail to acquire age-equivalent spoken language skills and why some older-implanted children adopt trajectories that can diminish the gaps.

Unilateral, bilateral or bimodal hearing

So, if early cochlear implantation is a good and reliable option in compensating for insufficient hearing, why are not all children with bilateral severe-profound HI given bilateral, simultaneous implants? Two implants would hypothetically be a better option than unilateral CI, as NH individuals hear with two ears. Recent studies also show a clear benefit of bilateral listening for developing spoken language (Sarant, Harris, Bennet & Bant, 2014; Niparko et al., 2010). Hughes and Galvin (2013) report that for some children listening effort seem to be reduced. They conclude that this might affect the attention level and therefore will be positive for learning processes. However, there are some hearing, ethical and medical-technical issues that control the decision for providing one or two CI to a child. Children with deafness caused by meningitis may experience ossification of their cochlea. Therefore it is important to operate as soon as possible, otherwise it could affect their possibility of having an optimal bilateral CI use. Secondly, provided that bilateral implantation is found to be the best option by the investigating CIC-team, it is still always the decision of the caregivers to decide whether their child should have an additional CI or not. Today, the majority of the youngest generation of children with CI, who are implanted after the year of 2010, have bilateral CI in Sweden. Most of them have been implanted sequentially for medical reasons, as their balance might be affected as a consequence of simultaneous implantation (Karlton et al., submitted). However, in a study by Boons et al. (2012) it was found that expressive language development benefit of simultaneous rather than sequential implantation.

Nowadays, a so-called bimodal approach is also fairly common with a group of children who have one CI and one HA, mainly due to the changed operation criteria of CI candidates and the fact that they have enough hearing on one ear with HA (Dettman, D'Costa, Dowell, Winton, Hill & Williams, 2004). This gives them an opportunity of developing spoken language with both acoustic and electric hearing. Some of the children in this group with bimodal hearing might be bilateral CI candidates later in life, if their hearing loss progresses. In many cases, within this group, the CI “takes over” and the child starts to rely primarily only on their CI hearing and has difficulties in continued use of the HA, even if the speech recognition data

might indicate that the HA should be beneficial for them, at least to a certain degree. In these cases, children are also probably candidates for bilateral CI. Today, there are some research studies showing that one should not wait too long if children are in the “grey zone” for being candidates of bilateral CI (Dettman, et al, 2004).

Aetiology

Cause of deafness can be genetic or acquired and is often connected to other issues besides the HI, which are of importance for outcome results and clinical management (Joint committee of infant hearing, 2013, Alford et al., 2014). Around 50 % of sensorineural hearing loss (HL) cases are caused by genetic reasons, of them 30 % are syndromatic and 70 % are non-syndromatic (Alford et al., 2014). The other 50 % are caused by environmental factors like infections (Alford et al., 2014). The phenotype of the different causes is heterogeneous in its nature and may affect the linguistic and cognitive outcome with CI.

One of the most common non-syndromatic genetic causes is Connexin 26 (cx26). Children who are deaf due to cx26 or other non-syndromatic and genetic reasons seldom have additional disabilities besides their HI, at least that are related to the cause of deafness (Genetic Evaluation of Congenital Hearing Loss Expert Panel, 2002). There are several different syndromatic causes of sensorineural HI like Usher’s syndrome, Jervell and Lange-Nielsen syndrome (JLNS) and Waardenburg syndrome. There are also many children with presumed hereditary causes of HI not diagnosed in detail. These children might have siblings who also have a HI. A major cause of acquired and non-hereditary congenital deafness is cytomegalovirus (CMV) resulting in a variation of different deficits besides HI. In a recent study by Karltorp et al. (submitted), it was found that a majority of children with cCMV-caused HI had late onset of walking, affected balance ability, vision problems and a higher incident of additional diagnoses like ASD and ADHD. Since the event of general vaccination 2009 in Sweden there are less Swedish children who become HI due to meningitis. As for children in the rest of the world, especially in the developing countries, the incidence of infection-acquired HI with accompanying, sometimes severe additional deficits is still a major problem (WHO, 2014).

For children in the present thesis, there was a large group with an “unknown” reason for their deafness. This is no exception, but a general reality, as it is still difficult and expensive to find out the exact cause of deafness and sometimes it is also difficult to determine if the cause is genetic or acquired or if the two factors coexist and cause the HI (Alford et al., 2014). Around 30-40 % of all children with CI have additional disabilities that may affect their outcome (Ching, Dillon & Day, 2009). Eze, Ofo, Jiang and Fitzgerald O’Connor (2013) examined results for children with CI and additional disabilities in a review article. They concluded that children with developmental disabilities might not benefit from CI, based on traditional assessment tools but appeared to improve their quality of life and environmental awareness. Eze et al. (2013) also concluded that there was a need of more work on how to define the term benefit in these subgroups.

Speech perception and cognitive processes

Traditionally, auditory outcome with CI has been measured with e.g. speech recognition tests examining levels of hearing non-verbal sounds and speech at different frequencies and with different amplitudes. Additionally, when children started to get bilateral CI, the level of localisation and listening ability in ambient noise have been included as measures of hearing ability in children with CI (Litovsky et al. 2012; Asp et al., 2012). A pure-tone audiometry test result gives us good diagnostic information about the child's threshold hearing in different frequencies, but does not alone predict how a child is actually functioning in a real life situation regarding listening (Wie, Falkenberg, Tvette and Tomblin, 2007). Wie et al. (2007) found that use of CI predicted speech recognition the most and that non-verbal cognitive ability also was correlated to better speech recognition level. In addition, mode of communication and choice of educational setting, in favour of spoken language and mainstream options, explained better results of speech recognition, in line with other studies (Archbold, Nikopoulos, Tait, O'Donoghue & Lutman, 2000; Geers, Brenner & Davidson, 2003). However, recent data shows that measures of speech perception results are not always associated with outcome and do not explain the variation of higher linguistic functions, like lexical-semantic ability, in the new generation of children who are implanted at fairly low ages (Karltr p et al., submitted). In a recent study by Caldwell and Nittrouer (2013) children with CI had more difficulties in recognizing speech on group level, both in quiet and in noise, and in comparison to NH children. They conclude that it is important to maximize signal-to-noise levels in the classroom and that all children with CI need extra support to acquire optimal language and phonological skills.

More research points in the direction of cognitive deficits due to the initial auditory deprivation that might be causing some of the variation in i.e. phonological awareness (Cupples, Ching, Crowe, Day & Seeto, 2013) or executive functioning (Kronenberger, Pisoni, Henning & Colson, 2013). Finding instruments that not only capture speech perception on peripheral level but also measure higher-level functions that are more related to cognition and linguistic learning would therefore be beneficial. There is clear evidence that PWM aspects are involved in the listening process and affect listening and spoken language development, in children with HI, including children with CI, but also in NH children and other clinical groups (Lyxell et al., 2009; Masaroula and Gathercole, 2005). In the case of children with CI, the focus of research has been the immediate PWM process leading to worse storage, while less interest has been shown for the level of knowledge in the mental lexicon (Jerger, Damian, Tye-Murray, Dougherty, Mehta & Spence, 2006).

In a recent study by Nittrouer, Caldwell-Tarr and Lowenstein (2013), the authors argue that the problem children with CI have is more specifically in the storage of spoken language, not in the processing mechanisms, except for receiving the signals in an impoverished way. They conducted an experiment where both parts (process and storage) were examined at the same time, by using word lists designed to manipulate storage and processing skills independently. The results showed that children with CI recalled equally many items as children with NH, but showed less accurate recall of serial order than children with NH. Their results did not support the conclusion that children with CI have atypical development of working memory capacities. Nittrouer

and colleagues (2013) concluded that children with CI instead have poorer storage capacity but normally functioning processing. This result is also in line with results for adults with acquired HI, showing the same kind of pattern for episodic memory (Rönnberg et al., 2011).

Considering that children with CI or HA make up a very heterogeneous population, it is important to measure auditory outcome on both peripheral and central levels for technical, listening, cognitive and spoken language developmental reasons (Fitzpatrick;). Just like it is beneficial to have a broad test battery for examining spoken linguistic development, it is also an advantage to have a broad set of assessment tools for examining auditory and higher functions of listening that border on more cognition-related skills (Geers, 2009). Assessing listening in ambient noise is especially important, as this is one way of trying to examine how the child hears in common noisy everyday situations like the school canteen (Asp et al., 2012; Litovsky et al. 2012). This measure will also give information of cognitive functions, like attention level, which are part of the higher-level executive function system. Besides assessing the hearing and listening ability in formal tests, it is also possible and useful to perform parent and/or child evaluation forms especially designed to describe the listening (Ching, Day, Seeto, Dillon, Marnane & Street, 2014).

2.3.3 Characteristics of environmental factors

Influence of spoken language stimulation from caregivers

Hearing-impaired children who receive CI learn spoken language partly under different conditions than NH children (Petersen et al., 2010; Karl and Sharma, 2012). At the time they are implanted, they have already missed a large part of the language experiences and verbal interactions that form the basis of spoken language development in NH children (Bloom, 2002; Kuhl, 2010; Lacerda, 2005). Therefore, a targeted and individually based support is desirable for children with CI and their families (Dettman et al., 2007; Grant et al., 2006). In a study by Desjardin and Eisenberg (2007) it was found that mothers' level of participation and confidence in communication with their children with CI influenced positive on their quantitative and qualitative linguistic stimulation and the children's spoken language development. Szagun and Stumper (2012) found that linguistic development is largely explained by mothers' linguistic input and education level. Parents' level of education has previously been shown to be an important factor in NH children's language development (Hart and Risley, 1995). A study by Pressman et al. (1999) showed a significant association between expressive spoken language ability in children with hearing loss and parents who were attentive to their child's own initiatives and involvement in communication.

Choice of communication mode. Most Swedish children with CI, implanted during the last 10-15 years, uses spoken language as their primarily communication mode both in interaction with peers and family and for knowledge-based learning (Karlörp et al., submitted). Some of them also develop a sign-based language using either supported signs or sign language. Others develop two or more spoken languages in parallel with or without a supporting sign-based system. Ninety-five percent of the parents who have a HI child are NH themselves and have no prior knowledge about deafness, hearing impairment, experience of sign language or consequences of having a child with special

needs. Today, when a majority of children with severe-profound deafness are identified early in life, most parents choose spoken language as their first and primarily communication option (Karlton et al., submitted; Percy-Smith et al., 2012).

Family intervention options

Today, most intervention options in Sweden are focused on individualised EI by using either an auditory-oral (AO) or AV approach. Initially, there were few SLPs working with individual AV-based intervention for families with HI children in Sweden. Today, there are several SLPs and teachers of the deaf who work with an individual family-centred AV approach, for families with children who have HA or CI. Auditory verbal practice focuses on family empowerment and individualized coaching of how caregivers can stimulate spoken language development primarily through the listening of their children with CI or HA, aged 0-3 years (Percy-Smith et al., 2012; Estabrooks, 2013). The AV approach has also been accompanied by other more auditory-oral related options in Sweden, for families who need and choose inclusion of sign-supported communication. However, the situation in the whole country still varies regarding possibilities for individual families who want to choose e.g. an AV-based approach.

Preschool and school options

Many Swedish children with CI who have undergone a CI intervention since the beginning of 20th century are today integrated in mainstream settings or part of classes in special schools for hearing impaired. The situation for children who are integrated in mainstream preschools and schools with age-matched NH children varies, with more or less individual and general support. There are sometimes a reduced number of pupils in the class or a resource person in the classroom dedicated to support the child with CI, as well as the use of FM-systems. For children in special classes for hearing-impaired there is an adjusted environment, with smaller classes, FM-systems, sign-supported communication for those who need it and sometimes the possibility of individual speech training. However, the spoken language support varies and is not uniform, in either of the options. In Sweden, fewer children with CI attend schools for the deaf with sign language as the main education language, except for those that have deaf parents. Other cases include that they were diagnosed late and as a consequence had higher ages at their CI operation or have an additional diagnosis besides their HI influencing their ability to learn spoken language primarily through listening and thereby to be integrated in a mainstream environment.

2.4 LANGUAGE DEVELOPMENT WITH CI

Early developmental phase (0-24 months): due to an initial period in life with auditory deprivation, deaf-born children will not be able to recognize their mother's voice or songs that have been sung to them before birth like NH infants (Granier-Deferre et al., 2011). The severity of consequences varies depending on the length of this deprivation period and the severity of their HI (Schramm, Bohnert & Keilmann, 2010). They might still be able to vocalize like NH babies initially in life but have a later onset of canonical babbling (Moeller et al., 2007a). Besides the necessity of wearing their hearing devices full-time, infants and young children need support in learning to listen and e.g. to be aware of how to use their voice with intent (Yoshinaga-Itano, 2014).

