Children with Cochlear Implants

Cognition and Reading Ability

Malin Wass

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Abstract

The present thesis investigated cognitive ability in children with severe to profound hearing impairment who have received cochlear implants (CIs). The auditory stimulation from a cochlear implant early in life influences most cognitive functions as a consequence of the plasticity of the brain in the young child. It is important to understand the cognitive consequences of auditory stimulation from CIs in order to provide adequate support to these children. This thesis examined three specific aspects of cognitive ability (working memory, phonological skill and lexical access), and reading ability in children with CIs, as compared to children with normal hearing in the same age. The relations between cognitive abilities and reading skills were also investigated, as well as the associations between demographic variables (e.g., age at implantation and communication mode), cognitive abilities and reading skills. The children with CI generally had lower performance levels than the normal hearing children in tasks of phonological and general working memory, phonological skills and lexical access. They had specific problems in tasks with high demands on phonological working memory, whereas their performance levels in tasks of visuospatial working memory were on par with the hearing children. A majority of the children with CI demonstrated reading skills within the normal range for hearing children, both for decoding and reading comprehension. The relations between demographic factors and cognitive skills varied somewhat between the studies. The patterns of result are discussed with reference to contemporary theories of working memory, phonological skills, and lexical access.

Contents

List of papers

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

I. Wass, M., Lyxell, B., Sahlén, B., Asker-Árnason, L., Ibertsson, T., Hällgren, M., Larsby, B., & Mäki-Torkko, E. In search of cognitive correlates of orthographic learning: Is there a relation between orthographic learning and working memory? (manuscript).

II. Wass, M., Ibertsson, T., Lyxell, B., Sahlén, B., Hällgren, M., Larsby, B., & Mäki-Torkko, E. (2008). Cognitive and linguistic skills in Swedish children with cochlear implants – measures of accuracy and latency as indicators of development. *Scandinavian Journal of Psychology*, *49*, 559–576.

III. Asker-Árnason, L., Wass, M., Ibertsson, I., Lyxell, B., & Sahlén, B. (2007).The relationship between reading comprehension, working memory and language in children with cochlear implants, *Acta Neuropsychologica, 5(4),* 163–187.

IV. Wass, M., Lyxell, B., Sahlén, B., Asker-Árnason, L., Ibertsson, T., Mäki-Torkko, E., Hällgren, M., & Larsby, B. Reading Strategies and Cognitive Skills in Children with Cochlear Implants. (manuscript).

Introduction

The present thesis deals with cognitive ability in children with severe to profound hearing impairments (HI) who have received cochlear implants (CIs). A CI is a technical device which enables persons with severe or profound HI to perceive sound and speech. Candidates for cochlear implantation are children who do not benefit enough from conventional hearing aids to develop oral language. Today, it is estimated that about 98% of the children in Sweden who have a degree of hearing impairment where they would benefit from implantation, receive CIs (Socialstyrelsen, 2009). The use of a CI does not restore hearing to a normal level but enables a different course of development of speech and language function, than would have been possible without the CI (Geers, 2003; Houston, Pisoni, Iler Kirk,Ying & Miyamoto, 2003; Richter, Eißele, Laszig & Löhle, 2002; Spencer, 2004).

The auditory stimulation from a cochlear implant early in life should be expected to influence most cognitive functions as a consequence of the plasticity of the brain in the young child (Kral & Eggermont, 2007; Pisoni et al., 2008; Sharma, Nash, & Dorman, 2009). It is important to understand the cognitive consequences of receiving a cochlear implant, in order to be able to provide adequate support to children who use CIs in various settings. The present thesis is focused on three specific aspects of cognitive ability: working memory, phonological skill and lexical access.

The outline of the thesis

This thesis starts with a brief overview of hearing impairments, including definitions, etiology, cochlear implant functioning and candidacy. The concept of plasticity is also discussed as well as demographic factors and rehabilitation. In the second chapter, research on working memory, phonological skills and lexical skills in children with CI and other populations is reviewed. The third chapter is focused on methodological issues in research on children with CI in Sweden. Following a brief summary on the papers, I-IV, the findings on each of the cognitive skills, working memory, phonological skills, and lexical access are discussed, as well as the results on reading ability. Finally, a few ideas for future research are suggested.

Hearing impairments

Degree and prevalence

Hearing impairments are categorized according to degree of hearing loss based on better ear hearing level averaged over the frequencies 0.5, 1, 2, and 4 kHz ($BEHL_{0.5\text{-}4kHz}$). Over 20 dB HL or less than 40 dB HL is categorized as *mild* HI, over 40 and up to 70 dB HL is *moderate*, over 70 but less than 95 dB HL is *severe*, and 95 dB HL or more is classified as *profound* HI (Stephens, 1996). Persons with a hearing loss of 95 dB HL or more are commonly referred to as *deaf*. The term *Deaf* on the other hand, refers to persons who are members of the Deaf - World (Lane, 2005), i.e., a minority culture of people who use sign language as their primary language and do not consider their hearing loss to be a disability. In order to avoid confusion, the term profound hearing impairment will be used in this thesis to denote hearing loss of more than 95 dB HL.

Since screening for hearing impairment in newborns was introduced in Sweden recently (Socialstyrelsen, 2009), it is possible to identify children with congenital HI at an early age. Before screening of newborns was common practice in Sweden, only about 60% of the children with profound HI were identified before 18 months of age (Socialstyrelsen, 2009).

The number of children with profound HI has been estimated in a number of studies in order to project the number of CI candidates (Blanchfield, Feldman, Dunbar, & Gardner, 2001; Bradham & Jones, 2008; Davis, Fortnum, & O'Donoghue, 1995; Van Naarden, Decouflé, & Caldwell, 1999). Davis et al. (1995) assumed a prevalence of 0.37/1000 children for hearing impairments of 95 dB HL or worse in the UK for children, 3–9 years of age. American studies have reported prevalence rates of between 0.5/1000 children (Blanchfield et al., 2001; Bradham & Jones, 2008) and 1.1/1000 (Bradham & Jones, 2008; Van Naarden et al., 1999). A Finnish study (Mäki-Torkko et al., 1998) reported a prevalence of 1.2/1000 live births for HIs of 40dB HL or more. There are no exact figures on the number of implant candidates in Sweden. An extrapolation of American studies, however, suggests that a little less than 1 child per 1000 would benefit from implantation (Socialstyrelsen, 2009).

Cochlear implant candidacy

When a child is diagnosed with a permanent bilateral HI, he/she is always fitted with conventional hearing aids. Cochlear implantation is considered for children with severe to profound HI, for whom hearing on the best ear is not sufficiently restored by optimally fitted traditional hearing aids to enable oral speech and language development (Gravel & O'Gara, 2003). These decisions are made by the cochlear implant teams based on clinical experience and repeated evaluation of hearing and communication development. The cause of HI needs to be located in the inner ear in order for CIs to be effective (Socialstyrelsen, 2009).

Categorization according to onset of hearing impairment

Hearing impairments in children can be categorized according to various criteria, for example the time of onset of hearing impairment, i.e. *congenital* (from birth) or later acquired hearing impairments. Sometimes, a categorization into *prelingual* and *postlingual* HIs is made, on the basis of the child's level of (expressive) language development at the onset of HI. According to clinical experience, these categories refer to onset before or after 2 years of age (Arlinger, 2007, p.247). The term *perilingual* HI may be used when onset of HI is in the most intense period of language development (around the age of 2). About 80% of the children with severe to profound HI who would benefit from a cochlear implant, in order to develop spoken language, have a congenital hearing loss (Fortnum, Marshall, & Summerfield, 2002).

Causes of hearing impairment

At the physiological level, damage in the inner ear, *cochlear HI*, commonly caused by injured hair cells**,** is the most frequent cause of permanent HI in children (Arlinger, 2007; Socialstyrelsen, 2009). The etiological cause of HIs in children (better ear hearing level >=40 dB HL) is known in approximately 60% of the cases (Fortnum & Davis, 1997). Out of the total number of children with profound HI in Fortnum & Davis (1997), about 40% were reported to have at least one additional disability. Similar patterns of epidemiology are to be expected in Sweden. About 50% of all HIs are genetic. Thirty percent of those are syndromal and 70% are non-syndromal (Van Camp & Read, 2003).

Postnatal/acquired HIs are often caused by various infections, where meningitis is the most common cause of HI. Head trauma and tumours may also result in severe to profound HI (Davis et al., 1995).

Cochlear implants - a description

Cochlear implants consist of an external and an internal part (Figure 1). An external speech processor with a microphone (1) picks up and recodes sound into digital signals. The processor transmits the signals to a surgically implanted receiver (2). The receiver transmits electrical signals to the electrode array, which is inserted into the cochlea (3). The number of active electrodes in this array commonly varies between 8 and 24 and different electrodes are stimulated depending on the frequency of the signal. The active electrodes stimulate the nerve cells in the auditory nerve (4) and the signals are further transmitted to the brain where they may be perceived and interpreted (Arlinger, 2007).

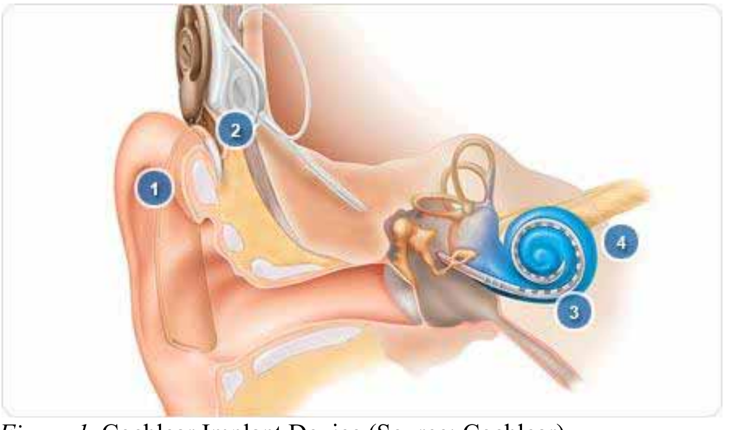

Figure 1. Cochlear Implant Device (Source: Cochlear).

Hearing with cochlear implants

The children receive auditory input from their CI but their auditory perception (the sensory stimulation) is not restored to normal level. This is, in part, because the electrically transmitted signals from the CI do not possess the same fine acoustic-phonetic details of the speech-signal as in acoustic hearing. Thereby, the brain receives a degraded signal which, in turn, may affect the development of long-term representations of spoken words and speech sounds in long-term memory (Harnsberger et al., 2001; Pisoni et al., 2008). The exact nature of auditory stimulation with a cochlear implant is, however, not possible to predict or describe since it is dependent on a number of factors, such as the auditory representations which were established before implantation, and how these representations are transformed due to plasticity in the brain (Sharma, Dorman, & Kral, 2005; Sharma et al., 2009). This topic will be further discussed below.

Brain plasticity and age at implantation

In general, development of the central nervous system is dependent on the interplay of two sources of information, genetic factors and input from the external world (Bischof, 2007). Input from the world around is mediated by the sense organs. Early sensory input affects the electrophysiological properties of single neurons in the nervous system. These changes can only occur within a limited period of time, which is referred to as a *sensitive period* (see Bischof, 2007, for a review). The developing brain has various sensitive periods during which information from the external world is used to modify and complete its neuronal networks. There are indications that the auditory system has a sensitive period of 3.5 years during which time the central auditory pathways are considered to be maximally plastic and primed for stimulation-driven development (Sharma et al., 2005; Sharma et al., 2009). Higher order cortices have been found to be cross-modally reorganized in deaf participants who have been deprived of auditory input for extended periods of time (Sharma et al., 2005; Sharma et al., 2009; see also Neville & Bavelier, 1998, for results on cortical reorganization in adults as a consequence of sensory deprivation). Similarly, length of auditory deprivation before implantation has been proved to affect brain activity in response to auditory stimulation from cochlear implants in children, implanted at different ages (Sharma et al., 2005). Cochlear implantation within the sensitive period therefore should provide optimal implant benefit.

Children implanted before 3.5 years of age have been found to have similar cortical responses to auditory stimulation as children with normal hearing (NH), although the exact patterns of activation may vary as a function of hearing impairment and length of auditory deprivation (Sharma et al., 2009). Children implanted between 3.5 and 7 years of age have demonstrated variable cortical responses to auditory stimulation, and children implanted after seven years of age show activation outside the auditory cortical areas (Sharma, Dorman et al., 2005; Sharma, Nash et al., 2009). After seven years of age, which is regarded as the end of the sensitive period for the auditory system, the primary auditory cortical areas are likely to be de-coupled from surrounding higher order cortices. Secondary auditory areas may thereafter, to some extent, re-organize to serve other modalities, such as vision and somatosensory functions (Sharma et al., 2009).

In addition to the sensitive period for central auditory development, language development in children with NH is in its most intensive phases during the first 4 years of life (Hoff, 2009). Even before birth, fetuses can respond to external sound and discriminate their mother's voice from other women's voices (Hoff, 2009). The first year of life is a very important period of language development, as children go through many important stages of interaction with their environment by means of, for example, discriminating between voices and sounds, crying, babbling, recognizing and producing the phonemes and first words of their maternal language (Arlinger, 2007; Hoff, 2009). For example, infants recognize their parents' voices within a few days after they are born (Hoff, 2009). Within the first months of life infants may generally discriminate between phonetic contrasts of any language, even those languages they have not heard (Kuhl, Williams, Lacerda, Stevens & Lindblom, 1992; Werker & Tees, 1999). By 6 months of age, their ability to discriminate between different phonemes has been found to be altered by their native language (Kuhl et al., 1992; Werker & Tees, 1999). That is, they may no longer be able to discriminate between phonetic contrasts that are not used in their native language. Most children have acquired basic language skills by 4 years of age (Hoff, 2009) and rich language stimulation during these years is crucial for optimal language development. Therefore, it is extremely important to start the auditory rehabilitation process, for example hearing aid fitting, as early as possible, and by 6 months at the latest (Socialstyrelsen, 2009), in order to maximally utilize the sensitive period for auditory and language development in children with severe or profound hearing impairment. Implantation before the congenitally deaf child is one year old is thus recommended and has become increasingly common in Sweden since 2005 (Socialstyrelsen, 2009).