There is evidence that deficits within speech perception and speech production have a negative influence on both the immediate language development and on later and higher cognitive abilities like reading, at school age in children with HI with either CI or HA (Geers, 2010). The good news is that the plasticity of the brain is at its greatest during the first 3 years in life (Kral and Sharma, 2012). This means that even congenitally deaf-born children can catch up and develop listening skills and spoken language comparable to those of NH children, if they are provided with early and best possible hearing amplification in conjunction with linguistic experiences with supporting caregivers (Quittner et al., 2013). Recent studies indicate difference in outcome if the child is implanted before or after 12 months (Nicholas and Geers, 2013; Karltorp et al., submitted). Few studies have examined babbling in children with severe-profound HI. However, Schramm et al. (2010) found that children who were CI-implanted at early ages developed canonical babbling in a faster rate than NH children, and acquired consonant phonemes in similar ways like NH children. A recent study by Sundquist et al. (2014) reports that children with early ages at 1st CI have age-equivalent ToM, a factor important for vocabulary development (Bloom, 2002). In addition, Bergeson, Houston & Miyamoto showed that early auditory stimulation is important for integration of audiovisual information, indicating that auditory deprivation affects multimodal perceptual processes. Furthermore, children with HI usually have a delayed onset of first word unless they are fitted early with HA or CI and this influences on later vocabulary outcome (Houston, 2012).

To summarize, children who are implanted early with CI can develop listening skills, canonical babbling, and ToM in a more synchronized way. These abilities are important for early word learning, in turn predicting of later vocabulary outcome.

One way of trying to explain how key language skills are interrelated has been proposed by Bloom and Lahley (1998). They divide language skills in three modules: use, form and content. However, they have not distinguished between expressive and receptive language and they have not included speech perception and aspects like attention and memory. One attempt at including some of these aspects in the model by Bloom and Lahley (1998) has been made below, while describing the overall findings of language results so far within the population with CI. The main purpose of this was to try to distinguish between domains and at the same time show the interrelationship of the linguistic and more cognitive related skills.

Use could primarily be described as the way language is used in communication with others, but also as an individual's internal cognitive processes for solving problems, planning and reasoning. Basically, use could be summarized as the way language is used for social and personal needs. Use can also be referred to as pragmatics. Semantics and pragmatics are in many senses interrelated, but are not examined specifically in this thesis. Language use is related to aspects like ToM (Bloom and Markson, 1998; Fischer et al. 2005). Children with HI might have difficulties to perceive prosodic information in spoken utterances like irony, jokes, and subtle emotional features that are easy to perceive if you are NH and have a typical cognitive development. In a recent study by Goberis, Beams, Dalpes, Abrisch, Baca, & Yoshinaga-Itano (2012) it was shown that children with HI had age-equivalent language scores on tests of understanding and vocabulary but the same children were many years behind in their pragmatic skills. One

explanation, besides less efficient auditory skills, could be that they do not have as many linguistic experiences or have not been naturally trained to interpret auditory linguistic-related social cues to the same extent as NH peers of the same age. Children with parents who have been sensitive to their young child's own communicative initiatives and who have been more interactive in their communication with their child have better long-term outcome (Quittner et al., 2012; Cruz et al. 2013). Maternal sensitivity has been found to be one important factor in promoting natural and well-function communication and language skills in children with HI (Pressman, Pipp-Siegel, Yoshinaga-Itano, & Deas, 1999).

Form refers to speech sounds; phonology, parts of words; morphology, combination of words in phrases and sentences; syntax are organized following universal and more language specific "rules" (Bishop, 1997). Today, most children with CI develop intelligible speech and in a shorter period of time than in the early days of CI intervention (Boons et al., 2012). This is the result of earlier CI intervention (Leigh et al., 2013; Wie, 2010). The infants who are implanted within their first year of life are more in synchrony with their biological age and many of them therefore catch up in speech quickly and easily, compared with children with CI in previous years (Leigh et al., 2013; Kral and Sharma, 2012). However, some children are still at risk of lacking linguistic experiences (Szagun and Stumper, 2012) and some studies have shown that children with CI may have phonological deficits affecting later phonological, grammatical skills (Wass et al., 2008). Some of these studies concern children who were older at the 1st CI and most of the previous studies were made with unilateral cohorts (Willstedt-Svensson et al., 2004). Still, recent studies show similar patterns for individuals in the new generation of children with younger ages at 1st implantation and bilateral CI. Interestingly, despite their lower ability to develop age-equivalent phonological skills, they develop age-appropriate literacy over time, indicating that they compensate for worse phonologically based higher-level abilities with other orthographic learning strategies (Geers and Hayes, 2011).

Even if children with CI are able to catch up in some linguistic and cognitive areas, studies show that they are still behind on grammar (Duchesne et al., 2010; Ramirez Inscio, Odell, Archbold & Nikolopoulos, 2009). It seems that this domain is more difficult to grasp for children with CI. One reason for this might be their difficulties to perceive low-ambient and unstressed linguistic markers like endings and prepositions in fast and acoustic speech stream (Svirsky, Stallings, lento, Ying & Leonard, 2002; Tribushinima, Gillis & De Maeyer, 2013). On a syntactic level, there are nowadays fewer children with syntactical errors, previously found in previous generations of children with CI. Duchesne et al. (2010) found in a study of grammar that not all children implanted between 1-2 years of age were able to reach the same grammar levels as NH children, even after 6 years of CI-use. In addition they identified small phenotypes within their sample of 27 children.

Content could be described as the meaning of words and groups of words and the network between words and concepts. In this thesis, this is referred to as lexical and semantic ability on word level. Language understanding is one of the skills that develop to an age-equivalent level for a majority of children with CI (Geers et al., 2009; Colletti et al., 2011; Karltorp et al., submitted).

Depth and breadth in lexical-semantic ability of children with CI

In the early stages when CI-intervention was implemented for children, there was little interest in examine more linguistic specific matters, with a few exceptions (Szagun, 2000; Szagun, 2001). Since, then there has been a period when much research focus has been on examining the influence of earlier ages at implantation. Researchers have used clinical tests for evaluating success of CI by determine e.g. their receptive vocabulary in comparison with norm data. Only lately has the research become more focused on cognition and developmental processes rather than test scores (Wass et al., 2008; Tobey et al. 2012; Houston et al., 2012).

There has been little previous interest in studying e.g. word learning ability in children with HI and CI (Houston et al., 2012; Willstedt-Svensson et al., 2004). In a Swedish study by Willstedt-Svensson et al. (2004), word learning was studied in a small group and with much higher ages at implantation. However, this study has influenced on later studies as it is one of the first studies that examined word learning in children with CI and also because the findings not only found correlations with age at CI but also cognitive factors like PWM (Schwartz et al., 2013). Houston et al (2012) found that word learning ability of toddlers influenced on later vocabulary development.

Further, the diversity within the groups that have been studied is not equal regarding background characteristics, especially not chronological ages and ages at 1st implantation, which may be some of the most important factors for developing early vocabulary skills (Houston et al., 2012). However, there are also some studies that point in a new direction, where the effects of age at implantation levels disappear as a function of increased age and that other factors may affect a positive development of e.g. vocabulary in children with later ages at 1st CI (Szagun and Stumper, 2012). One important factor in this process is the level of quantitative and qualitative use of language stimulation from caregivers (Quittner et al., 2013; Nott et al., 2010) as well as the child's own inherent abilities, like intelligence (Geers, 2009). Children with additional disabilities besides their HI have more difficulties in developing vocabulary, regardless of age at 1st CI (Cupples et al., 2014).

Several studies have reported that children implanted before 2 years of age reached age-equivalent receptive vocabulary (Hayes et al., 2009; Percy-Smith et al., 2012; Fagan et al., 2010; Colletti et al., 2011). However, recent studies indicate that there are differences in outcome when children are implanted before 12 months (Leigh et al., 2013; Nicholas and Geers, 2013). In addition, Karltorp et al. (submitted) reported that there were some children who did catch up on receptive vocabulary, even if they were implanted somewhat later. This shows that there are other factors involved in the learning process that are more cognition-related and influence receptive vocabulary. In the same study, there was a group of younger implanted children who did not reach expected levels despite early age at 1st CI, on e.g. expressive vocabulary. Many of them came from homes where both parents spoke another mother tongue than Swedish, a factor that could have affected the results (Teschendorf, Janeschik, Bagus & Arweiler-Harbeck, 2011).

Aragon and Yoshinagana-Itanto (2012) showed in their study of bilingual children that it is not two languages per se that have a negative influence, but SES related factors, also found in other studies, on both NH and HI. Interestingly, the researchers were able to show a positive correlation with family-centred intervention and use of the language environmental analysis (LENA) technology (Aragon and Yoshinaga-Itano, 2012). Intervention with LENA has recently showed positive results in many studies, of both NH children and children with HI (Ambrose, Van Dam, & Moeller, 2014; Aragon & Yoshinaga-Itano, 2010; Leffel and Suskind, 2013). Dushesne, Sutton, & Bergeron (2012) examined receptive vocabulary and found that there seemed to be phenotypes within the sample. Expressive vocabulary was influenced by age at implantation and phonological awareness and these two factors were important for literacy, which in turn affected continuous development of receptive vocabulary (Dushesne et al., 2012). Expressive vocabulary had a somewhat different path although closely connected to receptive vocabulary. A supporting environment and early onset of expressive linguistic use seem specifically important (Quittner et al., 2013; Cruz et al., 2013; Szagun and Stumper, 2012). A recent study by Nicastra et al. (2014) indicate the possibility that some of the variation seen in children with CI concerning oral language and reading understanding, could be explained by poor ability of decoding higher-level semantic knowledge e.g. to understand and use figurative language.

In summary, the research field relating to semantic knowledge and lexical-semantic aspects has generally become more narrowed over the years as a consequence of better possibilities of early, bilateral implantation in new subgroups of children with CI. It is now possible to study how children with CI develop linguistic and cognitive skills more in synchrony with their chronological age. The change has also broadened the field as a consequence of our expanded knowledge about this heterogeneous population and subgroups. However, depth in lexical-semantic ability has not yet been the focus of research, with a few exceptions.

Spoken language ability in early school years (6-9 years): The variation in outcome is larger in children with CI at this age, than in NH children (Geers, 2009). Some children with CI might catch up with NH children at school entry on many aspects, despite continuously worse phonological abilities (Wass et al., 2008). Despite of this, many children learn to read and have age-appropriate reading abilities (Lyxell et al., 2009). Several studies report that children with CI in this age may have age-equivalent vocabulary, but less developed grammar (Dushesne et al., 2012). Others report that children with CI in early school ages have less developed vocabulary and delays in pragmatic skills (Tobey et al., 2013, Aragon and Yoshinaga, 2012).

3 THE PRESENT STUDY

3.1 AIMS OF THE THESIS

The general aim of this thesis was to explore and investigate lexical-semantic ability of children with CI aged 6-9 years in a prospective cohort study. Background characteristics like age at implantation (1st and 2nd) as well as level of non-verbal cognitive ability and other demographic factors were investigated in relation to lexical-semantic ability.

-The hypothesis was that children with CI would have a more shallow and narrow vocabulary than age-matched NH peers.

Study I: To examine word fluency performance in children with CI and in comparison with age-matched NH controls. Additionally, a second aim was to explore the use of strategies in the retrieval process of words from the long-term memory.

Study II: To explore lexical-semantic ability of children with CI and in comparison with age-matched NH controls and children with language impairment (LI) or autism spectrum disorder (ASD). Furthermore, a semantic response analysis of the erroneous responses of a picture-naming test was performed.

Study III: To examine lexical-semantic ability on sentence level in relation to cognitive and linguistic factors, including verbal learning in children with CI and in comparison to age-matched NH controls.

Study IV: To follow up and examine the influence of age at implantation on lexical-semantic development in a subgroup of children with CI. One hypothesis was that a subgroup of children with younger age at 1st CI would catch up on receptive vocabulary over time and thereby show a less atypical developmental pattern than another group of children with higher mean age at 1st CI.

3.2 METHODOLOGICAL ISSUES: POSSIBILITIES AND LIMITATIONS

Children with CI would have been deaf without a CI intervention. Still, without the outer part of CI they have no, or a severely reduced hearing (in the case they have bimodal listening). However, the electrical stimulation of the auditory nerve enables this group of children to develop listening and spoken language skills as well as literacy (Geers, 2009; Lyxell et al., 2009). Not in the same way as if they had been NH, but still in a way no one would have thought possible in the beginning of the 80's when the first children were implanted in Sweden. Considering the relatively short period of CI intervention, many things have happened and developed on individual, subgroup and population level, in the direction of more specific and also broader clinical educational and research perspective (Peterson et al., 2010). In addition, considering that auditory sensory periods are crucial for this population we can also learn more about the importance of these sensitive periods in general (Houston et al., 2012).

One major limitation in studying children with CI is that the group is small and heterogeneous in most aspects like age at identification, age at implantation, aetiology and SES and many other factors (Geers and Seedy, 2011; Boons et al., 2012). This is a challenge and limits the possibilities of large randomised studies or designing studies with hypothesis-driven testing. Furthermore, considering the limited size of the population in a small country like Sweden, there are high standards on ethical issues to avoid exposing the children. A second limitation is that our level of knowledge regarding their linguistic and cognitive development is relatively low, at least in comparison with other populations of children with disabilities that affect their linguistic and cognitive development (e.g. children with dyslexia; Melby-Lervåg et al., 2012).

Initially, research in the population was more medical-technical directed. The research in Sweden and Scandinavia has developed over the years and now incorporates aspects like aetiology (Karlton et al., submitted; Henricson, Wass, Lidestam, Möller, & Lyxell, 2012), social wellbeing (Anmyr, Olsson, Larsson & Freijd, 2012), bilateral outcome (Asp et al., 2012), cognitive aspects like phonological training (Nakeva von Mentzer et al., 2013), reading and writing (Lyxell et al., 2009) and ToM (Sundqvist et al., 2014) besides from listening and spoken language development (Wie et al., 2007; Wie, 2010; Karlton et al., submitted; Percy-Smith et al., 2012). However, less is known about development of vocabulary, especially of semantic knowledge in relation to cognitive capacity compared with age-matched groups. In addition, there is sparse knowledge about semantic knowledge in general in the whole research field, especially concerning lexical-semantic organisation, picture naming and semantic feature knowledge in children with HI, regardless of type of technology (Kenett et al., 2013; Jerger et al., 2013; Schwartz, Steinman, Ying, Mystal, Houston, 2013).

There has been a fast development in Scandinavia within the population of children with CI in all areas. Especially concerning the implementation and influence of earlier ages at 1st CI, bilateral implantation and more AV-based intervention options (Percy-Smith et al., 2012). Therefore, it is somewhat difficult to compare studies of language, such as lexical-semantic development, in children within different “generations”, meaning children that underwent CI intervention at different periods in time, since 1990. The mean age at implantation in the 1st generation of children implanted in

Sweden was around 3 years; today the youngest implanted child was 5 months and the group mean is around 12 months (Karlton et al., submitted). In addition, the CI-technique has developed and the general individual family-support has changed focus and as a consequence from sign language to a more spoken language based communication approach.

The selection criteria were set as an attempt to assess lexical-semantic ability in a group of children with CI, in the best conditions. Therefore, to rule out some of the known factors that can affect spoken language development a group of children within the population with additional disabilities that might affect their linguistic and cognitive development was not invited to take part in the studies. This does not mean that these children should not be included in future studies, but given the high degree of heterogeneity in the population of deaf children with CI we need to build up a solid base of theoretical knowledge for deaf children with CI – but without additional disabilities. When this is accomplished, we can also study children with additional disabilities and to relate their linguistic and cognitive performance to children without other disability than a severe-profound HI or deafness.

3.3 METHOD

3.3.1 Participants

Children with CI - Studies I-IV

The cohort of children with CI had all received their CI and follow-up treatment from the Cochlear Implant Centre (CIC), Karolinska University Hospital, Stockholm, Sweden. At the time of recruitment in the year of 2009, there were 104 children in the age range 6-9 years enrolled at the CIC. Besides the determined age range of around 6-9 years, there were additionally four inclusion criteria for participation in the study; (1) to be full-time users of their 1st CI, (2) to be Swedish-speaking, (3) to have had their 1st CI for at least two years and (4) not to have any identified cognitive disabilities like LI or ASD besides their deafness.

Among the 104 children, there were 24 children who came from families where Swedish was not the first language spoken at home. Eight children were diagnosed with additional disabilities. Eight children had moved and received follow-up services at other CICs. Two children had worn their CI for a shorter period than two years and four children were only part-time users of their CI. The remaining group, who met all the inclusion criteria for the study at the time of recruitment, included 58 children. They were all invited to participate in a letter of invitation. Thirty-six of them agreed to participate in the study. However, two children were later excluded because they had been diagnosed with LI at the time of the study. In the end, there were 34 children left (15 girls) in the study group, aged 7;7 years (5;9-9;11), see Figure 4.

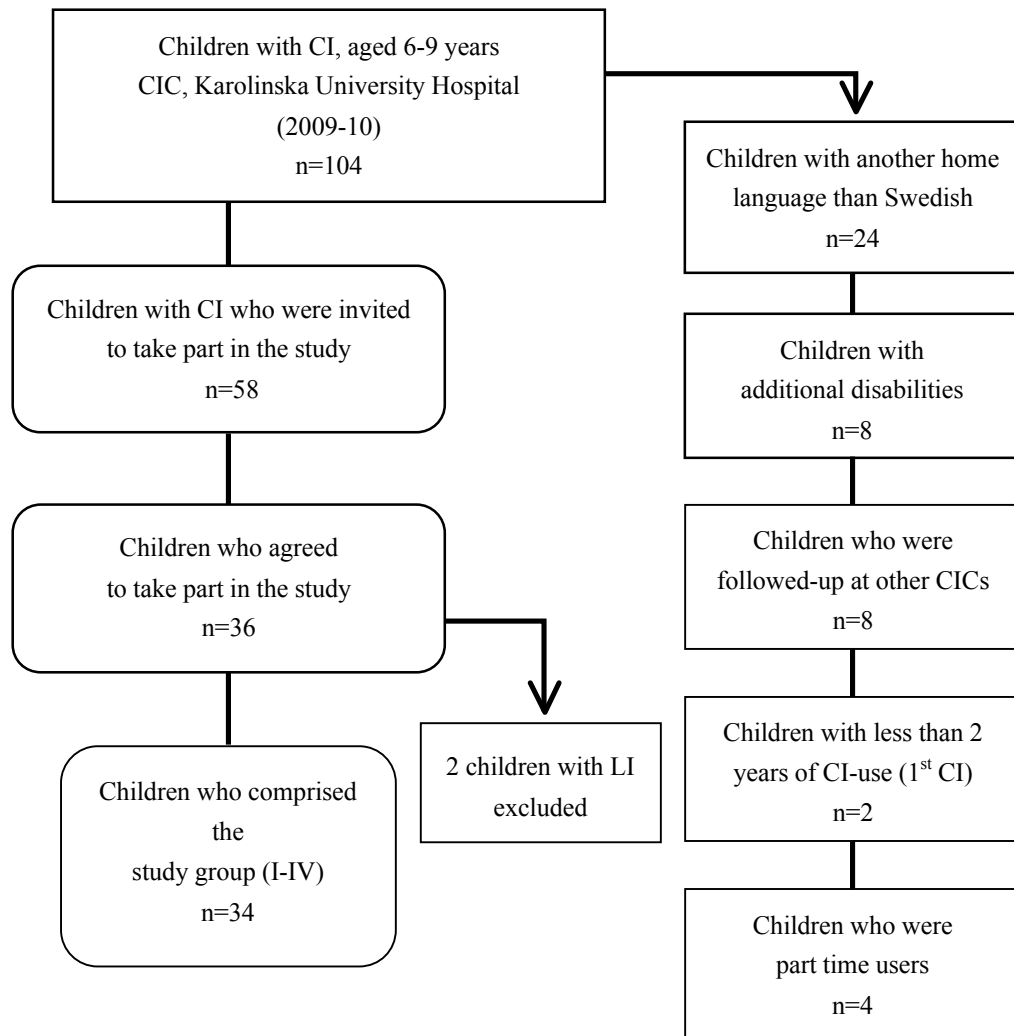


Figure 4. Flowchart representing the selection process of children with CI (Studies I-IV).

Hearing background

Eleven children were identified with deafness or severe hearing-impairment at birth through universal newborn hearing screening. Unaided PTA dB HL (pure-tone average threshold) at the time of diagnosis in the sound field at 500, 1,000, 2,000 and 4,000 Hz was measured pre-implantation on the best ear. The results for the study group were 116-130 dB HL for 17 children, 101-115 dB HL for nine children and 82-100 dB HL for eight children. Twenty-three children were fitted with HA before six months of age, one child had HA between 6 and 12 months and four children between 25 and 36 months of age. The five cases with meningitis and one late-diagnosed child did not receive HA prior to the CI intervention.

Aetiological background

Twelve children were deaf due to genetic causes: non-specific hereditary cause (5 children), Connexion 26 (cx26) (3 children), Usher's syndrome (2 children) Jervell and Lange-Nielsen syndrome (JLNS; 2 children) and another seven children had an unknown cause of deafness. There were eleven children who were deaf due to non-

genetic causes of deafness: meningitis (5 children) and congenital cytomegalovirus (cCMV) (6 children).

Cochlear implantation

Twenty-eight children had bilateral CI. Five had both CI implanted simultaneously. There were two children with unilateral CI and another four children had a bimodal hearing situation (CI+ HA). The two children with unilateral CI use were both deaf due to meningitis, one was implanted unilaterally due to medical reasons and the other child stopped using one of his devices due to technical and medical reasons. Thirty children in the study group had devices from Med-El and four children had devices from Cochlear Corp. The mean age at 1st CI implantation in the cohort (n=34) was 22±13 months (7-61) and 30±14 months (7-59) at the 2nd CI implantation (n=28). Eight children received implants before 12 months, nine children between 12 and 17 months, and six children between 18 and 23 months of age. Twenty-three of the children in the study group received CI intervention before their second birthday. Among the remaining participants, there were five children who received CI between 24 and 35 months and six children received CI between 36 and 61 months of age. The mean time period with the 1st CI (hearing age) was 69 ±18 months (28-99) (n=34) at the time of test occasion 1 (Studies I-IV). The eighteen children who took part in the follow-up testing in study IV had a hearing age of 104±8 months (90-119) at test occasion 2.

Demographic characteristics

Fifty percent of the children with CI came from the middle or northern part of Sweden and the remaining from the Stockholm area. All of the children used spoken Swedish as their main communication mode and had parents who spoke Swedish at home, confirmed by medical records and clinical observation. Twenty-seven children went to mainstream schools and seven children attended special classes for hard-of-hearing children in regular schools where spoken Swedish was the main language of education. None of the children attended special schools for deaf where sign language was the main language.

Characteristics of children with CI who declined to participate

There were 22 children who declined to take part of the study, 8 girls and 14 boys. This group had somewhat higher age at their 1st and 2nd implantations. The mean age at 1st CI implantation was 24 months (10-58) (n=22) and 35 months (18-59) (n=18) at 2nd CI implantation. Twenty children had Med-El devices and two children had devices from Cochlear Corp. Eighteen children had bilateral implants, two had unilateral CI and two children had a bimodal hearing situation (CI +HA). Two of the children were deaf due to meningitis, five due to hereditary reasons and fifteen due to unknown reasons. There was a difference in school setting between the study group and the children that declined to participate. Four of the 22 children went to special schools for deaf children with a mainly sign language approach, while there were none in the study group. Eight children attended special classes for hearing-impaired children and 10 children (45 %) were mainstreamed. In the study group the majority, 27 children (79 %) were mainstreamed. The reasons for declining to take part in the studies are unknown.

Table 1. Background characteristics for children with CI on individual level: aetiology, number of children detected with OAE, age at 1st and 2nd CI (months), time between 1st and 2nd CI (months) and number of children in mainstream school setting.

Child	Aetiology	OAE	Age CI-1	Age CI-2	Time C1-2	Mainstream
1	Hereditary		20	36	16	X
2	Unknown		20	54	34	X
3	Unknown		29	34	5	X
4	CMV		22	30	8	
5	Unknown		17	40	23	
6	Hereditary	X	11	26	15	X
7	Unknown	X	47	59	12	
8	Usher		17	30	13	X
9	CMV		15	27	12	X
10	Cx26	X	11	26	15	X
11	Unknown		14	18	4	X
12	JLNS	X	10	18	8	X
13	Hereditary		38	-	-	X
14	Unknown		14	23	9	X
15	JLNS	X	13	13	-	X
16	Unknown	X	24	24	-	X
17	Meningitis		16	16	0	X
18	Meningitis		17	17	0	X
19	Cx26		22	28	6	X
20	CMV		23	45	22	
21	Hereditary	X	11	47	36	
22	Unknown		42	-	-	X
23	Meningitis		27	-	-	X
24	Unknown	X	61	-	-	
25	CMV		48	48	0	
26	CMV		18	31	13	X
27	Meningitis		11	-	-	X
28	Unknown		30	-	-	X
29	Unknown		26	26	0	X
30	Usher	X	13	31	18	X
31	Cx26	X	9	22	13	X
32	Unknown	X	9	18	9	X
33	CMV		44	44	0	X
34	Meningitis		7	7	0	X
Total		11/34	22(13)	30(13)	11(10)	27/34

3.3.2 Comparison groups

Age-matched children with normal hearing (NH) - Studies I-IV

A control group of NH children (n=39) was recruited with the help of a school nurse, from two different schools in a small town. Only Swedish-speaking children with typical development and presumed NH were recruited from around 160 children aged 6-9 years.