In sum, in order to utilize the sensitive period for auditory development and the most intensive period of language development, the rehabilitation process should start as early as possible for children with a severe or profound HI. Those children who would benefit from a CI should be implanted as early as possible, when it is safe to perform surgery from an anesthesiological point of view (commonly from the age of 6–8 months).

Frequency of cochlear implant patients

The first cochlear implants with multiple electrode channels were surgically inserted in 1978 in adults. Today, more than 100 000 people worldwide have received a cochlear implant (Socialstyrelsen, 2009). Sweden has the comparatively highest ratio of implantations in relation to population size (Socialstyrelsen, 2009). By the end of 2008, there were approximately 1600 CI-users in Sweden. Almost 600 of those were children and 263 of the children were bilaterally implanted (E. M. Mäki-Torkko, personal communication, October 13, 2009). About 55 children per year in Sweden are considered for cochlear implantation and today most of them receive bilateral implants (Socialstyrelsen, 2009).

So far, the long term benefits of bilateral implantation in children has not been extensively investigated but a growing number of research studies report improved sound localization (Litovsky, Johnstone, & Godar, 2006) and better speech recognition in quiet and in noisy conditions (Scherf et al., 2007). The cognitive consequences of bilateral implantation have hitherto not been addressed.

Rehabilitation

Fitting of traditional hearing aids before the child is 6 months of age, at the very latest, is one of the measures that need to be taken to maximize auditory stimulation and allow the child to perceive at least some aspects of the ambient oral language (Gravel & O'Gara, 2003). The goal of the rehabilitation processes for implanted children is generally that they should have acquired age appropriate, or near age appropriate, oral language by the age they enter formal education (Specialskolemyndigheten, 2005). After cochlear implantation, the children and their families attend regular follow-up appointments at their cochlear implant programs where individual support is provided by technicians, pediatric audiologists, speech and language pathologists, and special education teachers. There are many issues to discuss about rehabilitation in children with CI. This topic is, however, outside the scope of this thesis.

Communication modes and educational settings

According to an American study by Gravel & O'Gara (2003), the communication modes that children with CIs may use can be considered to vary along a continuum from auditory/spoken language to purely visual sign language, with a number of different combinations of auditory/visual communication in between. Similar variations in communication mode are found in Swedish children with CI. Furthermore, sign language is offered to all children with severe to profound HI born in Sweden and their families (Ibertsson, 2009).

Children who use CIs in Sweden may choose between a number of different educational settings. Some children are individually integrated in mainstream education for hearing children. Others attend classes for children with HI in mainstream schools. They may also attend special schools for children with HI where they are instructed in oral language. Other options are special schools for children with severe to profound hearing impairments where education may be in sign language or oral language with signed support, depending on the linguistic and communicative abilities of the child (Ibertsson, 2009).

Effects of demographic factors on the development in children with cochlear implants

A number of variables associated with the child and with the implant may explain varying proportions of variance in outcome after implantation (Geers, Tobey, Moog, & Brenner, 2008; Stacey, Fortnum, Barton, & Summerfield, 2006). For example, the use of oral communication mode at home and in the educational setting has been found to be associated with larger capacity to remember phonological information for a short period of time (Pisoni & Cleary, 2003; Schorr, Roth, & Fox, 2008) and with higher educational achievements compared to other communication modes (Geers et al., 2008; Stacey et al., 2006). This relationship may be causal (Geers et al., 2008) but, as suggested by Stacey et al. (2006), it may also be that the choice of communication mode in education is influenced by the child's performance level before entering education. Younger age at implantation and longer periods of implant use have a positive effect on implant benefit which is manifested in, for example, spoken word recognition (Geers et al., 2008; Geers, Moog, Biedenstein, Brenner, & Hayes, 2009; Holt, Svirsky, Neuburger, & Miyamoto, 2004; Nicholas & Geers, 2006; Pisoni et al., 2008), various aspects of preverbal (Houston, Ying, Pisoni,& Iler Kirk, 2003;Tait, Nikolopoulos & Lutman, 2007) and verbal language development (Geers et al., 2008; Pisoni et al., 2008; Schorr et al., 2008; Tomblin, Barker, Spencer, Zhang, & Gantz, 2005), e.g., receptive vocabulary (James, Rajput, Brinton, & Goswami, 2008; Schorr et al., 2008), and reading skill (Geers et al., 2008; Marschark, Rhoten, & Fabich, 2007; McDonald Connor & Zwolan, 2004). Other factors which are evidenced to have an effect on the outcome after implantation are implant technology factors, such as processing strategy of the speech processor in the implant (Geers, Brenner, & Davidson, 2003; Manrique et al., 2005), and number of active electrodes in the implant (Friesen et al., 2009; Geers et al., 2003). A larger number of active electrodes provide a better frequency resolution and thereby better speech recognition (Friesen et al., 2009). The above-mentioned variables are only some examples of demographic factors which have been found to influence implant benefit in children.

Cognitive functions

Cognitive psychology refers to the study of mental processes such as perception, memory, and attention (Eysenck & Keane, 2005; Sternberg, 2006). These mental processes are essential for our ability to perceive and attend to the world around us. They are also fundamental for our thinking and problem solving. The auditory stimulation from a cochlear implant early in life should be expected to influence most cognitive functions as a consequence of the plasticity in the brain of the young child (Kral & Eggermont, 2007; Pisoni et al., 2008; Sharma et al., 2009). Thus, in order to fully understand the cognitive consequences of receiving a cochlear implant, research has to be performed in all of the areas of cognitive ability. The focus of this thesis was to study three specific aspects of cognitive ability, namely working memory, phonological skills and lexical access. The rationale for studying these skills is that they have proved to be fundamental to a number of more complex cognitive skills such as reading ability, arithmetic skills and various aspects of communication in children with NH. For example, these cognitive skills have proved to be affected in children with various problems, for example ADHD (Rapport et al., 2008; Willcutt, Doyle, Nigg, Farone, & Pennington, 2005), dyslexia (Bishop, Adams, & Frazier Norbury, 2004; Griffiths & Snowling, 2002; Roodendrys & Stokes, 2001), and specific language impairment, SLI (Archibald & Gathercole, 2006; Gathercole & Alloway, 2006), which will be further discussed below. These three abilities are particularly important to study in children with CI, since children with severe to profound HI who have not been implanted, have demonstrated poor levels of reading skills (Paul, 2003) as well as phonological skills in oral language (Blamey, 2003). It is thus of critical importance to find indications of how the auditory stimulation from CIs may influence cognitive skills and reading skills in children.

Working Memory

Working memory (WM) refers to the limited capacity to store and process new information for a short period of time (Baddeley & Hitch, 1974; Baddeley, 2009; Daneman & Carpenter, 1980; Daneman & Hannon, 2007; Miyake & Shah, 1999; Repovš & Baddeley, 2006). This ability is crucial in various situations in everyday living, for example in remembering a telephone number while looking for paper or pencil to write it down, or in keeping a French phrase in memory while figuring out what it means. Various types of information, e.g., phonological, visual, and spatial are stored and processed within working memory (Baddeley, 2003; Repovš & Baddeley, 2006).

The theories that have been most influential for our understanding of working memory can be categorized in various ways. One set of theories regard both the storage and processing components of dual tasks (i.e., storage and prcessing) to be *domain specific* (e.g. Andersson & Lyxell, 2007; Daneman & Hannon, 2007; Miyake, Friedman, Rettinger, Shah, & Hegarty, 2001). According to this view, two independent resources are responsible for the processing and storage of phonological and visuospatial information, respectively. Another set of theories (Baddeley, 2003; Engle, Kane & Tuholski, 1999; Repovš & Baddeley, 2006) claim that working memory has a domain-general component (executive) responsible for processing of information in dual tasks and domain-specific subcomponents dedicated to short-term storage of information.

Further distinctions can be made according to how the working memory resources are distributed between storage and processing of information. The *resource-sharing hypothesis* (Daneman & Carpenter, 1980; Daneman & Hannon, 2007) assumes that the same mechanism within working memory is responsible for both storage and processing of information and that there is a trade-off between processing and storage demands. Thus, if the processing component of a working memory task is very demanding, only a small amount of working memory capacity is left over for storage and vice versa. In contrast, the task-switching hypothesis (Towse, Hitch, & Hutton, 1998; 2000) considers tasks which require simultaneous storage and processing to be dependent on the ability to switch attention back and forth between the tasks of storing and processing information (Towse, Hitch & Hutton, 1998; 2000; Saito & Miyake, 2004). This hypothesis further assumes that processing and storage are independent in that they are not dependent on the same mechanism (Saito & Miyake, 2004).

The domain-general account of WM may be represented by the multi-component model (Baddeley, 2003; 2009; Repovš & Baddeley, 2006). This model assumes that WM includes four components, each serving a specific purpose. A central executive serves as a modality-independent control system, which directs and divides attention between tasks. It is involved whenever manipulation of information within working memory is required (Repovš & Baddeley, 2006). The central executive, thereby, controls the function of the subsidiary storage components, i.e., the phonological loop, the visuospatial sketchpad and the episodic buffer (Baddeley, 2003; 2009; Repovš & Baddeley, 2006). The functioning of the phonological loop is referred to here as phonological working memory. It is represented by a phonological store, which can hold phonological content for a few seconds before it fades, and an articulatory rehearsal process where phonological information may be rehearsed in order to refresh the memory trace (Baddeley, 2003; 2009; Repovš & Baddeley, 2006). The phonological loop has a limited capacity, such that when the number of items to be rehearsed in the articulatory rehearsal process increases, at a certain point, the words may have faded before they can be rehearsed.

The visuospatial sketchpad is dedicated to the storage and manipulation of visual and spatial information (Baddeley, 2003; 2009; Repovš & Baddeley, 2006). Recent results suggest that this component of working memory can be further divided into separate subsystems for visual- and spatial information, respectively, where each subsystem has its own mechanisms for storage, maintenance and manipulation (Repovš & Baddeley, 2006).

In the present work, visual and spatial aspects of visuospatial working memory are not treated as separate abilities. In the context of this thesis, the term visuospatial WM thus refers to temporary storage and manipulation of both visual and spatial information.

The episodic buffer, hitherto relatively unexplored (see, however, Baddeley, Hitch, & Allen, 2009; Rudner, Fransson, Ingvar, Nyberg, & Rönnberg, 2007) is assumed to be a temporary store, where information from the multiple sensory modalities and from long-term memory may be integrated (Baddeley, 2000; 2003; 2009; Repovš & Baddeley, 2006). The integration of information within the buffer is assumed to occur through binding of information from different sources, which results in multi-dimensionally coded information (Allen, Baddeley & Hitch, 2006; Baddeley, 2000). Thereby the episodic buffer may be regarded as an interface between the subcomponents of WM, perception and long-term memory (Baddeley, 2000; Repovš & Baddeley, 2006).

In this thesis, the term general working memory refers to the simultaneous storage and processing of information, as defined either by the functional perspective of working memory (e.g., Hannon & Daneman, 2001a; 2001b; 2007; Miyake, 2001; Miyake et al., 2001), or by the concurrent involvement of the central executive and a subsidiary storage component, according to the multi-component view (Baddeley, 2003; Repovš & Baddeley, 2006). General working memory is therefore measured in dual-tasks, taxing both the executive control function and storage. In relation to general working memory, the theoretically older term *short-term memory* is sometimes used with reference to tasks which require temporary storage but not significant concurrent processing of information (cf. Archibald & Gathercole, 2006; Miyake et al., 2001). In the present thesis, tasks with demands on storage but minimal processing requirements are referred to as visuospatial or phonological working memory tasks, depending on the storage component and the type of stimulus modality involved.

Phonological working memory

In children with typical development, phonological working memory is important for vocabulary learning, both in the native language and in foreign languages (Baddeley, 2003; 2009; Gathercole, 2006a; Repovš & Baddeley, 2006). The key feature in this relationship is that the storage of an unfamiliar phonological representation in working memory mediates the establishment of this particular phonological representation in long-term memory (Archibald & Gathercole, 2006; Gathercole, 2006a). High phonological working memory capacity is thus intimately associated with better word learning skills. Phonological working memory also influences the development of reading skills (e.g., Goff, Pratt, & Ong, 2005; Nikolopoulos, Goulandris, Hulme, & Snowling, 2006). This relationship has frequently been regarded as secondary to the effects of working memory on other variables known to influence reading ability, e.g. new word learning and verbal language learning ability (Baddeley, 2003; Gathercole, 2006a; 2006b; Majerus, Poncelet, Greffe, & Van der Linden, 2006), letter learning (Torppa, Poikkeus, Laakso, Eklund, & Lyytinen, 2006), phonological skills and the quality of phonological representations (Durand, Hulme, Larkin, & Snowling, 2005; Gathercole, Alloway, Willis, & Adams, 2005; van Daal, Verhoeven, van Leeuwe, & van Balkom, 2008).

Impaired phonological working memory function is suggested to be one of the key factors for the language problems in children with SLI (Archibald & Gathercole, 2006; Baddeley, 2003; Gathercole & Baddeley,1990; Gathercole, 2006a; Marton & Schwartz, 2003). Phonological working memory predicts reading ability in children with reading disorders (Bishop et al., 2004; Griffiths & Snowling, 2002; Roodendrys & Stokes, 2001). Specific deficits in phonological working memory capacity are characteristic of children with Down's syndrome, in that their deficit in working memory capacity is more severe than their general language problems (Gathercole & Alloway, 2006). Deficits in phonological working memory capacity have also been reported in children with ADHD (Rapport et al., 2008; Willcutt, Doyle, Nigg, Farone, & Pennington, 2005). Recent studies report that children with CI have a more limited phonological WM capacity than children with NH (Dawson, Busby, McKay, & Clark, 2002; Pisoni et al., 2008; Spencer & Tomblin, 2009). Phonological WM, however, also plays an important role for cognitive functioning in children with CI. For example, phonological working memory has proved to be important for novel word learning (Willstedt-Svensson, Löfqvist, Almqvist & Sahlén, 2004), word recognition, and vocabulary development (Cleary, Pisoni, & Iler Kirk, 2000), composite measures of grammar and lexicon (Dawson et al., 2002), and reading skills (Dillon & Pisoni, 2006). Phonological WM capacity has also proved to be related to the type of communication mode mainly used by the child at home and at school, i.e., oral communication is associated with higher phonological WM capacity, as measured by forward digit span (Pisoni, & Cleary, 2003) as well as age at implantation (Schorr et al., 2008).