Children with language impairment (LI) – Study II

Recruitment of children with LI was done together with an SLP at the Karolinska University Hospital and at special classes or schools for children with LI. All participating children had been investigated and diagnosed with specific LI of the general or expressive type according to the classification of diagnosis, ICD-10 (2000), by a SLP and a psychologist before the study. This was done by the team for neurodevelopmental disorders at Karolinska University Hospital. All children with LI had an IQ over 85. Half of these children were boys.

Children with autism spectrum disorder (ASD) – Study II

Only children with high functioning autism and Asperger syndrome were invited to participate. The participants with ASD were all investigated and diagnosed by a multidisciplinary team for neurodevelopmental disorders at Karolinska University Hospital and in accordance with the DSM-IV-R. The sample included a high proportion of boys and this is also common in clinical practice (Bishop, 1997).

Table 2. Background characteristics on group level; CI (n=34), NH (n=39), LI (n=12), ASD (n=12) for gender, chronological age and non-verbal cognitive ability (Ravens), paper II.

	CI	NH	LI	ASD
N	34	39	12	12
Girls/Boys	15/19	20/19	4/8	3/9
Age (months)	92±15	94±17	89±12	84±14
Ravens (raw scores)	26±5	26±5	17±8	17±9

There was a significant difference between groups as regards non-verbal cognitive ability, which was controlled for in the study when conducting statistical analysis.

3.3.3 General procedure

The project consists of four separate studies with data collected during the period between 2009 and 2012. All children with CI and NH (6-9 years) were assessed by experienced SLPs. The first test occasion was divided in two separate parts (60 minutes each). SLP students collected data from children with LI and ASD in Study II. Finally, a follow-up study was conducted for 18 of the children with CI in the cohort (60-minute assessments). Children with CI were assessed when they visited CIC, Karolinska University Hospital, for regular follow-up visits, except three children who were tested at their school or home-based clinic. Children with NH and children in the other clinical groups (LI and ASD) were tested in quiet office rooms at their schools.

Instructions were only given orally and were presented in the same way to every child, following the test manual and predetermined instructions by the author.

3.3.4 Lexical-semantic ability – assessment battery

A broad and deep test battery was composed with a focus on the lexical-semantic domain, including both quantitative measures of receptive and expressive vocabulary and more qualitative analysis of error responses on the picture naming test and strategy use during retrieval of words from the LTM. Additionally, some phonologically based tests were included as well as a test of verbal memory.

Table 3. Linguistic and cognitive measures and test material used in Studies I-IV.

Measure/Test name	Study I	Study II	Study III	Study IV
Lexical-semantic ability				
Receptive vocabulary (PPVT-3)		X	X	X
Picture naming/Expressive vocabulary (BNT)		X	X	X
Semantic word fluency (animal)	X	X	X	X
Phonemic word fluency (FAS letter fluency)	X	X	X	X
Semantic feature knowledge (Questions & Pictures)		X	X	X
Grammatical sentence understanding (TROG-2)			X	X
Cognitive capacity				
Non-verbal cognitive ability (Ravens)	X	X	X	X
Verbal learning (RAVL test)			X	
Non-word discrimination (SIPS)			X	
Phonological representations (SIPS)				X

Receptive vocabulary - Studies II-IV

The Peabody Picture Vocabulary Test (PPVT-3) was used to examine receptive vocabulary (Lloyd, Dunn, & Dunn, 1997). This is a closed-set test where the child hears a word and is asked to choose among four alternatives (a picture that correspond with the word). Data were collected at test occasions 1 and 2 and were used for different purposes in Studies II-IV. Retrospective test results were included in the fourth study. The total number of correct responses was calculated in accordance with the test manual in study IV and stanine scores were also obtained.

Expressive vocabulary including error response analysis - Studies II-IV

The Boston Naming Test (BNT) was used to examine picture naming ability and expressive vocabulary (Kaplan et al., 1983). The BNT consists of 60 pictures that the child should name orally. The child has to retrieve words from the semantic LTM and name the pictures with the correct lexical-semantic label. Semantic or phonological prompting were avoided and not allowed to gain scores. However, prompting was used in some cases as a strategy for the test administrator to encourage the participants to proceed with the test. An analysis of error responses was conducted for determining if participants in Study II had semantically relevant error responses or not. The test administrators made notations of all verbal responses from the children, including erroneous utterances, and the test was also audiotaped for further analyses.

Words named in accordance with the target word, including synonyms and subordinated words, were counted as correct responses, in accordance with previous research (Tallberg, 2005; Brusewitz and Tallberg, 2008; Storms et al., 2004). This procedure follows the original American procedure (Kaplan et al., 1983), but has been modified to make the error analysis possible.

Analysis of Lexico-Semantic Error Types on BNT

Analysis of lexico-semantic error patterns was conducted on the BNT, using our own model inspired by previous studies of error types on BNT (Tallberg, 2005; Brusewitz and Tallberg, 2008). In the present study, classes of error types were consolidated into ten different categories with a focus on concept understanding. The categories were defined with respect to semantic closeness to the target words and to relevance and hierarchical organization within the lexico-semantic network.

Semantically Relevant Error Types of Responses; examples

1. Super-ordinated words like “statue” for sphinx.
2. Side-ordinated words like “ferret” for beaver.
3. Lexically incorrect responses on single word level but semantically relevant (phonological or morphological errors or phonological associations) e.g. “finx” or “sinks” for sphinx.
4. Semantically relevant single words that are newly formed, either as non-existing single words or as a compound from two correct Swedish words, e.g., “pyramid-lion” for sphinx. These words cannot be found in a Swedish dictionary.
5. Semantically relevant utterances/clauses e.g., “a statue near the pyramids” for sphinx.

Semantically Irrelevant Types of Responses; examples

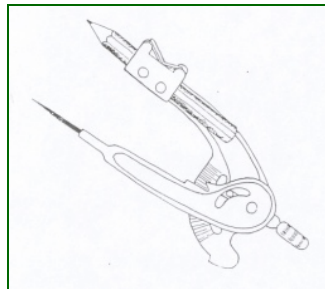
6. Contextual associations of the target word, e.g. “lion” for sphinx.
7. Unrelated words without a direct or close contextual association to the target word, e.g., “cat” for sphinx.
8. Semantically irrelevant single words that are newly formed like “fattice” (non-existing word) for trellis or as a compound from two correct Swedish words, e.g., “time-watch” for protractor. These words cannot be found in a Swedish dictionary.
9. Semantically irrelevant utterances/clauses, e.g., “we have one at home” or “the doctor has one” for stethoscope.

Omitted responses

No responses, responses like “I don’t know” and unfinished responses like “fi:” for sphinx, were counted as omitted responses.

Picture from the BNT (Kaplan et al., 1983), including some examples of semantically relevant responses (in green) and semantically irrelevant responses (in red).

What is this?



”Circle machine”?

Sharpener?

Drawing material?

”Something you can do circles with”

”I have seen one”

?

Inter-rater reliability for the analysis of Error Classification

An inter-rater reliability measurement was conducted and calculated with the Pearson’s correlation coefficient for the picture naming (BNT) data material. One evaluator conducted the classification of error categories. The responses from 30 % of the material were randomly selected and a second evaluator did a blind classification. The correlation between the two evaluators’ separate classifications was calculated, and the correlation between the two classifications of error categories was $r=0.86$, ($p<.001$) for semantically relevant answers and $r=0.95$, ($p<.001$) for semantically irrelevant answers.

Word fluency including cluster analysis Studies I-IV

Word fluency measures the ability of retrieving words from semantic LTM and at the same time gives information about the lexical-semantic network and which strategies an individual uses while generating words. The participant is asked to say as many words as possible within a special category during one minute. The categories used in Study I were semantic word fluency (animal fluency) and phonemic word fluency (FAS letter word fluency). Besides counting correct words, a cluster analysis was conducted to analyse which strategies the children used in the process, similar to the method of Brusewitz and Tallberg (2008).

Semantic Feature Knowledge - Studies II-IV

The Semantic Feature Test is an in-house form, inspired by the repeat-and point-test by Hodges et al. (2008), with two subtests and was used for assessment of semantic knowledge. The first subtest consists of 19 sets of semantically and perceptually similar objects. This subtest examines the child’s ability to visually identify a member of a semantic category, for instance “alligator”, among five pictures of similar animals with some resemblance (chameleon, frog, dinosaur and lizard). The subjects were presented

nouns orally and instructed to point to each noun's pictorial referent. The second subtest consisted of a set of 17 questions concerning knowledge of the semantic features of different objects (for example “*Does the camel have tusks?*”). This subtest examines the child's ability to auditorily identify the exact or shared features of a concept. The subjects are instructed to respond immediately with either yes or no.

Example from the Semantic Feature test (Pictures), target word “alligator”.



Grammatical Sentence Understanding - Study III.

To assess GSU, we used a Swedish version of the second edition of the Test for Reception of Grammar (TROG-2), originally developed by Bishop (2003). TROG-2 consists of 80 items divided into 20 blocks. Each block included 4 target sentences and 12 distractors, aiming to evaluate the same grammatical constructions. The blocks are supposed to be presented in order of increasing difficulty. The examiner read a sentence and the child was told to choose the one of the 4 pictures, which illustrated the sentence in accordance with the test rules. The results were counted for total scores, number of achieved blocks on TROG-2 and percentile scores (Swedish norm data) for group (CI vs. NH) and age (6-7 vs. 8-9 years) comparisons. Additionally, mean scores on individual blocks (1-20) were compared between groups (CI vs. NH) with the aim of examining possible differences or similarities of different grammatical construction abilities.

3.3.5 Cognitive-related capacity and hearing ability

Non-verbal cognitive ability - Studies I-IV

Ravens Colored Progressive Matrices was used to assess non-verbal cognitive ability (Ravens et al. 2003). This test assesses a child's ability to discover and interpret visual patterns in 30 images with different difficulty levels and is often used in language research as a screening tool of fluid intelligence (gF), in this project referred to as non-verbal cognitive ability.

Phonological processing skills - Studies III-IV

A non-word discrimination task was used to assess receptive phonology ability. It consisted of 16 minimal pairs divided into 2 parts with 8 minimal non-word pairs in each part (Wass et al. 2008). The minimal non-word pairs had a change of only one consonant phoneme in one of the two non-words within a pair and only in one of the parts (e.g. sallotan - sallovan). The total score was 8 - both minimal pairs of non-words within each of the 2 test parts had to be correct for a full score of 1. The test was presented through loudspeaker. Similar tests have been used in several previous studies of children with NH and clinical populations including children with CI and NH (Henricson et al. 2012; Lyxell et al. 2009; Reuterskiöld-Wagner, Sahlén, & Nyman,

2005; Wass et al. 2008). The presentation of each stimulus was presented only once. A test of phonological representations was used in Study IV for 18 children with CI who were examined by means of a longitudinal design. The task was to identify three different phonemes (s, t and n) in initial, middle and final position of a real word. The quality of the phonological representations of real words was measured by asking the participants to determine which one of five alternative productions of a word that was correct or not without audiovisual information and with live-voice presentations by the test administrator (Wass et al., 2008).

Learning ability - Study III

Rey Auditory Verbal Learning Test (RAVL) was used to investigate verbal learning (Rey, 1941). The child is told to listen carefully and try to remember 15 nouns, commonly used in everyday communication. The words are read aloud by the test administrator five times in a row and after each time the child is asked to repeat the ones that he/she remembers. The test investigates immediate verbal memory span and learning ability. The test was performed in accordance with the standard administration rules. Scores from trials 1-5 were summarized and counted as total score.

Early spoken language abilities - Study IV

Retrospective data was collected from medical records for children with CI after one year, one and a half years and two years with their 1st CI. Language understanding had been assessed with The Reynell Developmental Language Scales (RDLS-3) (Edwards, 1997). Only the comprehension scales of the RDLS-3 had been assessed. Total scores were counted and compared between groups of children with CI. Additionally, the speech intelligibility rating scales (SIR-2) (Allen 2003) were included in the analysis of early language abilities in Study IV. Furthermore, expressive grammar level had been rated by SLPs at the CIC, using a local in-house rating scale from 0-8, which illustrates an increased use of syntax in particular.

Level	Description
1	No use of voice with intent
2	Use of voice with intent
3	Emerging cv-babbling such as /mamama/, /bababa/
4	One-word utterances
5	Successive one-word utterances without grammatical hierarchy such as “car big”
6	Two-three word utterances
7	Multi-word sentences with atypical or incorrect grammar (articles, word order, morphology, conjunctions)
8	Typical or correct expressive grammar and sentence level

Table 4. Expressive grammar level: an in-house rating scale, CIC, Karolinska University Hospital.