General working memory

General WM capacity has been found to predict reading ability at various levels in children with typical development (e.g. Baddeley, 2003; Cain, Oakhill, & Bryant, 2004; Gathercole et al., 2005; Goff, Pratt & Ong, 2005; Savage, Cornish, Manly, & Hollis, 2006). In these children, general working memory capacity predicts unique variance in areas important for most academic domains, e.g., language comprehension (Miyake, 2001), reading comprehension (Cain, Oakhill, & Bryant, 2004; Seigneuric, Ehrlich, Oakhill, & Yuill, 2000; Swanson & Ashbaker, 2000), arithmetic skills (DeStefano & LeFevre, 2004; Rasmussen & Bisanz, 2005), and reasoning and problem solving (Baddeley, 2003; Miyake, 2001). General WM capacity is also significantly related to severity of reading problems in children with reading disabilities (Gathercole et al., 2005) and in children with attention problems (Savage et al., 2006). Children with mathematical difficulties have been found to have specific problems with concurrent storage and processing of numerical and visual information (Andersson & Lyxell, 2007). Substantial impairments in tasks that require simultaneous processing and storage of information are also characteristic of children with SLI (Archibald & Gathercole, 2006; Gathercole & Alloway, 2006). These general working memory problems may not be the causal deficit of SLI but are assumed to play an important role in the characteristic difficulties in reading and mathematics which children with SLI experience (Archibald & Gathercole, 2006; Gathercole & Alloway, 2006).

General working memory has not been extensively studied in children with CI. Some studies indicate that children with CI have lower levels of general working memory capacity than children with NH (Burkholder & Pisoni, 2003; Pisoni et al., 2008; Willstedt-Svensson et al.,2004), whereas other research has not found significant differences between these groups of children (cf. Sahlén, Willstedt-Svensson, Ibertsson, & Lyxell, 2008). It should be noted that these studies use different measures of general working memory. That is, in the studies by Willstedt–Svensson et al. (2004) and Sahlén et al. (2008), the tasks required a high degree of semantic processing, whereas the backward digit recall tasks used by Burkholder & Pisoni (2003) and Pisoni et al. (2008) did not require semantic processing to any great extent. Poorer performance of children with CI in tests of general WM may, however, depend on the modality of the task since they, like children with SLI (Gathercole, 2006a), may experience specific problems in auditory perception and/or phonological WM which could have negative effects on their performance in tasks of general working memory, when the test items to be processed and stored are auditorily presented. General working memory has furthermore been found to be related to lexical and grammatical development in children with CI (Willstedt-Svensson et al., 2004).

Visuospatial working memory

In comparison to phonological and general working memory, the effects of visuospatial WM on various cognitive skills are less well understood. Visuospatial working memory has been found to predict reading comprehension in children with typical development (e.g., Swanson & Berninger, 1995), and poor visuospatial WM is characteristic of children with reading disabilities, although the roots of this relationship is not well understood (Del Giudice et al., 2000; Gathercole et al., 2005). In comparison with phonological and general working memory, however, the storage (and processing) of visuospatial information is considered to be less important to the development of reading skills (Gathercole et al., 2005; Goff, Pratt, & Ong, 2005; Meyler & Breznitz, 1998). Previous results indicate that visuospatial WM capacity is associated with arithmetics and other mathematical skills in children with typical development (DeStefano & LeFevre, 2004; Rasmussen & Bisanz, 2005), particularly for younger children in preschool (Holmes & Adams, 2006). Children with mathematical difficulties have also displayed problems with concurrent storage and processing of numerical and visual information (Andersson & Lyxell, 2007).The mechanisms of these relationships are, however, not fully understood (Holmes & Adams, 2006).

The results on visuospatial working memory in children with CI are scarce and inconsistent. Cleary, Pisoni, & Geers (2001) reported lower performance levels than children with NH in tasks of visuospatial working memory and suggested that children with CI have atypical development of both phonological and visuospatial components of working memory. In contrast, a study by Dawson, Busby, McKay, & Clark (2002) did not find significant differences between children with NH and children with CI in tasks of visuospatial working memory, where the test items were unlikely to be recoded into verbal form. These inconsistent results may indeed be due to different test designs, since the task used by Cleary et al. (2001) may have primed the children to rely on phonological coding of the stimuli. Dawson et al. (2002) further found spatial working memory capacity to be a strong predictor of a composite measure of receptive language ability, e.g., syntax, linguistic concepts, and word classes.

In sum, research on the various aspects of working memory capacity in children with CI has hitherto been relatively scarce. Preliminary studies indicate that children with CI have poorer levels of phonological working memory capacity compared to hearing peers. Studies investigating general and visuospatial working memory have generated varying results, where some studies indicate normal performance compared to hearing peers and other studies report poorer results of children with CI.

Phonological skills

Definitions - an umbrella term

Phonological skills is an umbrella term which refers to the awareness of, and sensitivity to, the sound structure of language (e.g., Anthony et al., 2002; Anthony & Francis, 2005; Dally, 2006; Kjeldsen et al., 2003). Phonological skills are typically involved in a range of tasks, which require the ability to detect whether words are similar or dissimilar, for example, if they rhyme or whether they start or end with the same sound (e.g. Snowling, Gallagher, & Frith, 2003). This type of skill is also used when manipulating the sounds in a word, e.g., when reversing the order of the sounds, or when blending, removing and adding sounds (Kjeldsen et al., 2003; Oakhill & Kyle, 2000; Snowling et al., 2003). Phonological skills is thus a very broad term, which can be further categorized into a number of subskills (Anthony et al., 2002; Anthony & Francis, 2005; Kjeldsen et al., 2003; Oakhill & Kyle, 2000; Snowling et al., 2003), and there is no consensus whether these subskills are tapping into exactly the same fundamental skill (cf. Anthony et al., 2002; Anthony & Francis, 2005; Oakhill & Kyle, 2000). Consequently, it is important to define exactly what aspect of phonological skills is measured when studying these abilities. However, irrespective of whether tests commonly used to assess phonological skill measure different skills, or the same underlying skill along a continuum, the level of difficulty is considered to increase when the size of the phonological units to process decreases. That is, the processing of single phonemes is viewed as more difficult than that of rimes and syllables (Anthony & Francis, 2005; Vloedgraven & Verhoeven, 2008).

The present thesis takes a cognitive perspective on phonology and development of phonological skills. The focus is therefore on phonological representations and not on phonological production processes, as is the case in disciplines more strongly influenced by a structural linguistic perspective (for example, in speech pathology).

Here, the term phonological representation refers to the phonetic structures of speech sounds and spoken words represented in long-term memory. The quality or distinctness of those long-term memory representations in children with CI and the extent to which fine phonetic contrasts may be represented is of main interest. The tests used to investigate phonological representations in the present thesis require some kind of processing of phonological information, for example, discrimination of phoneme contrasts, in (Swedish) words e.g., /cykel/-/nyckel/ and non-words , e.g., /patina´dru:p/ - /patina´vru:p/, as well as identification and segmentation of phonemes in words and nonwords.

The present thesis is, thus, not concerned with other aspects of phonological skill such as processes involving manipulation of words, e.g., rhyming, deletion of phonemes etc. The rationale for studying phonological representations in this population of children is that the auditory stimulation from a cochlear implant does not restore normal hearing (Harnsberger, 2001; Pisoni et al., 2008) and, as a consequence, we do not know exactly what phonological contrasts they can and cannot perceive. Thereby, we do not know which level of distinctness or stability their mental representations of words and speech sounds may reach. The quality of phonological representations plays an important role for the development of phonological and linguistic skills in children with NH (Boada & Pennington, 2006; Elbro & Nygaard Jensen, 2005). Therefore, knowledge about the development of phonological representations in children with CI should be considered fundamental in research on phonological skills in this population. In other words, basic knowledge about phonological representations in this population is required before we can go on to measure other aspects of phonological skill.

The development of phonological representations starts at an early age (Kuhl, Williams, Lacerda, Stevens & Lindblom, 1992; Swingley, 2003; Näätänen, 2001; Swingley & Aslin, 2000) and is intimately related to the development of long-term memory and working memory (Baddeley, 2009). For example, the perception of phonetic contrasts (such as vowel contrasts, /i/ - /y/) is altered by the native language already when the child is 6 months of age (Kuhl et al., 1992). Furthermore, children 18–23 months of age are able to discriminate between correctly and incorrectly pronounced words in the ambient language (Swingley & Aslin, 2000).

Phonological skills versus phonological working memory

Phonological skills and phonological working memory are intimately related although the exact nature of the relationship is not completely understood (Jacquemot & Scott, 2006; Oakhill & Kyle, 2000). Jacquemot & Scott (2006) suggest that the models of phonological working memory and phonological processing should be combined for a more complete understanding of these abilities, and suggest a model which comprises both constructs and has separate buffers for input and output (Jacquemot & Scott, 2006). Furthermore, some theoretical accounts use the definition *phonological processing* with broad reference to phonological skills, lexical access and phonological working memory (e.g., Bowey, 2006; Clarke, Hulme, & Snowling, 2005). The advocates of this view claim that these skills represent the same underlying ability but use the terms *implicit and explicit phonological processing*. With these definitions, implicit phonological processing refers to phonological working memory and lexical access (speeded naming) and explicit phonological processing denotes the metalinguistic ability in which reflection and manipulation of language is required (Clarke, Hulme, & Snowling, 2005). Nonword repetition has been found to be a clinical marker and phenotype of SLI (e.g., Bishop, 2006). According to the phonological processing account, nonwords repetition is a measure of phonological processing rather than phonological working memory (Bowey, 2006). However, regardless of whether non-word repetition is a test of phonological processing or of phonological working memory, performance in this test is considered to be a language learning device for acquisition of vocabulary and syntax in children with normal hearing and typical development (cf. Baddeley, Gathercole, & Papagno, 1998; Bowey, 2006; Gathercole, 2006a).

Relations between phonological working memory, phonological skills and lexical access

In this thesis, phonological skills are viewed as separate from phonological WM and lexical access, although all three abilities are regarded as interdependent. Specifically, in tasks measuring phonological skill (such as nonword discrimination and phoneme identification), the processing of phonological information is considered to be dependent on the working memory system, such that the phonological information is briefly stored in the phonological loop while being processed (Repovš & Baddeley, 2006). Working memory is also important for the establishment of phonological representations (Baddeley, 2009; Gathercole, 2006a). This relation is further discussed in the section about working memory above. Lexical access (i.e., the ability to recognize spoken words and be able to quickly come up with the correct word for what one intends to say) is, in turn, facilitated by distinct phonological representations (Gaskell & Marslen-Wilson, 2002; Norris, Cutler, McQueen, & Butterfield, 2006). The latter relations are further discussed in the sections below.

The influence of phonological skills on other abilities

Irrespective of the exact definition of phonological skills employed, previous research agrees that these skills are important predictors for the development of language and reading skills in children with typical development (e.g., Kjeldsen, Niemi, & Olofsson, 2003; Muter, Hulme, Snowling, & Stevenson, 2004). Children who have problems detecting or manipulating speech sounds are likely to experience problems when learning to read. Poor phonological skills have further been found to be one of the main characteristics and the underlying cause of developmental dyslexia (Anthony & Francis, 2005; Joanisse, Manis, & Keating, 2000; Marshall, Snowling, & Bailey, 2001; Vellutino, Fletcher, Snowling, & Scanlon, 2004). This is an incomplete list of examples but still it illustrates the importance of studying phonological skills in children with CI in order to increase our understanding of many aspects of language and academic skills in this population.

Training of phonological skills

Previous research indicates that training of phonological skills is beneficial for the development of language and readings skills, for example, in children with typical development (Anthony & Francis, 2005; Gustafson, Ferreira, & Rönnberg, 2007), with or at risk of developing dyslexia (e.g., Kjeldsen, Niemi, & Olofsson, 2003; Lyytinen, Erskine, Aro, & Richardson, 2007), with phonological impairment (Bernhardt & Major, 2005) and with apraxia of speech (Moriarty & Gillon, 2006). However, oral phonological training may not result in permanent effects on reading skill unless the intervention is also focused on the associations between graphemes and phonemes (Lyytinen, Erskine, Aro, & Richardson, 2007). In typically developing children, the relation between phonological skills and reading ability has been suggested to be at least partially reciprocal, with substantial improvement in phonological skill as a consequence of reading instruction (Anthony & Francis, 2005; Castles & Coltheart, 2004; Holopainen, Ahonen, Tolvanen, & Lyytinen, 2000). This effect is most obvious in transparent languages, such as Finnish and Turkish (Anthony & Francis, 2005; Holopainen, et al., 2000), even though it has been found in languages with highly irregular orthographies, such as English (Castles & Coltheart, 2004).Thus longer intervention periods are needed for children learning to read in languages with deep orthographies (Lyytinen et al., 2007).

Phonological skills in children with cochlear implants

Phonological skill is poorer in children with CI compared to children with NH (Ibertsson, Willstedt-Svensson, Radeborg, & Sahlén, 2008; James et al., 2008; Schorr et al., 2008; Spencer & Tomblin, 2009). Furthermore, children with CI are reported to use their orthographic representations of words to a greater extent than normal hearing children, when solving auditorily presented, phonologically demanding tasks which use real words as test items (James et al., 2008).That is, they may use their knowledge about the spelling of a word in order to solve tasks intended to measure phonological skill. Variation in phonological skill within the group of children with CI is also substantial and may be a consequence of, for example, demographic factors such as age at implantation (e.g., James et al., 2008). Another cause of this variation may be the quality of phonological representations in individual children with CI (Pisoni et al., 2008; Harnsberger, 2001). Since the development of phonological representations starts within the first few months of life (Kuhl, Williams, Lacerda, Stevens & Lindblom, 1992; Swingley, 2003; Swingley & Aslin, 2000), auditory deprivation before implantation may cause atypical development of phonological representations due to plasticity of the brain, even when the child receives his/ her implant at a young age (Pisoni et al., 2008). Furthermore, the auditory stimulation provided by CIs does not restore normal hearing (Fallon et al., 2007; Houston, Carter, Pisoni, Iler Kirk, & Ying, 2005), for example due to lower frequency resolution. The quality of the auditory signal from the CI may thus result in less well specified phonological representations of words and speech sounds in children with CI. The phonological representations may also vary considerably between children, as a consequence of the unique pathology of the individual child (Harnsberger, 2001). In line with this, previous research reports varying results regarding phonological skills of children with CI (e.g. Chin & Lento Kaiser, 2000; Young & Killen, 2002). Chin & Lento Kaiser (2000) reported inferior phonological performance compared to children with NH in the same age range. Young & Killen (2002) studied six prelingually deaf children with CI implanted between 3;0 and 6;10 years of age. Four of these children had expressive vocabularies within the average range as measured by naming tests. It is important to consider test validity when assessing phonological skills in children with CI, in order to minimize the risk that their test performance is confounded with their ability to hear the test items (Spencer & Tomblin, 2009). Test validity for children with CI was assessed by Spencer and Tomblin (2009) on a number of tests commonly used to test phonological skills in hearing children. The children were tested in auditory-only as well as in auditory–visual conditions in tasks of phoneme deletion, phoneme blending and phonological working memory. The authors found that in most of the tests there were main effects for presentation condition (both groups had higher performance in the auditory-visual condition). There were further always main effects of group (the children with CI had lower performance than the hearing children). No significant interaction effects between group and presentation condition were found, however, indicating that tests which are typically used to measure phonological skills in hearing children may be valid measures even for children with CI (Spencer & Tomblin, 2009).

In sum, there is no consensus regarding whether the broad term phonological skills reflects one underlying ability, or a range of separate abilities. It is thus important to be specific about which aspect is measured when assessing phonological skills. The present thesis focused on phonological representations, since little is known about distinctness of phonological representations in children with CI as a consequence of the auditory stimulation they receive from their implants. The development of phonological representations is related to working memory and is further fundamental for other aspects of phonological skills, both in children with CI and in children with NH.

Lexical access

Lexical access refers to the efficiency, in terms of speed and accuracy, of retrieving language representations and/or conceptual/semantic information from long-term memory (Anthony & Francis, 2005; Gaskell & Marslen-Wilson, 2002; Norris, Cutler, McQueen, & Butterfield, 2006; Penolazzi, Hauk, & Pulvermüller, 2007). Lexical access is inherent in speech recognition (Gaskell & Marslen-Wilson, 2002; Norris, Cutler, McQueen, & Butterfield, 2006), for example when we hear the word 'dog' we instantly think of a furry creature with a tail that can bark and run after cats (the semantic representation). Probably we also see a picture of a specific familiar or stereotypical dog before our eyes. Lexical activation also goes in the opposite direction in speech production (e.g., Goldrick & Rapp, 2007; Navarrete & Costa, 2005; Schwartz, Dell, Martin, Gahl, & Sobel, 2006). For example, when we see a dog, automatically the word 'dog' comes to mind (the lexical/phonological representation).

These processes occur without our conscious reflection but most of us know that they may fail sometimes, for example, when we cannot come up with a specific word even though we know that it is included in our mental lexicon, i.e., the tip-of-the-tongue phenomenon (e.g., James & Burke, 2000). The efficiency of lexical activation of any specific word is influenced by various factors related to familiarity, such as the age at which the word was learnt (e.g., Cuetos, Alvarez, González-Nosti, Méot, & Bonin, 2006; Gershkoff-Stowe, 2002) and the frequency with which it is encountered in language (Gaskell & Marslen-Wilson, 2002; Goldrick & Rapp, 2007). The linguistic context where the word or concept occurs, word length (Penolazzi, Hauk, & Pulvermüller, 2007), and the word's neighbourhood density, i.e., the number of phonologically similar words (Gaskell & Marslen-Wilson, 2002; Goldrick & Rapp, 2007), also affects lexical access.

Lexical access is thus very important for our processing of information and often includes the activation of both semantic and phonological representations (Gaskell & Marslen-Wilson, 2002; Johnson, Clark, & Paivio, 1996; Norris, Cutler, McQueen, & Butterfield, 2006; Schwartz et al., 2006). In many models of lexical access, for both recognition and production of speech, phonological and semantic information about words in long-term memory are conceptualized as separate representations, although these representations are considered to be interconnected in a network (e.g., Goldrick & Rapp, 2007; Navarrete & Costa, 2005; Norris et al., 2006; Schwartz et al., 2006). Some preliminary support for this structure of lexical access is provided by neuroimaging studies of individuals suffering from a stroke (DeLeon et al., 2007) and amnesia (Meinzer et al., 2006) indicating that a network of brain regions supports naming (DeLeon et al., 2007).

Theories of speech recognition typically conceptualize lexical access as a process in which perceptual input activates lexical representations from long-term memory (Gaskell & Marslen-Wilson, 2002; Norris et al., 2006). Whether a specific word in long-term memory is activated or not is determined by how well the incoming speech signal matches the stored phonological representation (Gaskell & Marslen-Wilson, 2002; Norris et al., 2006). In this view, recognition of any specific word occurs when the level of activation of a representation in long-term memory reaches a certain threshold. It should also be noted, at this point, that the process of lexical access in speech recognition may be facilitated by the factors mentioned previously, such as, the context in which the word is perceived (e.g., Gaskell & Marslen-Wilson, 2002; Norris et al., 2006). The literature on lexical access in speech recognition and speech production reviewed above, rarely considers the influence of other cognitive factors (e.g., working memory and attention) or discusses the cognitive implications of lexical access in speech perception and production. The process of matching an incoming speech signal to a lexical representation in long-term memory is, however, also linked to phonological WM. In a process called *redintegration* (Gathercole, 2006a; Lewandowsky & Farrell, 2000; Turner, Henry, Smith, & Brown, 2004), the incoming speech signal, possibly incomplete due to hearing problems and/or noise, is held in the phonological loop until a matching long-term representation is activated. Redintegration may improve working memory performance such that familiar words, which are easily accessed from long-term memory, are better recalled than unfamiliar words (Turner et al., 2004). Lexical access and phonological working memory are further related through vocabulary, since a larger vocabulary is beneficial for efficient retrieval of lexical and semantic representations (Gathercole, 2006a). Furthermore, lexical access and phonological WM may be related through vocabulary, as a consequence of the strong relation between novel word learning and phonological WM (Gathercole, 2006a). See also Rudner, Foo, Rönnberg, & Lunner (2009) for a working memory model of language understanding, including lexical access in adults.

It has been debated whether lexical access represents an ability which is separate from phonological skill (Vellutino et al., 2004). As mentioned in the section about phonological skills, some argue that lexical access, together with phonological WM, is part of a system dedicated to implicit phonological processing (Clarke, Hulme, & Snowling, 2005; Vellutino et al., 2004). Lexical access and phonological skills are indeed related to each other developmentally, but an increasing number of studies indicate that lexical access skills should be considered as separate from other aspects of phonological skill (cf. Clarke, Hulme, & Snowling, 2005; Frost, 1998; Powell, Stainthorp, Stuart, Garwood, & Quinlan, 2007; Savage & Frederickson, 2005). Lexical access and phonological skills make independent contributions to reading ability (e.g., Clarke et al., 2005; Di Filippo et al., 2005; Georgiou, Parrila, & Kirby, 2006; Plaza & Cohen, 2003; Savage & Frederickson, 2005) and spelling (Plaza & Cohen, 2003; Swan & Goswami, 1997).

In this thesis, lexical access is regarded as an ability separate from phonological skill and working memory. The relation between lexical access and reading skill is not well understood (Clarke et al., 2005; Powell et al., 2007; Vellutino, Fletcher, Snowling, & Scanlon, 2004). In children with typical development, lexical access, as operationalized by performance in rapid naming tasks, has been reported to be a relatively stronger predictor of orthographic (word) decoding and reading speed, whereas phonological awareness tasks, such as, phoneme deletion and phoneme substitution, predict a larger proportion of unique variance in phonological (nonword) decoding (Clarke, Hulme, & Snowling, 2005; Manis, Doi,& Bhadha, 2000; Savage & Frederickson, 2005) and reading comprehension (Savage & Frederickson, 2005). A possible cause for these relationships is that rapid naming reflects processes involved in the establishment of orthographic representations (Clarke, Hulme, & Snowling, 2005; Manis, Seidenberg, & Doi, 1999). Orthographic representations are, in turn, important for efficient and fluent reading (Castles & Nation, 2006; Conrad, 2008; Nunes & Bryant, 2009; Share, 2004), a topic which will be further addressed below. Furthermore, neuroimaging results from a study by McCrory, Mechelli, Frith, & Price (2005) on adults with dyslexia indicate that picture naming, a commonly used measure of lexical access, and word reading result in similar patterns of activation in the brain (i.e., reduced activation in the left occipito-temporal region, McCrory, Mechelli, Frith, & Price, 2005). The authors suggest that the relation between the poor picture naming and word reading skills seen in persons with dyslexia may be the result of a general impairment in retrieving phonological information from visual input (McCrory et al., 2005). These results provide additional support for the view that rapid naming may reflect the ability to establish orthographic representations, since retrieval of phonological information from orthographic representations should be considered inherent in this process.

Lexical access in children with cochlear implants

Few studies have investigated lexical access in children with CI. Two recent studies, however, focus on lexical access in speech production (Schorr et al., 2008; Spencer & Tomblin, 2009). Schorr et al. (2008) found that 56 children with CI in the age range from 5–14 years had a significantly lower performance level than a hearing comparison group on a composite measure of rapid naming (colours, objects, digits and letters). Seventy-eight percent of the children with CI in that study (as compared to 94% of the hearing children) performed within the average range for hearing children. Spencer & Tomblin (2009) assessed rapid naming of letters and digits in 29 children with CI and found no significant difference between these children and a hearing comparison group on either of these tests. The children with CI, however, had higher and less varying results on letter naming compared to digit naming. In that group of children, rapid naming of letters was, further, significantly correlated with word reading and reading comprehension. These results indicate relatively high performance levels in lexical access in speech production for children with CI. Lexical access in speech recognition has not to our knowledge previously been assessed in this population of children. Correct recognition of a spoken word is dependent on how well the incoming speech signal matches the corresponding stored representation in the mental lexicon (Gaskell & Marslen-Wilson, 2002; Norris et al., 2006). Correct semantic representations are, in turn, needed to identify the meaning of the spoken word. We may hypothesize that lexical access in speech recognition is particularly difficult for children with CI because correct identification of words requires both distinct phonological representations of words in long-term memory (Elbro, Borstrom, Klint Petersen, 1998; Norris et al., 2006) and stable perception of input signals (Harnsberger, 2001; Pisoni et al., 2008). These two prerequisites for lexical access in speech recognition may be compromised for children with CI. That is, their phonological representations may be less distinct as a consequence of their anomalous auditory experience (Harnsberger, 2001; Pisoni et al., 2008; Svirsky, Robbins, Iler Kirk, Pisoni, & Miyamoto, 2000), and the input signal has also been reported to vary largely from time to time for the individual child (Pisoni et al., 2008). Lexical access in speech recognition may therefore be problematic for children with CI. In the present thesis, the efficiency of lexical access in speech recognition is examined. In two of the tests of lexical access used (Semantic Decision making and Passive Naming), both phonological and semantic representations need to be accessed in order to solve the task, whereas another task (Wordspotting) primarily measures access to phonological representations. In the latter task, recognition of the phonological form of a presented word is enough to solve the tasks. The children therefore do not need to know the meaning of the test items.

Reading ability

Typical reading development

Reading comprehension, which is the ultimate goal of reading, is composed of two component skills, namely the technical aspects of reading which are commonly labelled *decoding* (i.e., transforming strings of letters into words) and comprehension (e.g., Gough & Tunmer, 1986; Høien & Lundberg, 1992; Gustafson, 2000; Oakhill, Cain, & Bryant, 2003). For reading comprehension to occur, written words thus need to be correctly decoded, and comprehension is required in order to understand the message conveyed by words and sentences. These components of reading ability are considered to be independent although intimately related abilities (e.g., Cain, Oakhill, & Bryant, 2004; Oakhill, Cain, & Bryant, 2003). Decoding is fundamental in reading comprehension since the written words in sentences or text passages must be deciphered before they can be interpreted (Oakhill, Cain, & Bryant, 2003). Thus, for example young children, who still have problems with decoding, and persons with dyslexia will consequently also have problems with reading comprehension (e.g., Snowling, Gallagher, & Frith, 2003; Vellutino et al., 2004). The opposite is not necessarily true; words may be perfectly decoded without being comprehended. We can, for example, often correctly decode foreign words which we do not know the meaning of. High decoding skills but without the ability to understand what is read may also be found in clinical populations, such as, persons with hyperlexia and autism (Grigorenko et al., 2002; Newman et al., 2007).

Phonological decoding

The decoding component can be further divided into two types of decoding strategies, *phonological* and *orthographic* decoding. Phonological decoding refers to the process of mapping visually presented letters onto speech sounds (de Jong & Share, 2007; Snowling, 2000; Ziegler & Goswami, 2005). Phonological decoding is the fundamental key to reading ability, which the beginning reader typically has to master in order for subsequent reading development to be smooth and efficient (e.g., Castles & Nation, 2008; Kyte & Johnson, 2006; Share, 2004; Snowling, 2000; Sprenger-Charolles, Siegel, Béchennec, & Serniclaes, 2003). Phonological decoding is also used by more advanced readers at later points in reading development, for example, when decoding unfamiliar words, not previously seen in written form, or when decoding words of a foreign language for the first time (Gustafson, 2000). The mechanisms behind phonological decoding skill have been investigated in numerous studies, where the ability to process and store various units of phonological information in working memory has proved to be the most important prerequisite for development of phonological decoding ability (e.g., Anthony et al., 2002; Dally, 2006; Muter, Hulme, Snowling, & Stevenson, 2004; Ziegler & Goswami, 2005). These relationships are discussed in further detail in the separate sections about cognitive skills.