Hearing ability - Studies I-IV. Speech recognition level was tested for all children with CI by experienced audiologists at the CIC in quiet, bilateral listening conditions, using phonemically balanced lists of words developed for children at 65 dB SPL level (Asp et al. 2012). Time with 1st and 2nd CI as well as time between CI were included as possible independent variables in the statistical analysis of Studies I-IV. Hearing status was also assessed in the typically developed and NH participants. The hearing levels of all controls with presumed NH were screened at their school using a portable audiometer. The equipment had recently been calibrated at the local department of audiology. Screening took place in a small office room with a low ambient noise level. All participating children showed NH (0-20 dB HL at 500, 1,000, 2,000, 4,000, ND 6,000 Hz). Children within the LI and ASD groups were not tested for their hearing at the time of data collection.

3.4 STATISTICAL ANALYSES

In all four studies, descriptive statistics were first conducted.

Study I: two groups of children (CI and NH), each divided into two age groups (6-7 years vs. 8-9 years), were compared regarding performance on FAS and animal fluency, using two-way ANOVA. The dependent variables were first the total score on phonemic and semantic word fluency tasks, and then specific variables regarding strategies. The possible influence of background factors on phonemic and semantic word fluency tasks were analyzed by entering background factors as covariates in the two-way ANOVA. For non-verbal cognitive ability, the Raven score was used as covariate. For the other background variables: age at implantation (two groups according to median split), duration of CI-use (two groups according to median split), cause of deafness, school setting (mainstream vs. special class for children with HI) and speech recognition (two groups according to median split) were used together with age (6-7 and 8-9 years) in two-way ANOVA when analyzing the effect on performance of phonemic and semantic word fluency tasks within children with CI. Post-hoc analyses of single group comparisons were performed by means of Student's t-test. An inter-rater reliability measurement was conducted for word fluency tasks and this was calculated with the Pearson's correlation coefficient.

Study II: one-way analyses of variances (ANOVAs) were performed to examine possible group differences as regards picture naming including error response analysis, and receptive vocabulary, respectively, as well as semantic feature knowledge. Language test results for all four groups were controlled for age, gender and non-verbal cognitive ability, using covariate analyses. For the group of children with CI, language test results were also controlled for background factors, like age at implantation, time with CI, and speech recognition, using covariate analyses. Post-hoc analysis (Scheffe's) of single group comparisons was performed by means of Student's t-test. An inter-rater reliability measurement was used for the error response analysis on BNT, and this was calculated with Pearson's correlation coefficient.

Study III: a regression analysis was performed to analyze which factors were associated with GSU, besides conducting group comparisons (NH vs. CI).

Study IV: the relationship between age at 1st and 2nd implantation as well as early language development after 1 and 2 years with 1st CI, linguistic test results at the age of 8-9 years including receptive and expressive vocabulary, and word fluency abilities

were investigated by means of Pearson's correlation coefficient in the CI-Y group and in comparison to children with CI in the CI-O group.

Statistical calculations were performed using the SPSS for Windows (Statistical Package for the Social Sciences, 2010, version 19-22).

3.5 ETHICAL APPROVAL

The population of children with CI is small and heterogeneous, demanding extra care concerning ethical considerations to avoid that individual children are identified. Therefore, some individual information like chronological age and gender has not been presented in tables. All caregivers signed a form for letting their child take part in the studies and children who could read also signed an agreement to take part in the study. Children were told that they could decide to stop participating, at any time during the assessment and in some cases the test administrator interrupted individual tests before they were finished because children indicated that they did not manage to complete single tests, due to fatigue or for unclear reasons. The studies in the PhD-project were approved by the Regional Ethical Board in Stockholm (Dnrs 2009/1724-31/1, 2010/2083-32/1).

4 RESULTS

Study I. Word Fluency Performance and Strategies in Children with Cochlear Implants: Age-Dependent Effects?

Aims To examine word fluency ability in children aged 6-9 with CI (n=34) and in comparison to age-matched children with NH (n=39). Another aim was to explore whether children with CI used different strategies in the retrieval process compared with NH children. Finally, non-verbal cognitive ability in both groups and background characteristics of children with CI were examined in relation to the linguistic results.

Method The cohort was divided in two age groups; 6-7 years and 8-9 years for developmental reasons; eighteen children aged 6-7 years with CI in comparison to twenty children with NH and sixteen children aged 8-9 years with CI in comparison to nineteen children with NH. Two different word fluency tasks; one phonemically and one semantically based, were used to explore word fluency ability in conjunction with exploring the use of strategies in the retrieval process.

Results & Discussion The results showed that there were no significant differences in retrieving words from LTM between younger children (CI vs. NH) aged 6-7 years, regardless of whether they were performing semantic or phonemic word fluency tasks. They also used the same type of strategies in the retrieval process. However, children with CI aged 8-9 years had significantly poorer results on the phonemic word fluency task, and close to significantly worse results on the animal fluency task. Furthermore, this group of children with CI used less efficient strategies in the retrieval process compared with controls. Age at 1st and 2nd CI implantation was not associated with better results in either of the age groups. However, the results indicated a possible difference between age groups. Considering the equally low results on the phonemic word fluency task of children aged 6-7 years regardless of group (CI or NH), which is typical for age, it was difficult to state if this group had closed the gap or if they would lose ground and there would be a gap opening over time. The findings of this study led to the planning of a fourth follow-up-study, as an attempt to examine if the group of younger children with CI would continue to have age-equivalent word fluency results at the age of 8-9 years or if there might be a possible gap opening in performance across age for all children with CI, especially for the phonemically based FAS letter fluency task. The results from this study showed that age was an important factor, indicating that children with CI, at least partly followed the same kind of developmental patterns like age-matched NH controls.

Conclusions Age correlated with results on word fluency, both in children with CI and NH, aged 6-9 years. Differences in results with age-appropriate outcome at school entry in one group and poorer outcome two years later in another group of children with CI, indicated either a general gap opening or group-specific differences in the sample, possibly due to unknown background characteristics, which were most likely related to earlier ages at 1st CI in children aged 6-7.

Study II. Lexical and semantic ability in groups of children with cochlear implants, language impairment and autism spectrum disorder

Aim. The primary aim was to explore the lexical-semantic ability of children with CI aged 6-9 years in relation to age and non-verbal ability. Additionally, another aim was to compare lexical-semantic ability among the group of children with CI with age-matched NH controls and with two other clinical groups (LI and ASD).

Method Ninety-seven children participated and were divided into four groups: CI (n=34), NH (n=39), LI (n=12) and ASD (n=12). A battery of linguistic tests, including picture naming, receptive vocabulary and knowledge of semantic features was used for assessment. Moreover, a semantic response analysis of the erroneous responses on the picture-naming test was performed.

Results & Discussion Children with CI exhibited a naming ability comparable to that of the age-matched group of children with NH, and they possessed a relevant semantic knowledge of certain words that they were unable to name correctly. Children with CI more often had semantically relevant responses than omitted responses or semantically irrelevant answers, thereby demonstrating semantic knowledge in cases when they lacked the lexical term. Children with LI and children with ASD showed a more atypical response pattern and were more likely to not respond at all (LI) or give semantically irrelevant responses (ASD) when they could not name the picture correctly. This result indicates less semantic knowledge. Furthermore, children with CI had significantly better understanding of words compared with children with LI and ASD, but worse receptive vocabularies than children with NH. This demonstrates a smaller and less developed breadth of receptive vocabulary in children with CI. The significant differences between groups remained after controlling for age and non-verbal cognitive ability. Children with CI showed a somewhat atypical pattern compared with normal development, with better expressive than receptive vocabulary. Again, like in Study I, age at CI implantation was not significantly associated with the level of lexical-semantic ability in children with CI.

The findings from the second study also influenced the planning of the fourth follow-up study, especially regarding examination of the atypical pattern of expressive and receptive vocabulary in the group of children with CI.

Conclusions Dissimilar causes of neurodevelopmental processes seemingly affected lexical-semantic ability in different ways in the clinical groups. Children with CI aged 6-9 years had expressive vocabularies comparable to those of NH controls, but significantly poorer receptive vocabulary.

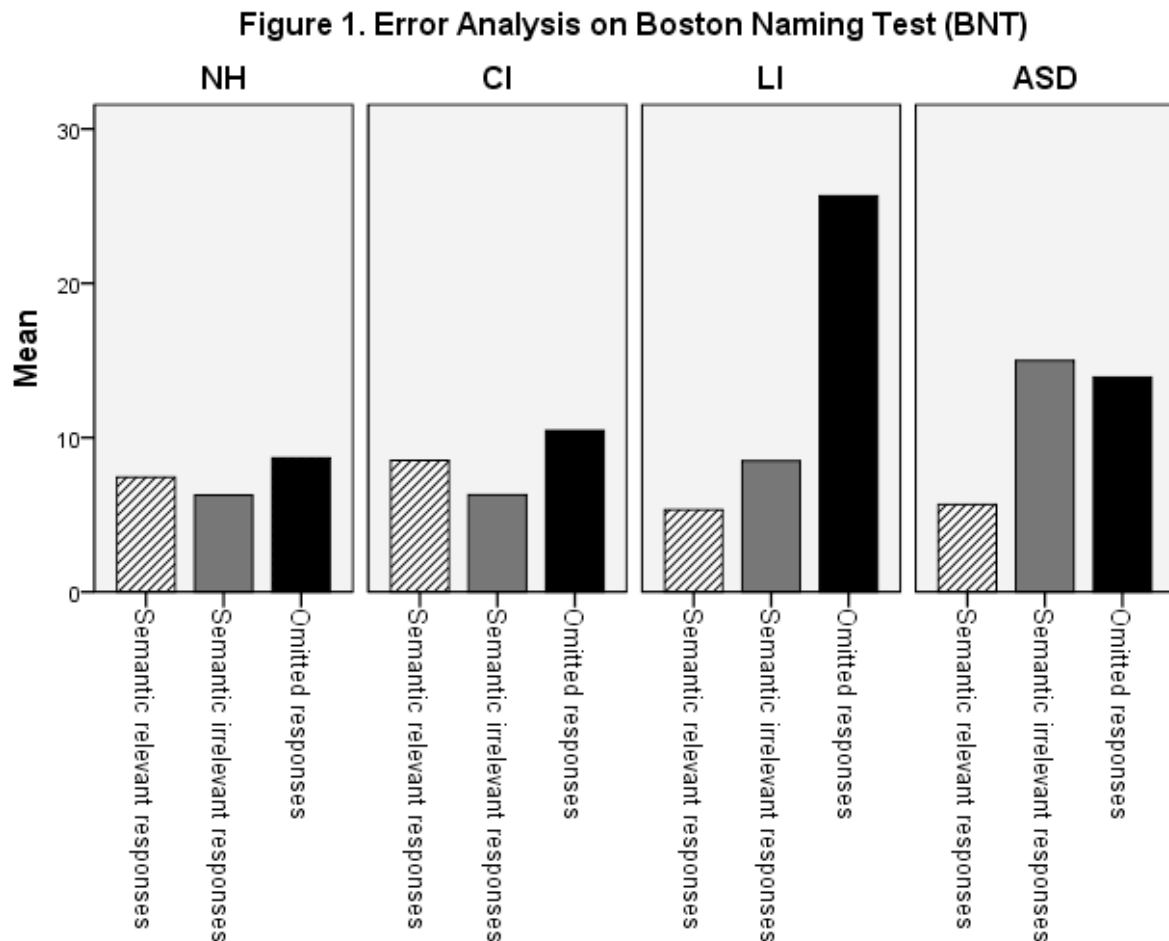


Figure 5. Group means (total scores) for semantically relevant responses, semantically irrelevant responses, and omitted responses from the error analysis of the Boston Naming Test (BNT) for four groups (NH, CI, LI and ASD) (*Study II*).

Study III. Non-verbal cognitive ability and Semantic capacity are associated with Grammatical Sentence Understanding in Children with Cochlear Implants

Aim To examine lexical-semantic ability on sentence level in relation to cognitive and linguistic factors in children with CI and in comparison to age-matched NH controls.