Orthographic decoding

After the child has acquired the basic phonological decoding skills, with reading experience, she/he starts to recognize words orthographically, “by sight”, without having to decode in a letter-by-letter fashion (e.g., Apel, in press; Cunningham, Perry, & Stanovich, 2001; Share, 2004). Orthographic decoding allows for faster and less cognitively demanding reading than phonological decoding (Castles & Nation, 2008; Conrad, 2008; Nunes & Bryant, 2009; Share, 2004). Orthographic decoding is possible for those words of which the child has established specific orthographic representations in long-term memory (e.g., Apel, in press; Cunningham, Perry, & Stanovich, 2001; Share, 2004). Thus, skilled orthographic decoding requires a large internal lexicon of distinct orthographic representations (Burt, 2006; Castles & Nation, 2008; Conrad, 2008; Perfetti, 1992). For the beginning reader new orthographic representations need to be acquired constantly to increase the orthographic lexicon. The transition from phonological decoding to an orthographic reading strategy has been suggested to be mediated through a process called orthographic learning (Castles & Nation, 2006; de Jong & Share, 2007; Share, 1999; 2004). In this account, children acquire detailed orthographic representations through basic phonological decoding, and efficient phonological decoding makes the child attend to the order and identity of letters and how they map onto the phonological representation (de Jong & Share, 2007; Share, 2004).This process of learning orthographic representations has been found to occur as children practice reading on their own, both in oral and in silent reading (Cunningham, 2006; de Jong & Share, 2007). Every correctly decoded new word is regarded as an opportunity to learn the specific orthographic representation of this word (de Jong & Share, 2007; Share, 2004).

Reading comprehension

Decoding skills are essential but not sufficient for reading comprehension to occur. A number of other factors are known to influence this complex cognitive skill (Cain, Oakhill, & Bryant, 2004; Hannon & Daneman, 2001a; 2001b; Seigneuric et al., 2000). Examples of such factors which, together with decoding skill, affect reading comprehension include vocabulary and verbal skills (e.g., Seigneuric et al., 2000; Cain, Oakhill, & Bryant, 2004), prior (world) knowledge (Cain, Oakhill, & Bryant, 2004) and the accessibility of this information from long-term memory (Hannon & Daneman, 2001a; 2001b), inference making (Hannon & Daneman, 2001a; 2001b), knowledge about story structure, working memory (Oakhill, Cain, & Bryant, 2003), and motivational factors (Seigneuric et al., 2000). Early in reading development, reading comprehension is more dependent on decoding skills than later in development (Cain et al., 2004). This is because decoding is still very demanding for the child and thereby a lot of processing capacity is used, which results in few resources left for text comprehension processes, such as integration and inference (Cain et al., 2004).

Reading in children with cochlear implants

Research examining reading skills in children with CI has resulted in diverging outcomes. Some studies indicate that around 50% of children with CI have age-appropriate levels of both decoding and reading comprehension at an early age (Dillon & Pisoni, 2006; Geers, 2003; Marschark, Rhoten, & Fabich, 2007). Recent results from Geers et al. (2008), however, indicate that the proportion of children who have reading comprehension skills within the normal range may be smaller in high school than in elementary grades. Another recent study by Ibertsson, Asker-Árnason, Hansson, Lyxell and Sahlén (2009), of seven teenagers (14–19 yrs of age) indicated that all seven participants had age-appropriate decoding skills in tasks tapping orthographic decoding strategies, whereas three out of seven were more hampered than hearing teenagers in reading tasks where phonological strategies had to be employed.

Other researchers (Dillon & Pisoni, 2006) suggest that children with CI may, to some extent, use phonological decoding at an early stage of reading development (7–9 yrs of age), since they found that nonword repetition was correlated with decoding skills as well as reading comprehension at this age. A number of other factors, e.g., post-implant vocabulary, language skills, nonverbal intelligence, family socioeconomic status, and early auditory experience also have been found to influence reading outcome (Marschark et al.,2007; McDonald Connor & Zwolan, 2004).

Method

Methodological challenges

Children with CI constitute an exclusive population for studying the effects that auditory stimulation from an implant may have on cognitive ability in children who have a severe to profound hearing impairment. This population of children is, however, small and heterogeneous, characteristics that make research on this group of children a challenging and demanding task. The first challenge pertains to the heterogeneity of the population. Children with CI are heterogeneous with respect to, for example, their diagnosis (i.e., cause of hearing impairment), age at onset of hearing impairment, age at implantation (e.g. Geers, Tobey, Moog, & Brenner, 2008; Holt, Svirsky, Neuburger & Miyamoto, 2004; McDonald Connor & Zwolan, 2004), type of implant, i.e., number of active electrodes, type of processor etc. (Geers, Brenner, & Davidson, 2003), anatomical aspects of the cochlea and thereby exact fit of the electrodes of the implant (Papsin, 2005). Other variables that may vary between children is their main mode of communication, oral, sign or various combinations of the two (Geers et al., 2008; Gravel & O'Gara, 2003), educational settings and pedagogical profiles at school, as well as the access to both human and technological resources. These are only examples of the large number of variables which may vary between children with CI, and which may directly or indirectly influence implant benefit and cognitive development in children who use these devices.

A further challenge is the fact that the children are geographically spread nationwide. This poses logistical problems as we cannot easily access our participants and the time for testing the children is very limited, at least in the perspective of writing a doctoral thesis.

As in all research, ethical aspects have to be taken into consideration when studying children with CI. There are also a number of specific issues that needs to be taken into account when studying small and heterogeneous populations, such as children with CI. For example, we may not be able to test the children as often and as long as might be required in order to answer all types of research questions. Testing times for children should always be kept to a minimum, but this should be even more important to take into account for this population since they are regularly tested within their cochlear implant programs. They are also frequently asked to participate in various other research studies. The small size of the population further makes participants relatively easy to identify. This has implications for the way in which the results of the studies may be presented and published, i.e., all relevant information about for example etiology, is not always possible to present.

Children with NH are used as comparison groups in the present thesis. This poses problems of measurement (i.e., ceiling effects), in some cases, as the children with NH have, and should be the expected to have, a performance level of 100% correct (e.g., in phoneme discrimination).

Another important factor, which sets the limits to the types of studies that may be conducted, is the low level of knowledge regarding cognitive development in children with CI. As mentioned in the introduction, relatively extensive research has investigated the medical and technological aspects of implantation in children. Behavioural research on this population has mostly studied the effects of various demographic variables for implant benefit (Geers et al., 2008). Compared to these areas of research, the cognitive effects of cochlear implant hearing have not been as extensively explored (see Pisoni et al., 2008; Marschark et al.,2007, for reviews). Thus, the level of knowledge in this area, in combination with heterogeneity of the small population, makes a research strategy where we employ "sharp" hypothesis testing designs more difficult in most cases. Therefore, approaches based on more descriptive research questions may be the most fruitful, if not the only, way to go in the initial stages of research within this area. In other words, we need some form of description of the population before hypothesis-testing strategies are adopted.

Test modality and validity

Almost all cognitive tests used in the present thesis, were presented in the auditory modality. The rationale for this choice of method was that the results should be as ecologically valid as possible. That is, the auditory stimulation that children perceive through their implants is different from that of hearing children, but still it is the auditory perception that they have to manage in every-day situations. This is particularly evident since 50 percent of the children used oral communication as their main communication mode at home and at school. The other 50 percent of the children used combinations of oral communication and sign. It is thus important to study the characteristics of the cognitive skills which are dependent on the auditory modality, since children with CI have a unique development of these skills compared to other populations, and we need to find out how well they may perform with the auditory perception that they have.

There is always a risk that auditory perception may bias the cognitive performance of children with HIs in tests with auditory test presentation (James et al., 2005; 2008). In order to minimize the risk that performance would be impaired by auditory perception, the volume was individually adjusted to a comfortable level before the start of every test session.

Recent findings from Spencer and Tomblin (2009) suggest that phonological skills may be accurately assessed in children with CI in a number of standardized tests used to measure phonological skills in hearing children. At this point, it should also be noted that tests intended to assess phonological skills with written stimuli as test items may not constitute better alternatives, since orthographic rather than phonological skills may be tapped by these tasks (Castles & Coltheart, 2004). This is particularly important to consider as persons with hearing impairment have been reported to use orthographic information when making phonological judgements whenever applicable (James et al., 2005). For the purpose of the present thesis, text-based test presentation was not an option since young children with little or no reading skills should be able take the tests. Their reading ability would thereby have influenced their results.

A second alternative to using auditory-based tests of phonological skill may be to present the test items in picture-form. Some studies have successfully used this strategy for testing phonological awareness in children with CI (James et al., 2005; 2008). We chose not to adopt this methodology for the following reasons. First, picture-based tasks would require all the test material in the tests of general and phonological working memory and phonological skills to consist of real, easily depicted words. Recall of real words is known to be easier than recall of nonwords since semantic information from long-term memory may be used to improve performance (Baddeley, 2009; Repovš & Baddeley, 2006). Recall of real words is therefore, considered not to measure phonological working memory with the same precision as recall of nonwords, since semantics and long-term memory affect performance (e.g., Baddeley, 2003; Gathercole, 2006a; Repovš & Baddeley, 2006). A second disadvantage of picture based tests is that another cognitive step would be added to every task. That is, before any storage or processing of information can take place, the picture has to be identified and labelled. Thus another confounding variable would be added which may complicate the interpretation of results and make most of the tests less pure. The lexical access tests may be best suited for picture-based presentation, since rapid naming (RAN) tasks typically involve speeded naming of pictured objects. With that approach we would, however, have measured lexical access in speech production rather than in speech recognition. With consideration to the preceding arguments, and with the purpose to measure the ability to store, make decisions about and process auditory/verbal information we decided to use the auditory mode of presentation in all of the cognitive tests except for the tests of visuospatial working memory and nonverbal ability.

Tests on auditory perception

Information about auditory perception was accessed from the medical case notes of almost all of the children who participated in the present studies. Since the children attended cochlear implant programs in different parts of Sweden, their hearing was, however, assessed by different tests. The hearing levels of individual children could therefore not be compared. Another issue concerns the fact that auditory perception for most of the children was assessed in speech recognition tests where the task was to replicate words or sentences. Since these tests use linguistic material to assess auditory perception, the performance level of a child tested with these measures may be a composite measure of their auditory perception, articulation skills, phonological working memory, vocabulary and/or lexical skills (since the child needs to be familiar with the words used). These characteristics of the tests may make them particularly uninformative when used for children with motor- and/or cognitive problems (see Blamey, 2003 for a discussion of similar problems in adults, children who use hearing aids and children with CI). A further potential problem is that some of the most commonly used tests contain words that are rare in contemporary Swedish vocabulary (Almqvist, 2004). Because of this situation we have not been able to relate the cognitive performance of the children with measures of auditory perception.

It should be noted at this point that there is a lack of tests on auditory perception for children in Sweden. It should also be noted that this could be a problem for hearing research in this population. The tests described above may be clinically useful measures of how the children manage in some situations requiring speech perception. However, when these Swedish auditory tests are used for a research purpose, we cannot be certain to which extent they are measures of auditory perception or of cognitive skills. It is thus obvious that we need to develop non-linguistic tests of auditory perception to be able to learn more about the influence of auditory perception on the cognitive performance of Swedish children with CI.

The challenges mentioned above are examples of factors that obviously affect research on this group of children. Thus, not all research questions are applicable to this population for practical and theoretical reasons. In order to meet these challenges, we used different research designs depending on the specific characteristics of every sample of children that we studied. Furthermore, comparisons with reliable reference material and theoretical comparisons to other populations are important features of the research designs applied.

Test battery

The cognitive tests used in the present thesis were mainly presented and performed by means of a computer-based test battery, specifically designed for this purpose. The test battery was named the *SIPS* (*Sound Information Processing System)*, analogous to the name of a similar test designed for adults, i.e., the *TIPS, Text Information Processing System* (Hällgren, Larsby, Lyxell, & Arlinger, 2001). In most of the cognitive tests within the SIPS, both accuracy and speed of performance are automatically recorded by the program. The fact that speed and accuracy are measured in the tests is an important feature since this enabled us to discriminate between children who had identical performance in terms of number of correct responses but who used different amounts of time to finish a test item, or vice versa, children with similar speed of processing but different levels of accuracy. This is because response latency reflects the speed of information processing and thereby the efficiency with which information is processed (e.g., Hällgren, 2005; Luna, Garver, Urban, Lazar, & Sweeney, 2004). Thus, the inclusion of response latency provides more information about cognitive performance than measures of accuracy alone.

In tasks of phonological skill, which require oral responses, performance could be restricted by impaired function in either processes reflecting the phonological skill (in the sense of ability to, for example, discriminate and manipulate sounds) or in the processes required to generate an oral response. Young children may for example be able to hear which pronunciation of a word is correct but still they cannot pronounce the word correctly themselves. These processes may be partly independent (Jacquemot & Scott, 2006). Therefore, very few of the cognitive tests included in the SIPS battery require oral responses. Instead, responses are made by pressing a computer key, thereby eliminating output phonology as a potential confounding variable. Verbal responses were however required in the tests of phonological and general working memory; Nonword repetition, Serial recall of nonwords, and Sentence completion and recall. In order to control for the phonological output factor, a test of output phonology (Hellqvist, 1995) was included in the fourth study. This test requires the child to name a number of familiar words. Performance in this test was subsequently compared with the results in the working memory tests, in order to find out whether performance may have been impaired by poor output phonology. Verbal responses were also common in a fourth test, the Phonological Representations test where the children had to answer 'yes' or 'no'. They were also allowed to respond by nodding or shaking their heads.

Reading tests

Six different tests were used to assess reading ability in the present thesis. These tests are described in further detail in the separate papers. All of the reading tests were presented in paper versions. The tests of reading comprehension used in the first data collection were standardized tests. In the second data collection we intended to assess other aspects of reading and therefore decoding tests for words and nonwords were added. We also used a different test of reading comprehension than in the first data collection. The reason was that the previously used tests were poor at discriminating reading ability in children.

Participants and recruitment

Cochlear implant teams in the southern and middle parts of Sweden and the national organization for children with CIs and their parents, *Barnplantorna,* assisted us in recruiting children with CI. A total of 28 children with CI participated in the studies. Most of the children with CI in study III also participated in study II. The children and their parents were contacted and informed about the purpose of the studies, the types of tests used, testing time and contact information to the researchers. They were also informed about confidentiality of the results and their right to withdraw their child from the studies without stating a reason for doing so. The children's parents had to sign a written informed consent in order for their children to participate. The participating children were tested in one of the following conditions: at a summer camp for children with CI and their families, at a regular follow-up at their cochlear implant program, or at their school. In those cases where the test sessions took place at school or at the cochlear implant clinic, it was done in agreement with the children's teachers or clinicians. A total of 118 children with NH constituted the comparison group. They were recruited at their schools. The same information as for the children with CI was distributed to their families and written informed consent from the children's parents was required for participation.

Levels of analysis

Given the challenges mentioned in the previous section, we are not always able to perform studies at a group level, even by applying quasi-experimental designs. This is because the population of children with CI in Sweden is so small that we cannot draw samples of individuals who are matched on all the particular demographic variables that may affect performance. Therefore, our studies are performed at three levels of analysis: group level (where applicable), subgroup level, and case level.

The research questions, and the extent to which the children can be matched on variables that may affect performance, have guided the designs used. At the subgroup level, the children were matched on some variable or variables of interest and case-studies were used where we do not have enough children who are homogeneous in the desired respects to be treated as a group or subgroup. Case studies, where we study individual children, were applied in order to generate new theoretical data. The rationale for performing research at the subgroup and case levels is that internal validity will increase as compared to conditions where less strict inclusion criteria are applied and all children are treated as one group (Todman & Dugard, 2001). That is, the various variables which may act as confounding variables and provide alternative explanations to our findings are controlled to a great extent in subgroup-and case designs. In these designs, data may thereby be analyzed by means of visual analysis in combination with statistical analyses (Todman & Dugard, 2001). The type of conclusions that we may be able to draw from our studies, and the extent to which the results may be generalized, thus depend on the type of question asked and on the research design used. That is, at the group level, we may draw general conclusions about the empirical pattern found in children with CI compared to hearing children. At this level of analysis, we may also be able to draw some general causal conclusions and generalize our findings to other populations of children with CI. With reference to the comparison between children with NH and children with CI, we may also theoretically relate the performance of children with CI to other populations, for example, children with dyslexia or SLI. More specific conclusions may be drawn from studies of subgroups and detailed conclusions may be drawn at the case level. A problem with the latter two types of study designs is that they do not allow us to directly generalize our findings to other populations of children with CI, due to the small number of participants. The findings may, however, be evaluated in comparison with research on other cases or groups of children, e.g. children with NH and typical development or children with reading impairment or SLI (Todman & Dugard, 2001).

The main issues discussed here must be taken into consideration when we study children with CI. The research presented in this thesis has sought to comply with these issues and still perform research on questions that are theoretically interesting.

General aims

The general aims of the present thesis were to study various cognitive abilities in children with CI in comparison to children with NH and how these skills are related to reading ability. A second aim was to develop a test platform (SIPS) for testing cognitive skills that allows for testing in different modalities, as well as recording responses in terms of response time and level of accuracy (Study I). The following more specific research questions were addressed in the studies:

1. What are the levels of cognitive capacity in children with CI compared to children with NH (Study II & IV)?

2. What level of reading skill do children with CI possess compared to children with NH, and in relation to their cognitive skills (Study III & IV)?

3. Which demographic variables are associated with cognitive ability and reading skill in children with CI (Study II & IV)?

Summary of the papers

Paper I

Purpose

The first paper investigated the relation between orthographic learning, i.e., learning the spelling of words, and other aspects of reading skill (phonological, and orthographic decoding, and reading comprehension). Secondly, the relationship between working memory (phonological-, general-, and visuospatial WM) and orthographic learning was studied.

Method

A total of 108 children with NH, 53 boys and 55 girls, 10–25 per grade (1–6) participated in this study.

Their cognitive abilities were assessed by means of the SIPS. *Phonological working memory* was assessed in the Nonword Repetition task, and the Serial Recall of Nonwords task. *Visuospatial working memory* was studied in the Matrix Patterns test, and *general working memory* (simultaneous storage and processing) was studied in a Sentence Completion and Recall task, which may also be considered to tap the function of the central executive within the multi-component model of working memory (Baddeley, 2000; Repovš & Baddeley, 2006).

Decoding of words and nonwords, respectively, was tested by the *Test of Word Reading Efficiency* (*TOWRE*; Torgesen, Wagner & Rashotte, 1999, Swedish version by Byrne et al., 2009). Reading comprehension was assessed in one of the standardized reading comprehension tests, SL 40, SL 60 or OS 64 (Nielsen, Kreiner, Poulsen & Søegård, 1983, 1989, Swedish versions by Magnusson & Nauclér, 1997) or a Swedish translation of the *Woodcock Reading Mastery test – Revised* (Woodcock, 1987, Swedish version by Byrne et al., 2009).The test of orthographic learning was identical to the Swedish version of the test used by Byrne et al. (2008) where the child had to learn the spellings of nonsense words. Orthographic decoding was assessed in the Scandinavian version of Orthographic Choices used by Byrne et al. (2008). This test required correct identification of the spellings of words.

Results & Discussion

Orthographic learning was found to correlate with all reading measures and with the measures of visuospatial and phonological WM. The findings support the view that orthographic learning is important in reading acquisition. Linear regression analyses revealed that phonological WM and age significantly predicted orthographic learning. Nonword decoding was predicted by age and visuospatial and phonological WM, but word decoding was not predicted by any of the cognitive variables over and above the variance predicted by age. These results are interesting since previous research does not provide information about the cognitive factors associated with orthographic learning and orthographic processing. The results further revealed that nonword decoding was correlated with visuospatial and phonological WM. The correlations between visuospatial WM, the decoding measures (nonword decoding, and orthographic choices) and orthographic learning are in line with recent research in which visuospatial WM has been found to affect performance in reading and arithmetics. Reading comprehension was correlated with phonological WM, general WM and nonverbal intelligence. General and phonological WM were also significant predictors of reading comprehension.

Paper II

Purpose

The aim of the second paper was to investigate various aspects of working memory capacity, lexical access and phonological skills in children with CI, compared to age-matched children with NH. A second purpose was to study the relationships between these cognitive skills and a number of demographic variables, i.e., age at diagnosis, age at implantation, time interval between first and second CI, main communication mode, and school setting.

Method

Nineteen children with CI and 56 children with NH participated in the study. The group of children with CI consisted of 11 girls and 8 boys (age range 5;7 - 13;4 yrs). All of the children with CI had onset of severe to profound HI before 3;0 years of age and had received their first implants between 1;9 and 10;0 years of age. Eleven of the children had bilateral implants. The group of normal hearing children (28 girls and 28 boys) represented the same grades as the children with CI (i.e., grades 0, 1, 2, 4, 5 and 6 in the Swedish school system), and included 10 to 11 children in each grade.

The tests of working memory were identical to the tests used in Study I. Phonological skills were assessed in the Nonword Discrimination test, and lexical access was investigated in three tests: Passive Naming, Wordspotting and Semantic Decisions.

Results & Discussion

The children with CI had poorer performance than the children with NH on the tests of phonological and general WM, phonological skills (accuracy) and lexical access. No significant differences were found in the test of visuospatial WM.

Out of the cognitive tests, the children with CI had particularly poor performance compared to the children with NH on the measures of phonological WM. However, when the most lenient scoring criterion of the Serial Recall test was used, the difference between the groups was smaller and was further not significant in grade 4. These results were interpreted as an indication that children with CI have relatively less problems in tasks of phonological working memory, which require a lower level of detail in the phonological representations to be stored. The children with CI also had significantly lower performance than the hearing children on the general working memory measure. Lower general working memory capacity of children with CI may be an effect of poorer phonological working memory capacity and reduced speed and efficiency in the processes of sub-vocal rehearsal and serial scanning, as suggested by Burkholder & Pisoni (2003).These results may also stem from the fact that real words were used in the test of general working memory. Thus, support from phonological, semantic and lexical representations in long-term memory may improve performance in the general working memory task (Gathercole, 1999). It may be that children with CI benefit more from support from long-term representations, and the use of top-down processing strategies than children with NH, since this would make them rely less on auditory perception. Their difficulties with phonological working memory may be caused by problems in the phonological loop of working memory, according to Baddeley's (2000) model. They may also, according to Jacquemot and Scott (2006), stem from problems in the output buffer and/or the input-output conversion mechanism rather than problems in the input buffer, since these children performed relatively high on the phonologically demanding Non-Word Discrimination task without requirements on output phonology. Less developed motor aspects of articulation may also be a possible cause for these difficulties.

In the lexical access tests, approximately 50% of the children with CI, performed within 1 *SD* of their grade-matched comparison group on the latency measures and 20–30% performed within 1 *SD* of the mean of the hearing children on the accuracy measures. These findings could indicate that children with CI can access their phonological and semantic representations of words in long-term memory at a high speed, when the quality of these representations is high, i.e., when the words are highly familiar to them.

The children with CI had a significantly poorer performance than the children with NH on the accuracy measure of the Non-word Discrimination test. There were no significant differences between the groups on the response latency measure of the test. Demographic variables were only associated with cognitive performance in two of the tests measures. That is, the children with CI who used oral communication had a higher score on the Passive Naming accuracy measure of lexical access, than those children who to some extent used sign. They also had higher performance in one of the measures of phonological WM. There was no difference in performance between children attending different school settings.

Paper III

Purpose
The third paper aimed to study the relationships between working memory capacity and language skills, especially phonological skills, and reading comprehension in Swedish children with CI. A second purpose was to investigate the possible relationships between demographic factors (e.g., age at implantation, length of implant use) and reading comprehension.

Method
Sixteen children with CI, 6 boys and 10 girls (7.2 to 13.4 yrs of age) who had received a diagnosis of severe to profound bilateral hearing impairment before 36 months of age were included in the study. Nine of the children had bilateral implants. The performance of the children with CI were compared to age/grade references for children with NH (based on 10–12 children in each grade, 1–6) for the measures of visuospatial WM, phonological WM, general WM, phonological skill, and lexical access. Age references for the general WM measures in grades 4–5 were obtained from 27 children (Ahlgren & Grenner, 2005).The cognitive measures were identical to the tests used in paper I and II, except for the complex WM measure used for 3 children in grade 4, Competing Language Processing Task (the CLPT, Gaulin & Campbell, 1994, Swedish version by Pohjanen & Sandberg, 1999). Since two different tests were used to assess complex WM, a relative score for complex working memory (CVM) was developed, in which the scores were related to standard deviations in reference populations for the particular test.

Reading comprehension was assessed by the tests included in Study 1 (SL 40, SL 60 or OS 64).

Results & Discussion

Ten out of the 16 children (63%) performed at or above the 25th percentile and within 1 SD of hearing children in terms of percent sentences/words correct. The children with CI in this study therefore appear to have slightly better reading comprehension than the children studied by Geers (2003). This may be a consequence of the fact that the sample in Geers' (2003) study was more homogeneous with respect to age and time factors than the sample of children in the present study. A second explanation for the differences relates to the fact that Swedish and English differ in orthography and if children with CI, as suggested by Dillon and Pisoni (2006), use phonological decoding skills, then reading might be easier in Swedish, which has a more transparent orthography.

The demographic variables did not correlate with the reading measures in this study, which is in line with several previous studies of children with CI (cf. Dillon & Pisoni 2006; Sahlén et al., 2007). A number of significant correlations between cognitive skills and reading ability were found. The only association, which remained significant when age was controlled, was that between reading percentile and one of the complex working memory tasks (SCR).

A comparison of the best and the poorer readers confirm the important role of working memory for reading comprehension. It should be remembered that reading comprehension is a complex skill and much more than only comprehension of sentences and words is required.

Paper IV

Purpose

The main purpose of the fourth paper was to investigate phonological representations and phonological skills in children with CI in further depth. A second aim was to study reading strategies in children with CI and to find possible cognitive correlates of the strategies that they used.

Method

Six children with CI, in grades 1–3 participated in the study (one child in grade 1, three children in grade 2, and two children in grade 3). All six children had severe to profound HI with prelingual onset and had received their implants at between 1;7 and 3;7 years of age. Five of them had bilateral implants. Forty-three children with NH constituted the comparison group (12–16 children in each grade). The children were tested in 2 separate 50 min sessions at their school. Some of the children with CI were tested at a regular follow-up at their respective paediatric cochlear implant program. The same cognitive tests as in Papers I and II were used with the following additional tests of phonological skills: *Phoneme Identification,* *Phonological Output* assessed in a picture naming test, and *Phonological representations*, measured in a task which required recognition of mispronounced words. Reading ability was measured in the tests presented in Paper I, *TOWRE* (decoding of words and nonwords) and Woodcock *Reading Mastery test – Revised* (reading comprehension).

Results & Discussion

Out of the working memory measures, the children with CI had most problems with phonological WM but had relatively less difficulties when tasks with shorter and suprasegmentally less complex test items were used. Four out of six children with CI performed comparable to their respective grade-matched comparison group on the measure of general WM. All six children with CI performed on par with their comparisons group on the measure of visuospatial WM.

Their phonological skills varied considerably between the tests with relatively higher performance levels on the measures of output phonology, phoneme segmentation, phonological representations, and the response latency measure of the nonword discrimination test. The results suggest that the children with CI in the present study have fairly distinct phonological representations for phonological input of words, which may result from large amounts of exposure and practice. The results also indicate that this group of children are relatively skilled at manipulating familiar words for which they have distinct phonological representations, although orthographic representations may have been used to improve performance in some of the tests.

Lexical access performance also varied between tests, five out of six children had a performance equivalent to their normal hearing controls in the Semantic Decision task, whereas only two and three children, respectively, performed on this level in the Wordspotting and Passive Naming tests. These findings may reflect that children with CI use phonological and semantic representations of words in long-term memory when solving these types of tasks and that they, to a greater extent, use top-down processing strategies to compensate for distorted auditory perception.

Decoding skills were age appropriate for all six children, for decoding of words as well as nonwords. Reading comprehension was comparable to that of the NH comparison group for four out of six children. The two children with poorer reading comprehension scores had a poorer performance than both their comparison groups and the other children with CI on the measures of general and phonological WM and most of the measures of phonological skills and phonological decoding. This suggests that they use orthographic decoding to compensate for poorer phonological skills.