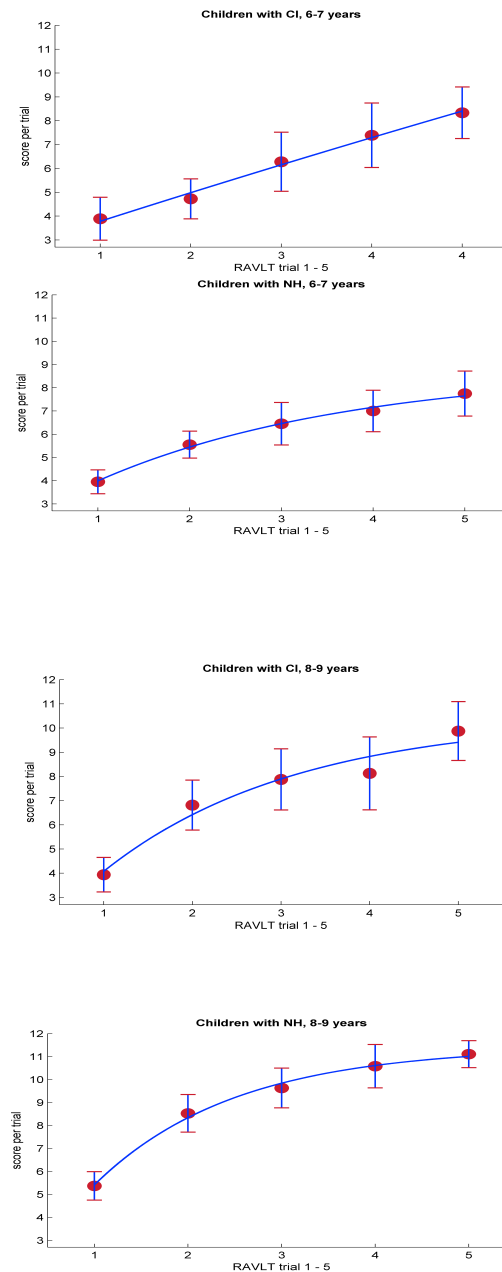
Method Grammatical sentence understanding (GSU) was examined using the TROG-2 test in the cohort of children with CI (n=34) and in comparison with NH controls (n=39). In addition, possible predictors of GSU including lexical-semantic ability, verbal learning, receptive phonology and non-verbal cognitive ability, were examined in a regression analysis.

Results & Discussion Non-verbal cognitive ability and semantic capacity were predictors of GSU in both groups (CI and NH). Age at 1st CI and receptive phonology did not explain the GSU outcome in the sample. There was a tendency toward an influence of younger ages at 2nd CI on a better outcome of GSU. Children with NH had significantly better raw scores on TROG-2 than children with CI. Children who were 6-7 years had age-equivalent scores on total number of blocks on TROG-2, as compared

with Swedish norm data. There was no significant difference between groups on verbal learning (see Figure 5). Children with CI basically used the same kind of learning strategies as NH children, although there was a greater variation of results for single trials (1-5) on the verbal learning task.

Conclusions The findings suggest that children with lower non-verbal cognitive ability and/or deficient expressive vocabulary are at risk of developing less adequate GSU ability, which in turn might influence their knowledge learning and social skills negatively. Another conclusion is that caregivers and interventionists should stimulate lexical-semantic abilities, to prevent some of the GSU variation within the group of children with CI.

Figure 5. Learning curves on the RAVL-test, trials 1-5, for children with CI and NH children in two different age groups (6-7 and 8-9 years).



Study IV. Influence of age at implantation on lexical-semantic development in children with cochlear implants

Aims One aim of the fourth follow-up study was to compare lexical-semantic ability between groups with younger (CI-Y, n=18) and older group mean ages (CI-O, n=16) at 1st CI-implantation and NH controls (n=19) after school entry, at the age of 8-9 years. A second aim of the fourth study was to examine the longitudinal results of a subgroup of children with CI (CI-Y, n=18) within the cohort. One hypothesis was that the lack of significant influence of age at implantation of lexical-semantic ability, as demonstrated in previous studies (Studies I-III), might change after school entry. Earlier ages at 1st CI was predicted to be associated with better linguistic abilities including phonological skills as a consequence of literacy. Another prediction was that an earlier age at 1st CI also would have influenced on early spoken language development.

Method Lexical-semantic ability was assessed using tests of vocabulary and word fluency at two occasions for a subgroup of children and at one occasion for comparative groups at the age of 8-9 years. In addition, phonological representations were measured in the subgroup at the age of 6-7 years. Retrospective test results from follow-up visits at CIC, after one year, one and a half years and two years with 1st CI, were collected from medical records. Then two correlation analyses were performed for this subgroup as well as for all children with CI, as an attempt to examine the influence of age at 1st CI, but also to follow up if there was a possible gap opening for children with CI for receptive vocabulary.

Results & Discussion Children with CI developed age-equivalent lexical-semantic ability over time. Younger ages at 1st implantation and a shorter period between 1st and 2nd CI affected early spoken language abilities significantly. Expressive language levels after 2 years with CI correlated with expressive vocabulary at the age of 6-7 years. Semantic knowledge demonstrated at the age of 6-7 years was associated with early spoken abilities and had a significant influence on most lexical-semantic abilities before and after school entry. There was a nearly significant difference between children in the CI-Y group and NH peers in regards to phonemic word fluency, indicating that children in the CI-Y group did continue to have age-equivalent abilities of retrieving words when the category was phonemic based. However, the same group had surprisingly poor semantic fluency, indicating a possible gap opening, and also somewhat worse expressive vocabulary compared with the group of NH children. One possible explanation is that they had more help from literacy ability and phonological awareness than the group of children with greater age at 1st CI, who had significantly worse results than NH controls on all lexical-semantic parameters, except semantic word fluency.

Conclusion Children with CI and typical non-verbal cognitive ability developed age-equivalent lexical-semantic abilities over time, though the developmental patterns were heterogeneous within groups. Age at 1st CI had a significant correlation with early spoken language and functioned as a starter engine for spoken language. Early expressive grammar after two years with CI and semantic knowledge before school entry were associated with better vocabulary at the age of 8-9 years.

Table 6. Individual test results in the CI-Y group (n=18) from test occasion 1 (1): 6-7 years and test occasion 2 (2): 8-9 years showing chronological age at testing (CA) in months, total raw scores of each test and a summary of mean/SD.

	PPVT-3	BNT	Semantic W.F.	Phonemic W.F.	Ravens
CI-Y Group					
C10 (1)	150	39	13	14	31
C10 (2)	146	40	14	18	34
C11 (1)	110	36	14	9	20
C11 (2)	-	42	18	11	30
C12 (1)	136	35	12	9	25
C12 (2)	144	43	10	11	32
C13 (1)	113	39	17	10	30
C13 (2)	144	41	19	30	31
C14 (1)	89	33	8	13	22
C14 (2)	143	-	11	17	21
C15 (1)	83	34	13	7	16
C15 (2)	120	38	14	15	27
C16 (1)	53	18	10	0	21
C16 (2)	-	23	12	8	23
C17 (1)	91	32	12	12	24
C17 (2)	132	41	15	19	31
C18 (1)	133	43	16	8	29
C18 (2)	118	45	18	17	30
C19 (1)	59	23	13	1	31
C19 (2)	130	39	16	11	34
C27 (1)	87	34	12	10	31
C27 (2)	137	40	12	14	33
C28 (1)	110	35	13	7	25
C28 (2)	118	36	16	16	34
C29 (1)	74	24	15	2	29
C29 (2)	131	35	15	14	36
C30 (1)	109	36	17	19	30
C30 (2)	131	38	16	26	36
C31 (1)	74	30	9	5	29
C31 (2)	141	48	16	28	30
C32 (1)	86	40	10	3	22
C32 (2)	153	38	21	24	35
C33 (1)	56	18	4	5	16
C33 (2)	138	41	16	23	27
C34 (1)	72	20	10	3	28
C34 (2)	91	28	19	9	29
Summary (1): M (SD)	93.61 (28.10)	31.61 (7.76)	12.11 (3.29)	7.61 (4.97)	25.50 (5.04)
Summary (2): M (SD)	132.31 (15.08)	38.59 (5.91)	15.44 (2.96)	17.28 (6.58)	30.72 (4.20)

5 GENERAL DISCUSSION

Children with CI and typical non-verbal cognitive ability did not have specific deficiencies in cognitive processing of lexical-semantic items, but some children had deficiencies in the storage of representations and semantic knowledge. Recent studies by Nittrouer et al. (2013) and Kennett et al. (2013) report similar findings. The results from the four studies contribute to the research field with some new pieces of information for the bigger picture, explaining part of the variation in language outcome in children with CI. This new information primarily concerns semantic capacity, with a focus on lexical-semantic ability on word level. Some of the overall findings will now be discussed and finally some clinical implications and recommendations for future research will be mentioned

Age-appropriate levels of lexical-semantic ability in children with CI 8-9 years

The thesis consists of four separate, although intertwined studies, all exploring the nature of lexical-semantic ability in the same cohort of children in the ages 6-9 years. One hypothesis was that school-aged children with CI would have poorer results both regarding breadth and depth of their mental lexicon in comparison with NH children.

Children with CI had age-appropriate group means on expressive vocabulary. This result did not confirm the original clinically based hypothesis for the PhD-project. The results also showed that many children with CI had semantic capacity demonstrated by, for example, an age-appropriate level of semantic feature knowledge. Another example was their use of semantically relevant responses when children with CI (6-9 years) could not find the correct lexical term while naming pictures. Especially the group of younger implanted children (CI-Y) had age-equivalent lexical-semantic outcome demonstrated in the fourth follow-up study at the age of 8-9 years. In the group of children with higher ages at 1st CI-implantation (CI-O) there were some children who also managed to close the gap and reached age-equivalent results, indicating that there are also other factors that influence on lexical-semantic ability besides from early age at 1st CI (Boons et al., 2012; Geers et al., 2011; Niparko et al., 2010).

Despite these overall positive results, which ruled out the initial hypothesis of poor performance, there were also contradictory results in the sample. Some children with CI had more problems with storage in the semantic LTM, including knowledge of words on lexical level, compared with age-matched children with NH. These results are also in line with recent findings by Jerger et al. (2013) and Kenett et al. (2013).

Children in the CI-O group had significantly poorer results on receptive vocabulary, phonemic word fluency and GSU. These three abilities have previously been explained by younger age at CI and better PWM or phonological skills (Willstedt-Svensson et al., 2004). Despite these results, the whole sample of children with CI had an age-appropriate level of expressive vocabulary and relevant semantic feature knowledge. These results are in line with similar findings within this population showing poor phonological skills but still age-equivalent linguistic related cognitive abilities like reading (Lyxell et al., 2009; Geer and Hayes, 2011).

How can we explain the differences between the subgroups?

Most demographic factors were equal in the two subgroups (CI-Y and CI-O), except for their chronological age at study start, age at implantation and to some degree school settings. There were no significant differences between groups in regards to hearing ability, hearing age, non-verbal cognitive ability or gender. However, one possible cause of the subgroup differences was the differences of mean age at 1st CI, and also age at 2nd CI. In addition, there were exceptions in both CI-groups, illustrating a larger variation within the population of children with CI, than in NH controls (Geers et al., 2009; Wass et al., 2008). Some children in the sample with relatively higher ages at 1st CI reached age-equivalent lexical-semantic results. Furthermore, some children with younger age at 1st CI did not reach age-equivalent results after school entry. Thus, indicating that other factors also influenced lexical-semantic outcome in children with CI besides age- and hearing-related factors, previously found in previous studies (Boons et al. 2012; Szagun and Stumper, 2012). Duchesne et al. (2009) found four different language profiles within their sample of only 27 children with CI with a mean age of 5 years and age at 1st CI of around 22 months. The range was from typical language levels of grammatical skills and vocabulary, to general language delay. Age at CI did not explain the language results. The results of Duchesne et al. (2009) are similar to the results of this PhD-project in that sense that different language profiles were found the sample, and that age at 1st CI was not directly related to the results.

In addition to the partly different individual and subgroup results of lexical-semantic ability in studies (I-IV), an overall atypical trajectory of lexical-semantic development was found in the whole sample of children with CI, with better expressive than receptive vocabulary levels. Although, this trajectory was partly different in the CI-Y and CI-O groups with more atypical developmental patterns in the CI-O group, indicating that a longer period with early auditory deprivation had affected brain development (Kral and Sharma, 2012). Chilosi et al., 2013 found a similar pattern in a group of Italian children who had better expressive than receptive vocabulary outcome. They suggested that this pattern is a result of the children's initial auditory deprivation and later different hearing with CI. Another explanation, suggested in this thesis, is that the unusual developmental pattern is explained both by their atypical hearing situation as suggested by Chilosi et al. (2013) but also for environmental reasons. One difference between subgroups was that children in the CI-Y group had higher levels of expressive language use after two years with CI than the CI-O group. This could have been influenced by their earlier CI intervention leading to a more synchronized spoken language situation or parents who have communicated more on spoken language, and thereby stimulated the child's own use of expressive language (Leigh et al., 2013; Szagun and Stumper, 2012). Cruz et al. (2013) found in their study of children with CI aged 0-3 years that higher level strategies, such as use of open-ended questions and also use of more word types by parents in interaction with their child predicted expressive language. The study by Cruz et al. (2013) contained ninety-three children and was based on video-analyses from dyads in free play situations. Weislader and Fernald (2013) also found an impact of parent interaction patterns for expressive vocabulary use in young children with NH. These two results strengthen the assumption that better expressive vocabulary in the current PhD-project could be explained by parent

involvement that have affected early interaction patterns in a favourable way, which have lead to better linguistic outcome. However, a prospective and longitudinal study would be necessary to gather more evidence that age at implantation is causing the atypical linguistic patterns, with better expressive than receptive levels in children with CI and whether environmental factors like parent-child interaction also is involved in the process.