The demographic variables seemed to provide explanations to some of the variance in performance on the cognitive tests, i.e. the children with the highest performance in most of the cognitive tests were diagnosed and implanted with their first CI at an early age whereas the two children who had relatively poorer performance were diagnosed and implanted later. Similarly, the two children with the highest performance in the cognitive tests were the only participants in this study who were integrated in mainstream education and used oral communication only, both at home and at school.

General Discussion

Summary Results

Generally the children with CI had poorer performance than the children with NH on measures of phonological and general WM, lexical access and phonological skills. These findings were largely expected with reference to previous research within this field (for results on phonological working memory, the reader should consult Burkholder & Pisoni, 2003; Dawson, Busby, McKay, & Clark, 2002; Pisoni et al., 2008; Spencer & Tomblin, 2009, for results on general working memory see Burkholder & Pisoni, 2003; Willstedt-Svensson et al., 2004; for results on phonological skills, see Ibertsson, Willstedt-Svensson, Radeborg, & Sahlén, 2008; James et al., 2008; Schorr et al. 2008; Spencer & Tomblin, 2009, and for results on lexical access see Young & Killen, 2002; Schorr et al., 2008; Spencer & Tomblin, 2009).

What is new?

This thesis contributes to previous research in that we can demonstrate more nuanced results on specific aspects of working memory, phonological skills and lexical access. The differences in cognitive performance between children with CI and children with NH were not significant for all children or subgroups of children, neither for visuospatial working memory, general working memory, phonological skills nor for lexical access, in study II and study IV.

Visuospatial working memory

The children with CI performed similar to their hearing controls on the measure of visuospatial WM in both study II and study IV. These findings are new since very little research has investigated visuospatial WM in children with CI and those studies that we know of have found different patterns of results (cf. Cleary, Pisoni & Geers, 2001, for results indicating lower levels of visuospatial WM in children with CI; see also Dawson, Busby, McKay, & Clark, 2002, for similar results as found in the present studies).

Phonological and general working memory

Most of the children with CI in study II and study IV had relatively higher general working memory capacity compared to their phonological working memory performance. A larger proportion of children performed within the normal range for hearing children on the general working memory test, than in the phonological working memory test, which is in line with previous findings (Burkholder & Pisoni, 2003; Sahlén et al., 2008). Phonological WM performance was further relatively higher when the test items to be recalled were shorter and suprasegmentally less complex.

Phonological skills

Performance in the tests of phonological skills varied considerably between measures and between children. The accuracy measure of the Nonword Discrimination test was significantly poorer for children with CI than for their controls (study II & IV), whereas their response latencies (for correct responses) in this test were not significantly slower.

Study IV, in which phonological skills were studied in further detail for six children, indicated relative strengths in tests of phonological skills in which real words, in contrast to nonwords, were used as test items, i.e., measures of output phonology, phoneme segmentation, and phonological representations. In some tests of phonological skill, some children further performed within the normal range for both accuracy and response latency measures.This pattern of results has not to our knowledge, been found in previous research.

Lexical access

In contrast to those previous studies on children with CI, which have measured lexical access in speech production (Schorr, 2008; Spencer & Tomblin, 2009), study II and study IV in the present thesis measured lexical access in speech recognition. Our results show that the relative level of performance in tasks of lexical access in speech recognition depends on the type of task and measure used. Performance in the tasks of lexical access also varied between children. A larger proportion of children in study II (around 50%) performed within the normal range (1 Standard Deviation) on the response latency measures for correct responses compared to their performance on the accuracy measures (which was found to vary between 20–33%). In contrast, 4–5 out of the six children in Study IV performed within the normal range on the latency measures, and between one and five out of six children performed within 1SD of their comparison group on the accuracy measures.

Performance in the Semantic Decision making test, in which semantic context may improve performance to some extent, was also relatively higher than in the two other tests of lexical access. That is, 33% of the children in Study II, and five out of six children in Study IV, performed within 1SD of their hearing controls on the accuracy measure of this test. Schorr et al. (2008) found similar results on lexical access in speech production. The finding that performance varied depending on the measure used is further in line with results from Spencer & Tomblin (2009) on speech production. In their study, rapid naming of letters was superior to rapid naming of digits. Their study, however, did not reveal significant differences between children with CI and children with NH on either measure of lexical access. Furthermore, our results revealed that subgroups and individual children did not constantly perform significantly below the level of their comparison groups, on neither accuracy nor latency measures of lexical access.

Demographic factors

The relations between demographic factors and cognitive skills varied somewhat between the studies. In study II, only one of the demographic variables was related to cognitive performance, i.e., children who mainly used oral communication had higher performance on a measure of lexical access and phonological WM. Demographic factors seemed more closely related to cognitive performance in study IV. However, these inferences are drawn based on visual analysis of individual data. Early age at diagnosis and implantation was in this study associated with higher cognitive test scores, which has also been found in previous research (e.g., Geers et al., 2008; Holt, Svirsky, Neuburger, & Miyamoto, 2004; McDonald Connor & Zwolan, 2004; Schorr et al., 2008). The two children with the highest cognitive test performance in this study were further the only participants in this study who were integrated in mainstream education and used oral communication only, both at home and at school (cf. Geers et al., 2002; 2008).

Reading skill in children with cochlear implants

A relatively large proportion, more than 60% of the children in studies III and IV, had reading comprehension within the normal range. This result is similar to findings from previous research (Dillon & Pisoni, 2006; Geers, 2003; Marschark et al., 2007, Fagan, Pisoni, Horn & Dillon, 2007). Geers et al. (2008) also found similar results for children in grades 1–3, but a follow-up study indicated that this proportion was smaller in high school. The individual children in study IV who had the highest reading comprehension scores were diagnosed and implanted earlier than the other children and mainly used oral communication at home and at school, which is in line with previous findings (Marschark et al., 2007; McDonald Connor & Zwolan, 2004). The two children with poorer reading comprehension in study IV also had poorer performance in tests of phonological WM, phonological skills and phonological decoding which is in accordance with findings from Dillon & Pisoni (2006).

The new findings related to reading ability in children with CI in this thesis is that general working memory was related to reading comprehension in both studies III and IV. Furthermore, the group analysis in study III did not indicate any relations between reading comprehension and demographic variables, which have been found previously (Marschark, et al., 2007; McDonald Connor & Zwolan, 2004). Decoding skills were age appropriate for all six children in study IV, both for decoding of words and nonwords. These results are in contrast with findings from Sahlén et al. (2008).

Reading and orthographic learning in children with normal hearing

In line with previous research (Cunningham, 2006; Cunningham et al., 2002; Nation et al., 2007), the findings from Study I indicate that the ability to acquire orthographic representations of words is closely associated with both decoding and reading comprehension. Our results further reveal that visuospatial and phonological working memory are related to orthographic learning ability, a finding which has not, to our knowledge, been demonstrated previously (cf. Castles & Nation, 2006; 2008; Cunningham et al., 2002). We also found moderate correlations between decoding skill and both visuospatial and phonological WM in children with NH in grades 1–3. These findings are new in that previous studies which have found relations between visuospatial working memory and reading skill are scarce and visuospatial working memory has, thus, been considered to play a minor role in reading ability.

Phonological working memory - a specific problem

The pattern of results from the studies in this thesis indicates that phonological working memory is a problematic area for children with CI. Burkholder & Pisoni (2003) has suggested that lower phonological working memory capacity in children with CI may partly be a consequence of reduced speed of articulatory rehearsal in the phonological loop according to Baddeley's model (e.g. Baddeley, 2003; Repovš & Baddeley, 2006). Our findings replicate and extend the findings from Burkholder & Pisoni (2003) such that the children with CI have generally lower phonological working memory performance than the children with NH but they have relatively less problems in tasks with shorter and less complex test items. That is, longer test items are generally more dependent on rapid subvocal rehearsal in order to prevent them from decay or displacement (Baddeley, 2003; Repovš & Baddeley, 2006). Lower levels of phonological working memory capacity in children with CI may further result in impaired performance in tasks where phonological working memory is a task demand. This is obvious in tasks where phonological information has to be simultaneously maintained and processed in working memory. Examples of such tasks may be tests typically used to measure phonological skill, such as, sound categorization, the nonword discrimination test used in studies II, III and IV (Oakhill & Kyle, 2000), and the phoneme segmentation test used in Study IV. Performance may, further, be particularly hampered when new words or nonwords are processed. This is because the retention of phonological information, for which there are no phonological representations in long-term memory, is more demanding on phonological working memory resources than retention of real words (Baddeley, 2003; Gathercole, 2006a). Recent results indicate that this is particularly evident in children with CI (Lyxell et al., 2008; Lyxell et al., 2009). Measures of phonological WM as measured by word span and phonological skills (sound categorization) have been found to correlate, although not strongly, in children with NH (Oakhill & Kyle, 2000). It has further been suggested that phonological skills contribute to the development of phonological WM but that individual differences in WM span do not explain variances in phonological sensitivity (Kail, 1997; Ferguson & Bowey, 2005). In children with CI, this could, however, be the case. Lower phonological working memory capacity may be a causal factor complicating general working memory (i.e., storage and processing) performance in these children when phonological information is to be processed. That is, even if the children are relatively efficient at processing information within working memory, problems with subvocal rehearsal prior to or after processing may result in poorer recall, and thus impair general working memory capacity.

It is also possible that the recognition of phonological information at the phoneme level is highly demanding on processing capacity for children with CI, even in tasks typically used to measure storage only of phonological working memory. If that is a valid explanation, the processing component of working memory (phonological and general) could be the limiting factor in tasks requiring storage and processing of phonological information. That alternative hypothesis would fit our findings with relatively strong correlations between general and phonological working memory in the children with CI (but not in children with NH) in Study II. The various theories of working memory may also provide alternative explanations to these correlations. According to the resource-sharing hypothesis (e.g., Daneman & Carpenter, 1980; Daneman & Hannon, 2007), the processing component of the general working memory task would limit the ability to store information since a trade-off has to be made between processing and storage (Daneman & Hannon, 2007). If our measures of phonological working memory tax storage only of phonological information, and the complex WM task measures both storage and processing, the relationship between these tests in children with CI may not readily be explained by this model.

The multi-component theory (Baddeley, 2003; Repovš & Baddeley, 2006), and possibly also the task-switching hypothesis (Towse, Hitch, & Hutton, 1998; 2000), of working memory seem to more easily fit our pattern of results. According to the former theory, the phonological information to be processed would need to be held in the phonological loop in order to be processed by the central executive. Thus, impaired function in the phonological loop would result in loss of information to be processed. Furthermore, intact function in the phonological loop but lower capacity of the central executive might result in poorer outcomes in tasks of general working memory (Repovš & Baddeley, 2006). This theory thereby may, at least partly, provide an explanation for the specific impairments in tasks requiring storage of phonological information, found for the children with CI in this set of studies. According to the task-switching approach, general working memory span reflects the gradual degradation of memory representations during the time spent on the cognitive processing task rather than as a function of processing intensity (Towse, Hitch, & Hutton, 1998; 2000). Consequently, similar to the multi-component model of working memory, the task-switching account acknowledges independent mechanisms affecting storage and processing within working memory (Towse, Hitch & Hutton, 1998; 2000). This model, thus, might explain different capacities, and impairments, in storage and processing within working memory.

In the measure of visuospatial working memory used in the present studies, the children were required to replicate a pattern of previously filled cells in a matrix. If, as demonstrated by Miyake et al. (2001), measures of storage only and measures of storage and processing are equally strongly related to executive functioning in the visuospatial domain, we may consider our measure of visuospatial working memory to tap general working memory of visuospatial information to a certain extent. Results indicating a relatively strong correlation between measures of storage only and storage and processing of visuospatial information, was also found by Alloway, Gathercole, & Pickering (2006) for children, 4–6 years of age. Following this line of reasoning, the high levels of visuospatial working memory demonstrated by the children with CI in the present studies would suggest that they may have similar general working memory capacity as their hearing peers.

The empirical findings in the present studies - that phonological working memory is a specifically problematic area for children with CI, which may in turn have consequences for other types of information processing tasks, such as, general working memory capacity or phonological skills is also supported by other researchers (Burkholder & Pisoni, 2003; Pisoni et al., 2008). Possible causal factors for these findings may be less efficient subvocal rehearsal processes or, alternatively, that recognition of phonological information is highly demanding on processing capacity for children with CI, thereby impairing their capacity to store information. The present set of data, also indicates that the problems with phonological working memory in children with CI were less pronounced for shorter and less complex test stimuli. This would provide support for the subvocal rehearsal hypothesis.

Another important finding from the present studies was that results on phonological working memory were obtained from two measures, commonly used to assess phonological working memory in populations with normal hearing. Spencer and Tomblin (2009) reported floor effects for children with CI on these tests, which were consequently not recommended as valid measures of phonological WM or, in the case of their study, phonological processing. However, Spencer and Tomblin (2009) used binary scoring methods which may explain their pattern of results. The present studies used more sensitive scoring criteria where the children received credits for correctly reproduced consonants and suprasegmental accuracy, which did not result in floor effects. The findings that our children with CI performed on par with their hearing peers on the visuospatial working memory measure is intriguing since this pattern of results has not been consistently found in previous studies (cf. Dawson et al., 2002; Cleary et al., 2001).

Phonological skills

As mentioned in the introductory section, phonological skills is a broad concept including a variety of different components and processes (e.g., Anthony et al., 2002; Anthony & Francis, 2005; Dally, 2006; Kjeldsen et al., 2003; Oakhill & Kyle, 2000; Snowling et al., 2003). The present studies focused on one specific aspect of phonological skill, i.e., the phonological representations of words and phonemes in children with CI.