Semantic knowledge and word retrieval ability

The study design aimed to explore lexical-semantic ability and not specifically to examine cognitive capacity, but to relate the results on linguistic assessments to non-verbal cognitive ability. In Study 4, it was shown that children in the CI-Y group did not have specific problems retrieving words in a phonemically based letter word fluency task, while they did have significantly poorer performance than age-matched NH children on the semantically word fluency task. Children in the CI-O group had significantly worse results than NH children in both categories, although showed better semantic word fluency ability. Previous research has shown that the phonemically based letter word fluency is related to memory processes like retrieving ability, while semantically based word fluency is more related to size and organization of the vocabulary (Riva et al., 2004; Storms et al., 2008). Children with CI aged 6-7 years used similar strategies in the process of retrieving words as age-matched NH children, both in semantically and phonemically based word fluency tasks. Children in the CI-O group used similar strategies but less efficient than controls. Further support for the argument that children with CI do not have specific problems with memory processing is that they used similar strategies while performing a verbal learning task (RAVL) and showed similar patterns of learning in the acquisition phase (Study III). This indicates that, especially earlier implanted children with CI did not have problems with retrieving strategies, but instead in some cases had a smaller and less developed mental lexicon, in line with results of Nittrouer et al., 2013. Children with CI in the sample had significantly lower results on the non-word discrimination task and had lower level of phonological representations. None of these tests were related to level of vocabulary or word fluency ability in the sample. Seemingly, lexical-semantic ability itself had an impact on word knowledge and the organisation of vocabulary.

Early age at 1st CI - a starter engine for linguistic learning?

The lexical-semantic ability in children with CI relies both on earlier ages at CI-intervention and earlier linguistic experiences (Niparko et al., 2010; Szagun & Stumper, 2012). In fact, Quittner et al. (2013) found in their study that the magnitude effects of observed maternal sensitivity and cognitive stimulation on oral language growth was similar to that of age at implantation. The lack of auditory stimulation during the auditory deprivation period from utero and until a deaf or HI child receives HA/CI negatively affects the development of neural networks important for spoken language development (Kral and Sharma, 2012). Several studies have shown the benefits of implantation before 12 months of age (Colletti et al., 2011; Karltorp et al., submitted; Wie, 2010). Earlier age at 1st implantation was not found to be directly associated to better lexical-semantic ability results in the sample of children with CI. However, when early spoken language results were analysed in relation to age at 1st and 2nd CI

implantation, some interactions were found. Earlier ages at 1st CI were important for early language development in the study group. Later on, expressive grammar levels after two years with CI and semantic capacity at school entry (6-7 years) were found to be important and associated with better lexical-semantic abilities at the age of 8-9 years in children with CI. Surprisingly, phonological abilities at school entry were not explained by younger age at implantation, nor did they explain the lexical-semantic ability in children with CI. Instead, phonological results were to some degree related to the lexical-semantic ability results of NH controls, despite children with CI having significantly worse results on e.g. non-word discrimination.

To summarize, age at 1st CI is not always predictive of age-appropriate lexical-semantic ability, shown in most studies of cognitive development where cognitive factors mostly correlate with age at 1st CI (see, Wass et al., 2008, for an overview). The results indicate that an earlier age at 1st CI is important as a starter engine of spoken language development through listening, but that other more linguistic-related factors and strategies were important for later language learning in the sample of children with CI.

Use of semantic learning strategies with a less robust phonology and poor lexical-semantic network?

When children with CI were provided with semantic information in a verbal learning task they performed at the same level as children with NH, whereas their performance in a non-word discrimination task without semantic content was not at the same level (Study III). This is also a replication of a number of previous studies (Cleary, Pisoni, & Kirk, 2003; Wass et al., 2008). In the CI-Y group there were no significant differences between NH peers on the ability to use phonological representations, which has been found previously (Lee, Yim & Sim, 2012). They also used the same kind of retrieval strategies in word fluency tasks when they were 6-7 years old. Two years later, they generated significantly fewer words in the semantic word fluency task (animal) but similar amount of words in the phonemically based word-fluency task. It seemed that the lexical-semantic network were less robust concerning the semantically related network or that the retrieval strategies were less efficient. Kenett et al., 2013 found in a recent study of verbal fluency in children with CI that the semantic network was less well specified. They found that children with CI had smaller lexical-semantic networks and that there were fewer connections in between concepts.

One possible explanation for this result is that children with CI have to struggle more to learn new spoken words in natural settings, both incidentally and consciously. Their lexical-semantic knowledge might be somewhat shallow in comparison to NH peers and in worst cases “fluctuating”, meaning that words that they have learned are forgotten unless they are used frequently enough. The age-equivalent results on animal fluency at 6-7 years and significantly worse performance two years later indicated specific difficulties in organizing animal words, which is in line with the results of Kenett et al. (2013). However, the group result of children in the CI-Y group was still within reasonable levels for their chronological age at the age of 8-9 years. Another reasonable explanation for their worse results on semantic fluency (animal) could be that their focus had changed somewhat and been shifted towards greater interest in letters and phoneme-grapheme after school entry.

Interestingly, despite worse receptive phonological ability and in some cases later ages at 1st CI, there were some children in the sample who reached age-equivalent lexical-semantic ability at the age of 8-9 years. Some of the children in the sample even had results that were above average of children with NH. However, it seemed that some children with CI had less developed lexical-semantic networks, in line with Kenett (2013). One explanation for this result might be that children with CI instead uses other, perhaps more semantically related strategies in learning words and processing language than children with NH. These results are also in line with previous findings concerning literacy ability in children with CI (Lyxell et al, 2009; Nakeva von Mentzer, et al., 2013; Geers and Hayes, 2011).

Different clinical groups show different lexical-semantic patterns

Children with CI showed different patterns compared with children with LI and ASD in Study II. They showed more semantically relevant knowledge and had larger expressive vocabulary, although there was no significant difference between children with CI and LI regarding receptive vocabulary level. Two years later, in the fourth follow-up study several children in the CI-Y group had receptive vocabularies above expected levels for NH children on group level. Seemingly, the nature of development and origin for less developed lexical-semantic representations are different in the three groups. Several studies have shown that children with LI have problems with both word-retrieval and storage, to some extent explained by both deficient phonological skills and semantic knowledge (Messer and Dockrell, 2006). Similar findings have been made among children with ASD, although their difficulties can largely be explained by their problems with ToM (Miniscalco et al., 2014). This group had the highest numbers of semantically irrelevant response in the error response analysis compared to other three groups. One overall conclusion to make from Study II is that children with CI had a more typical semantic response pattern than the other two clinical groups. What would have been interesting is to also have included children with children with dyslexia, with known phonological difficulties influencing on reading but age-appropriate language ability, for comparisons. Additionally, it would have been interesting to compare semantic knowledge in children that has lost another of the five senses, such as blind children.

Figurative language – next step of CI-intervention

Most children with CI in this thesis had age-appropriate lexical-semantic ability, shown in formal assessments and more quality-based analysis like linguistic cluster analysis and error response analysis. The semantically relevant responses indicate great creativity, but also, in some cases, a less developed network and fewer words in the lexicon. What was not measured was what kind of words that children used, if there were any differences regarding low- or high-frequency words. Another topic that was not measured was the level and use of figurative language, which could be an indicator of the depth in the language. Children at these ages, especially at 8-9 years, and presumably those who have gone through the syntagmatic-pragmatic shift, could have used some of these higher-level aspects of semantic knowledge. If children with CI learn receptive words as quickly as indicated in Study 4 and also have much higher level of phonemically word fluency, one assumption is that this is explained by better overall phonological awareness and literacy skills, but also lexical-semantic awareness. *Lexical-semantic awareness*, meaning to shift focus from the lexical form to different

semantic meanings of the same word units. Figurative language also includes irony and ambiguous language, which could be difficult to grasp for children with HI (Nicastri et al., 2014). Poor figurative language level could explain some of the lower pragmatic language skills reported, despite age-appropriate language (Goberis et al., 2012). The literature contains only sparse reports regarding how figurative language develops, but it is believed that cognitively matured children in later school years can understand and use metaphors. However, poor ability of handling figurative language may affect social interactions as well as problem solving in NH adolescents (Im-Boulter, Cohen, & Farina, 2013) and has therefore to be further explored and investigated in clinical groups, like children with CI. In one of the few studies that examined figurative language in children with CI, Nicastri et al. (2014) found that children with CI in the ages 6-15 years showed significantly worse ability to understand figurative meanings of metaphors.

What does a word mean? Well, it is not enough to just learn the names and lexical terms of objects. This will not be beneficial for language use in conversations (Bloom, 2002) or only to decode words orthographically while reading. Deeper knowledge of concepts and words is highly important for e.g. developing figurative language and to understand metaphors, irony and ambiguous language, commonly used orally and in written contexts (Nicastra et al., 2014). One interpretation of Patterson et al. 2007 would explain deeper word learning of a concept and a word like *the sea*, would preferably engage all senses at the same time for optimal semantic feature knowledge. Both the smell, the salty taste of the water, the sounds, colours and shapes of the waves rolling in to the shore and the feeling of *infinity* or *freedom*. Words are most often used for describing abstract concepts, emotions or as a language of force in argumentation with a rhetorical purpose in conversation with others or in literature. Children who have never seen the sea will learn the name and adopt the word in a lexical-semantic network, regardless of if she or he has experienced the sea with all senses. However, the deeper knowledge of the concept is less evident according to Patterson (2007) and will therefore not engage the same amount of semantic features in the hub, which will have a negative influence on the child's use and understanding of the word. When children reach school age, a new era starts in word acquisition and they learn many words every school year, mainly through literature and increasing knowledge.

Bloom (2002) would add another dimension in learning the word sea, namely that the word sea and the other words associated to the concept are learned in meaningful communication with the child. Meaningful means that caregivers not only name and talk about surrounding objects but that they also listens carefully for the verbal initiatives from the child. The child learns new words in communication (Kuhl, 2010). However, an objection to adopting the theory of Patterson et al. is that they build their hub-theory on adults, not children. Adults have already developed a language and therefore use more knowledge-based top-down strategies than children.

5.1 CONCLUSIONS

- Children with CI with typical non-verbal cognitive ability can develop an age-equivalent lexical-semantic ability over time, but not all.
- Children with CI had adequate semantic knowledge and used age-appropriate strategies in the process of storing and retrieving real words but had less developed ability of discriminating between non-words, indicating that they use semantically based strategies rather than phonologically based strategies for linguistic processing.
- Indistinct or incorrect storage of words on a lexical level due to unclear or sometimes non-existent speech signal may cause poor semantic knowledge of concepts and words in children with HI and CI. This in turn can provide a less effective and smaller vocabulary.
- Children with CI and typical non-verbal cognitive ability have better lexical-semantic ability than other clinical groups with NH, like children with LI and ASD.
- Non-verbal cognitive ability and expressive vocabulary predicts grammatical sentence understanding in early school years, both in children with CI and NH children.
- Semantic feature knowledge at school entry (6-7 years) and higher levels of expressive language abilities in early childhood are associated with higher levels of receptive and expressive vocabulary after school entry (8-9 years) in children with CI.
- Younger age at 1st implantation is related to better language understanding and higher levels of expressive language after two years with CI.
- Some children with higher age at 1st CI catch up over time and reach age-equivalent lexical-semantic ability and some children with younger age at 1st CI do not catch up over time.

5.2 CLINICAL IMPLICATIONS

The seedbed for this project was observed clinical findings of children who had poor expressive vocabulary in relation to their level of language understanding. The initial hypothesis at study start was therefore that children with CI would have poor semantic knowledge and small vocabularies. However, the results from this project only partly confirmed this assumption. Instead, many children reached age-appropriate scores on several tests of lexical-ability after school entry. There were some children in the sample with more atypical patterns and the variation of outcome was larger within the group of children with CI than for NH children.

One clinical implication that can be made is therefore that spoken language intervention in the population of HI children with CI and/or HA should not be uniform but instead individually driven. One result that was evident only in Study IV was that earlier ages at implantation had an influence on early language ability and that especially expressive language influenced later semantic feature knowledge and vocabulary.

Additionally, semantic capacity at the age of 6-7 years had an influence on receptive and expressive vocabulary two years later in children who had a mean age of 18 months at 1st CI. Another clinical implication is therefore that spoken language intervention should be based on prevention of potential later difficulties. If necessary, more specific training should also be included, as early as possible. Fewer children will need specific speech or language training if development can be accomplished within sensitive periods for linguistic learning. However, all children regardless of age at implantation could be at risk of having specific language problems due to other causes than HI or deafness, and therefore it is crucial that all children with severe-profound HI are followed regularly for evaluating both of their cognitive capacity and spoken language ability and development, by professionals like psychologists and SLPs.

Word learning is a life-long project that starts as soon as a child is born, or even before birth. Special efforts should therefore be made to provide all families with early intervention from specially trained professionals like teachers of the deaf or SLPs that can guide and support parents during the first, most important period when spoken language is established. Sensitive interaction and meaningful communication is the key for word learning (Bloom, 2002).

Even if most results did not have a direct link to ages at implantation, the longitudinal patterns revealed in a follow-up study confirm previous evidence of the importance of early implantation, especially for receptive vocabulary and phoneme based word fluency. Also, earlier ages at 2nd CI should be promoted, when a child meets the criteria, as it was shown to influence GSU to a certain extent. Earlier age at 1st CI is interpreted as a *starting engine* for cognitive capacity and spoken language development for children with severe-profound HI.

However, it seemed that it was the actual lexical-semantic development itself that promoted further linguistic development in this sample. Still, it is unclear how robust the lexical-semantic representations are and how well established the semantic hierarchical network is and how well-developed the associations between words are in

their LTM. Seemingly, another clinical implication is that intervention approaches should build on existing knowledge about phonological skills but also include a broader approach, with more lexical-semantic-specific training models, built on theories of typical development and with a specified but yet dynamic approach.

In summary, we can now have higher expectations on the ability of the new generation of early implanted children with CI to develop age-equivalent lexical-semantic ability. In addition, some children with higher ages at 1st CI are also able to catch up, at least to some extent. However, there are still children that need extra support and specific training to catch up with NH peers in both groups (younger and older implanted children). In addition, all children with CI need a conscious and supporting environment for developing optimal lexical-semantic ability. Some “white and red flags” have been identified in the sample that has been studied.

White flags Depth and breadth in vocabulary and specifically semantic capacity were associated to better vocabulary outcomes. Therefore, caregivers should provide opportunities for children to experience a rich spoken language and in meaningful situations, previously found to have an impact on NH children (Roberts and Kaiser, 2011) and children with HI (Quittner et al., 2013). Not only should they provide children with words, but also reflect in conversation of concepts in more depth and with all senses and in particular by listening and talking experiences. Children who used more expressive spoken language as toddlers had better linguistic outcome later in childhood. This indicates the importance of early language experiences. Still, there is need for more intervention studies for evaluating “which is the egg and which is the hen” in this case. Nevertheless, there is evidence that early family-based training have an impact on children’s later outcome in NH children and other groups, including children with HI (Aragon and Yoshinaga-Itano, 2012; Leffel and Suskind, 2013). Therefore, early dyad based options should be in favour for promoting phonology, grammar and lexical-semantic ability. Considering that families and children differ, including their needs and desires, it is necessary to offer different and optional intervention alternatives. Examples of such existing EI programs are AVT or individual-based programs including visual support with LENA (Percy-Smith et al., 2012; Aragon and Yoshinaga-Itano, 2012). One factor that should be further evaluated is the specific influence of *parents*. The aim of these mentioned EI alternatives is primarily to enhance parent awareness and parent empowerment. This will influence indirectly on the child’s opportunities to experience language, which seems to be an important factor for the expressive level as well as for the receptive level. These mentioned white flags could easily be adjusted as goals and specific targets in intervention work with families, and in close cooperation with caregivers. Prevention from early ages should be of outmost priority instead of later adjustment of already established atypical language patterns, which is more difficult to master.

Red flags Non-verbal cognitive ability and semantic capacity predicted grammatical sentence understanding in children aged 6-9 years, in both groups (CI and NH). Therefore, children with less efficient non-verbal cognitive ability and e.g. poor expressive vocabulary at school entry are at risk for worse lexical-semantic ability in early school years. Evaluation of children with CI should start early and always include a longitudinal approach, including cognitive assessments for providing the best possible individual support. Another red flag was less developed expressive spoken language in early childhood, also seen in other groups. The two rough measurements used in the study; speech intelligibility and expressive grammar level, are interpreted as indicators of early poor linguistic development of spoken language aspect like phonology and morphology, affecting lexical-semantic ability. Children with no or poor canonical babbling despite full-time of HA and/or CI use could also be a potential red flag (Oller et al., 1999). This needs to be confirmed in prospective studies with other assessment tools that investigate canonical babbling and other expressive language skills when the child gets older, like phonology and morphology, but also lexical-semantic knowledge. The influence of parents and other caregivers should be evaluated simultaneously, to gain more knowledge of the impact of environmental factors.

5.3 FUTURE PERSPECTIVES

Lexical-semantic organisation and picture naming ability has previously not been studied to a higher degree in the population. The results from this thesis show that semantic capacity at school entry is an important factor for later word knowledge. Therefore, more emphasis should be made on lexical-semantic ability in future studies of children with CI. There is a need of further studies on how children with CI learn and use words, both from a lexical and semantic perspective, preferably with a longitudinal approach and perspective. There is a clear advantage to studying lexical-semantic ability by using a variety and broad spectra of different tasks, as it seems that children with CI have atypical patterns, even if they may catch up over time.

In the relatively small sample of children with CI there were some interesting subgroup differences largely explained by different mean age at 1st implantation but also possibly a generation shift, presumably depending on general changes in perspectives, intervention options and school choices in Sweden. Given that the ages at implantation have fallen further since the children in this thesis were operated on, one should be cautious to compare both the results of earlier studies and of similar future studies with the results from this sample. NH control groups should therefore always be included in the study designs of studies conducted in the population. Single-subject design could be a good alternative for evaluating intervention options.

Considering the multidimensional process of learning words in early childhood a dyad approach should be adopted with focus both on the child and the caregiver, as it seems to be more of a mutual experience that promotes word learning in children with HI as compared with children with NH, who to a higher degree typically learn words seemingly effortless.

Future studies of lexical-semantic ability should include subgroups with known additional needs like children with bilingual background or additional diagnoses like LI

and ASD. The current thesis-work indicated that error analysis might be an efficient way of examining and identifying phenotypes within the clinical group.

5.4 SWEDISH SUMMARY

Bakgrund: Det föds ca 20 barn per år i Sverige med medfödd måttlig-grav hörselnedsättning och ungefär 10 barn per år blir döva på grund av progredierande hörselnedsättning eller någon förvärvad orsak som t.ex. meningit. Tack vare nyföddhetsscreening kan man idag upptäcka medfödd hörselnedsättning/dövhet redan på BB, vilket innebär att hörapparater kan provas ut redan när barnet är ett par månader gammalt. För de barn som inte hör tillräckligt bra med hörapparater finns möjligheten att operera in cochleaimplantat (CI). Cochleaimplantat är ett tekniskt hörhjälpmedel som via elektrisk stimulering av hörselnerven ger en ny, konstgjord hörsel. Somliga barn har ett CI och en hörapparat på andra örat, exempelvis om barnet delvis är hjälpt av hörapparat för att kunna höra och utveckla talspråk. De flesta svenska döva barn har dock bilaterala CI och ett fåtal använder bara ett CI, då p.g.a. medicinsk-tekniska orsaker. Omkring 500 gravt hörselskadade barn har opererats och fått CI på Karolinska Universitetssjukhuset sedan 1990 (maj, 2014). Det finns en stor variation i talspråksutvecklingen hos barn med CI, som är en liten och heterogen population. Viss forskning tyder på att barn med CI har brister i sitt ordförråd (Kenett et al., 2013; Fagan & Pisoni, 2010). Hittills har få studier undersökt lexikal-semantisk förmåga hos barn med CI och i relation till kognition samt i jämförelse med åldersmatchade barn som har normal hörsel.

Syftet var att undersöka ordförråds- och begreppsbildning hos barn med CI och i jämförelse med typiskt utvecklade barn med normal hörsel i samma ålder och delvis med andra kliniska grupper: barn med generell språkstörning och barn med autismspektrumstörning.

En hypotes innan projektstart var att barnen med CI skulle ha ett mindre och torftigare ordförråd.

Metod: 6-9 år gamla barn med CI (n=34) undersöktes i jämförelse med åldersmatchade barn med normal hörsel (NH) (n=39) och delvis med två andra kliniska grupper: barn med generell språkstörning (n=12) samt barn med autismspektrumstörning (n=12). Projektet bestod av fyra separata delstudier. I den första delstudien undersöktes förmågan att hämta ord från långtidsminnet utifrån två givna kategorier; fonologiskt baserat bokstavsflöde (FAS) samt semantiskt baserat ordflöde (djur) i gruppen barn med CI och barn med NH. Lexikal-semantisk förmåga undersöktes därefter i samma grupper (CI och NH) och i jämförelse med en grupp barn med språkstörning eller autismspektrumstörning (studie II). I den tredje delstudien undersöktes grammatisk meningsförståelse i relation till aspekter som inläring, icke-verbal förmåga samt kunskap om enskilda ord, hos barn med CI och normalhörande barn i åldern 6-9 år. Den fjärde delstudien var en uppföljningsstudie där lexikal-semantisk förmåga undersöktes i de två grupperna (CI och NH) samt med en longitudinell inriktning för 18 barn med CI som hade lägre medelålder vid sin första CI-operation än annan grupp barn med CI inom kohorten.

Många barn med CI uppnådde åldersadekvata lexikala- semantiska förmågor vid en ålder av 8-9 år. Semantisk kunskap och icke - verbal kognitiv förmåga förutspådde grammatisk meningsförståelse (GSU) i båda grupperna (CI och NH). Barn med CI använde samma sorts inlärningsstrategier som barn med normal hörsel och hade liknande kognitiva kapacitet som krävs för att hantera användningen av och kunskapen om ord. Barn med CI visade bättre resultat än barn med språkstörning eller autismspektrumstörning. I hela gruppen barn med CI fanns ett atypiskt utvecklingsmönster i studie II där barn med CI hade bättre expressivt and receptivt ordförråd jämfört med typiskt utvecklade barn med NH. Även variationen i resultat var totalt sett högre hos barn med CI (studie I - IV). En åldersrelaterad CI-gruppskillnad hittades i studie I vilket ledde fram till planering av en uppföljande studie, där också tidig talspråksförmåga hämtades från journaldata för statistiska analyser (studie IV). Barn med CI som var äldre vid operation med sitt första CI hade signifikant sämre resultat på receptivt ordförråd, ordflöde (bokstavsljud) och på grammatisk meningsförståelse jämfört med NH barn. Trots detta resultat hade alla barn med CI på gruppnivå åldersadekvat expressivt ordförråd vid 8-9 års ålder. Semantisk kunskap demonstrerades som en relevant förmåga att känna igen semantiska särdrag och att använda semantiskt relevanta svar då de saknade lexikala benämningar i samband med bildbenämning. Barn i gruppen med en yngre medelålder vid första CI-operation hade åldersadekvat receptivt ordförråd när de var 8-9 år samt åldersadekvat bokstavsflöde. Dessa resultat pekar på att barn med CI, med typisk utveckling, generellt sett inte har svårt att lära sig ord, men att det samtidigt är en stor variation i gruppen och att en del av förklaringen till olika subgruppresultat inom gruppen barn med CI kan vara yngre ålder vid CI men att andra mer lingvistiska och kognitiva förmågor har betydelse för en gynnsam ordförrådsutveckling.

Slutsatser: Barn med CI i åldrarna 6-9 år och typisk icke-verbal förmåga hade inte specifika svårigheter att processa, lagra och generera ord. De uppvisade semantisk förståelse och kunskap om ord de inte kunde benämna och hade god särdragskunskap men saknade i högre grad lexikala benämningar till begrepp, framförallt de som hade fått sina första CI senare. Variationen var dock stor, både hos de som fått sitt CI tidigt och de som opererats något senare. Därför är det viktigt att noga följa alla barns utvecklingsmönster individuellt över tid, både vad gäller lyssnande, talspråk och kognition. Tidig CI-intervention förordas tillsammans med individanpassad föräldrahandledning under barnets första levnadsår, med i första hand en preventiv inriktning. Tidig expressiv talspråksförmåga efter två år med CI samt expressiv ordförrådsförmåga och semantisk särdragskunskap innan skolstart tycktes vara associerat med bättre lexikal-semantisk förmåga två år efter skolstart. Framtida studier av lexikal - semantiska förmågor hos barn med CI bör ha en longitudinell inriktning och även inkludera mått miljöfaktorer som t ex föräldrars typ och grad av talspråksstimulans.

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