The reason for studying this subcomponent of phonological skill is that phonological representations are important for the development of phonological and linguistic skills in children with NH (Boada & Pennington, 2006; Elbro & Nygaard Jensen, 2005). Hearing children generally have well specified phonological representations for familiar words at an early age (Swingley, 2003; Swingley & Aslin, 2000). However, the phonological representations that children with CI acquire, may be different and possibly not as distinct, as a consequence of the auditory stimulation from the cochlear implant and early auditory deprivation before implantation (Harnsberger, 2001; Pisoni et al., 2008). Increased knowledge about phonological representations in children with CI is therefore fundamental for understanding the development of other aspects of phonological skills in this population. The results from studies II and IV indicate that children with CI have specific problems in tasks of phonological skills which use nonwords as test items. As discussed above, these problems may be attributed to impairments in phonological working memory. However, they may also reflect the fact that the children have no established phonological representations for nonwords. Consequently, processing may not be facilitated by the availability of representations from long-term memory when nonwords are used as test items. The results from the individual children in Study IV corroborate this interpretation since their performance in tasks that require discrimination of phonemes in real words was considerably higher (often similar to that of children with NH) than when nonwords were used. At this point it should, however, be noted that a few of the children with CI in study IV performed at the level of NH children even in the relatively more demanding tasks on phonological skills, where nonwords were to be processed.

These results indicate that children with CI may have relatively distinct phonological representations for real words that are highly familiar to them (cf. Boada & Pennington, 2006; Elbro & Nygaard Jensen, 2005; Swingley & Aslin, 2000; Swingley, 2003). This is a promising finding since distinct phonological representations are fundamental for other aspects of phonological skill and reading development (e.g., Elbro & Nygaard Jensen, 2005).The process of refinement of phonological representations may be slower and more dependent on abundant information for this population of children than for children with NH. Thus, rich linguistic stimulation at an early age is extremely important for children with CI as they may, just like children with poor phonological learning abilities, as proposed by Gathercole (2006a, p.515), "with time and sufficient exposure…succeed in forming stable lexical representations of the sound of a new word".

The relationship between phonological skills and acquisition of reading and spelling skills is reciprocal (Castles & Coltheart, 2004). That is, phonological skills improve reading and spelling ability and the latter two skills are also means for improving phonological representations and, as a consequence, other aspects of phonological skills (Castles & Coltheart, 2004). As suggested by Spencer & Tomblin (2009), this reciprocal relationship may be more evident for some children who use CIs than for children with NH. That is, a child who uses a CI may not be able to perceive all the phonemes of a word but comes to realize that some phonemes are included in a word only after seeing the word in print (Spencer & Tomblin, 2009). Thereby, their ability to “reflect on spoken words” may be improved by learning to read and write. This reciprocal relationship between phonological skills and reading may also go in the opposite direction even for children with CI. This is most evident for two of the children in Study IV, who seem to have more developed phonological representations, even for speech sounds in non-words. These representations may, in turn, be beneficial for their reading development (Castles & Coltheart, 2004).

Learning to read and spell, however, also improves the orthographic representations of words (Castles & Coltheart, 2004; Nation, Angell, & Castles, 2007; Share, 2004). Thus, once children have acquired orthographic skills, they may use these skills, either in addition to or instead of their phonological skills to solve phonological awareness tasks (Castles & Coltheart, 2004). Perhaps it is not a reckless speculation that the acquisition of orthographic skills provides effective strategies for reading acquisition in children with CI. That is, reading by means of orthographic strategies may improve their phonological representations and general language skills for both oral and written language (cf. Castles & Coltheart, 2004).

Lexical access

The present thesis assessed lexical access in speech recognition (Gaskell & Marslen-Wilson, 2002; Norris, Cutler, McQueen, & Butterfield, 2006) as opposed to lexical access in speech production (Goldrick & Rapp, 2007; Navarrete & Costa, 2005; Schwartz et al., 2006). This is an area that, according to our knowledge, has not been investigated previously in this population of children. The combined empirical results from the studies in the present thesis reveal that the children with CI had lower levels of correct identification of auditorily presented words compared to the children with NH. Furthermore, a relatively higher proportion of children performed within the normal range when semantic information was provided.

An additional inspection of data after the publication of the results revealed that the children with CI, in most cases, had longer response latencies for incorrect answers, or had no incorrect - only omitted- responses. This analysis indicates that they did not simply respond by chance, i.e., quick rather than correct for all test items.

Examples of the factors that should be most important for lexical access in speech recognition, both for children with CI and children with NH, are distinct phonological representations of words in long-term memory (Elbro, Borstrom, & Klint Petersen, 1998;

Norris et al., 2006), stable auditory perception (Harnsberger, 2001; Pisoni et al., 2008), familiarity with the word (Cuetos, Alvarez, González-Nosti, Méot & Bonin, 2006; Gershkoff-Stowe, 2002; Goldrick & Rapp, 2007), and the semantic context in which the word occurs (Aydelott, & Bates, 2004; McClelland, Mirman, & Holt, 2006; Obleser, Wise, Dresner, & Scott. 2007). We do not know the relative contribution of each one of these factors. A reasonable assumption is that they all interact to influence lexical access in children with CI. For example, the words for which the children can provide the correct responses may be easier to perceive with the implant, i.e., auditory perception influences speed of lexical access for correct responses. As a consequence, the phonological representation of that word will be more distinct, a factor which will also, in turn, improve lexical access.

The findings that the words which children with CI could access correctly were often quickly activated suggest, however, that in some cases the quality of the auditory input signal and the phonological representation must have been "good enough" for the process of matching between input and the phonological representation to be smooth and efficient. Factors such as familiarity, age of acquisition, semantic context, etc., certainly affect this process, although we do not know to what extent. The children's performance level in the semantic decision task was, however, relatively higher than in the other tests of lexical access compared to hearing peers. Since the Semantic Decisions test was the only test of lexical access that provided a semantic context, this result indicates that semantic context improves lexical access performance substantially in children with CI. No such effects were found in the NH children, since they performed, and should be expected to perform at ceiling in all three tests of lexical access. This finding was expected, as factors such as familiarity and context are known to improve speech recognition, particularly when the input signal is degraded or when the words are ambiguous (Aydelott, & Bates, 2004; McClelland, Mirman, & Holt, 2006; Obleser, Wise, Dresner, & Scott, 2007), when the phonological representation of a word is not particularly distinct, or when the input signal is of poor quality (cf. Cuetos, Alvarez, González-Nosti, Méot, & Bonin, 2006; Gershkoff-Stowe, 2002; Goldrick & Rapp, 2007), which may often be the case for children with CI (Harnsberger, 2001; Pisoni et al., 2008; Svirsky, Robbins, Iler Kirk, Pisoni, & Miyamoto, 2000). Thus, the use of such top down processes are always important for children with CI, just like for others who have a hearing impairment and for whom the auditory input signal is often compromised, due to, for example, lower frequency resolution compared to normal hearing.

In the study by Spencer and Tomblin (2009) on lexical access in speech production, rapid naming of letters was superior to rapid naming of digits for the children with CI, but not for the children with NH. The authors' interpretation was that the children with CI were relatively more skilled at retrieving the phonological representations associated with letters than with numbers. They further hypothesized that reading experience and letter-learning activities at school may have improved these children's phonological representations for letters compared to digits. The findings from Spencer and Tomblin (2009), thus, suggest that distinctness of phonological representations and familiarity with the test items contribute to lexical access in children with CI. Lexical access in a semantic context was not assessed in their study.

The empirical results on lexical access in speech recognition demonstrated in this thesis are in line with the findings from Schorr et al. (2008). These authors measured lexical access in speech production and found children with CI to have lower levels of performance than children with NH. Contrary to these results, Spencer and Tomblin (2009) did not find any significant differences between children with CI and children with NH on any of their measures of lexical access in speech production. Of course, lexical access in speech recognition, which was assessed in the present thesis, may be a relatively more difficult task for children with CI than lexical access for speech production, since the former ability is dependent on auditory perception. On the other hand, the present results were not confounded by the processes involved in phonological output and articulation, which, in turn, increased validity of our response-time measure. The findings from this thesis and the studies discussed above provide some support to the claim that children with CI may have relatively efficient processes of lexical access, for both speech production and speech recognition, when the quality of the phonological representation and the auditory input signal are high enough. Their performance is also improved when they are familiar with the words which are to be accessed and when the semantic context provides cues to which words are more plausible.

Not to forget, a few children with CI even performed similar to their hearing comparison groups on both accuracy and latency measures of lexical access, even in two of the conditions that did not provide semantic context. These findings are encouraging and motivate further research on how to improve lexical access in children with CI. Intervention programs including training of phonological representations, through reading and play with the phonological structures of words, may help children with CI to improve the quality of their phonological representations. Improved phonological representations would in turn, not only be beneficial for lexical access but also for phonological skills and reading (Kjeldsen, Niemi, & Olofsson, 2003; Moriarty & Gillon, 2006). Furthermore, top down strategies, e.g., the use of semantic context and familiarity, may be improved by intervention directed at increasing vocabulary size and improving general world knowledge in children with CI.

The pattern of results on cognitive ability found in the present studies, with relatively higher performance in tests which use real words as test materials, indicate that children with CI use top-down strategies and long-term phonological representations when solving cognitive tests presented in auditory modality. Cognitive performance in children with CI may thus not be reduced to the level of auditory perception.

Reading skills

Reading comprehension

Over 60% (63–66%) of the children with CI in the present studies had reading comprehension scores within the normal range for hearing children. Slightly higher levels of reading comprehension (75% of children with CI within normal range) were found by Lyxell et al. (2008).The fact that different tests of reading comprehension were used (within study III, depending on age) and between the studies (III and IV) may also improve the reliability of these findings. Although similar results have been reported in previous research (Dillon & Pisoni, 2006; Geers, 2003; Marschark et al.,2007), these results are unexpectedly high based on the children's general levels of phonological skills, lexical access and working memory. Furthermore, the results of reading skill found in the present studies are superior to those typically reported in children with profound hearing impairment who have not received a CI (e.g., Paul, 2003). In a longitudinal study by Geers et al. (2008), the proportion of children with reading comprehension within the normal range had decreased substantially by the time the children had reached the age of 14–16 years. The participants in our studies were younger (between 6 and 13 years of age). A follow-up study of their reading comprehension ability should, ideally, be conducted within a few years in order to find out whether the developmental pattern found by Geers et al. (2008) is similar in our group of children.

The finding that general working memory was related to reading comprehension in Study III and Study IV, may have been expected based on a vast amount of research in other populations (Baddeley, 2003; Cain, Oakhill, & Bryant, 2004; Gathercole et al., 2005; Goff et al., 2005; Oakhill, Cain, & Bryant, 2003; Savage et al., 2006). Our findings are, however, particularly important since our measure of general working memory required processing and storage of phonological information, presented in the auditory mode. The ability to simultaneously process and store information therefore appears to be an important component in reading comprehension also in children with CI. The correlation coefficients (around $r=.7$) further suggest that this relationship may be stronger in children with CI than in children with NH and typical development (cf. Cain, Oakhill, & Bryant, 2004; Oakhill, Cain, & Bryant, 2003). These findings also add to the growing body of empirical support regarding the importance of general working memory in individuals with hearing impairment. For example, results from Foo, Rudner, Rönnberg, & Lunner (2007) indicate that general working memory, as measured by reading span (Daneman & Carpenter, 1980) is a strong predictor of speech recognition in noise for elderly persons with mild to moderate hearing impairments, using traditional hearing aids.

The associations between reading comprehension and the demographic factors varied between the studies (III and IV). The individual children in study IV who had the highest reading comprehension scores were diagnosed and implanted earlier than the other children and used oral communication as their main communication mode at home and at school. These results are in line with previous findings (Marschark et al., 2007; McDonald Connor & Zwolan, 2004). The group analysis in study III, on the other hand, did not demonstrate any relations between reading comprehension and demographic variables. This variation in results may not come as a surprise, since a number of factors may affect reading comprehension in children with typical development, e.g., decoding skill, vocabulary and verbal skills (e.g., Cain, Oakhill, & Bryant, 2004; Seigneuric et al., 2000), prior knowledge (Cain, Oakhill, & Bryant, 2004) and the accessibility of this information from long-term memory (Hannon & Daneman, 2001a; 2001b), inference making (Hannon & Daneman 2001a; 2001b), knowledge about story structure, working memory (Oakhill, Cain, & Bryant, 2003) and motivational factors (e.g., Seigneuric et al., 2000). Thus, a relatively large proportion of children with CI have reading comprehension skills within the normal range of children with NH.

Reading comprehension ability in the children with CI in the present studies was further associated with general working memory capacity, perhaps even more strongly than in children with typical hearing.

Decoding skills

Decoding of words and nonwords were, in contrast to the findings from Sahlén et al. (2008), age appropriate for the six children in study IV. Some researchers suggest that children with CI use phonological decoding strategies in reading (Pisoni et al., 2008). Although we only had access to decoding results from six children, we could observe a trend in the empirical pattern, such that the children with CI in study IV were displaying larger differences between word decoding and nonword decoding than the hearing children. Results from Ibertsson et al., (2009) demonstrating teenagers (14–19 yrs of age) to have age-appropriate performance in reading tasks measuring orthographic decoding skill and slightly lower levels on phonological decoding measures, point in the same direction. This pattern of results may reflect the problems with phonological skills and phonological working memory that these children experience. Speculatively, children with CI may use orthographic decoding strategies more frequently than children with NH, as a means of compensating for their problems with storage and processing of phonological information.

The findings from study I, that orthographic learning was associated with phonological and visuospatial working memory in children with typical hearing, are intriguing and may have consequences for our understanding of orthographic reading skills, both for children with NH and for children with CI. A possible interpretation of these results is that visuospatial and phonological working memory affects orthographic learning through their effect on phonological decoding (cf. e.g., Bishop et al., 2004; Griffiths & Snowling, 2002; Roodendrys & Stokes, 2001). This explanation would be plausible since phonological decoding skill is considered to be the most important prerequisite for orthographic learning (Share, 2004) in children with NH. It may not be equally plausible for specific populations of children, who do not have a typical development of phonological skill or phonological working memory, such as the children with CI in our studies, and children with SLI or dyslexia.

The relations between visuospatial working memory, the decoding measures (non-word decoding, and orthographic choices), and orthographic learning, found in the children with NH in Study I are particularly interesting since relations between visuospatial working memory and reading and spelling ability have not been found in a consistent manner in previous research. This may suggest that the effects of visuospatial working memory are particularly important somewhere in the process of acquiring orthographic representations and not in orthographic reading per se. Findings from Predovan et al. (2009) that brain areas involved in visuospatial working memory show higher levels of activation in retention of the orthographic representations of non-words compared to real words would provide support for this line of reasoning.

Possible effects of visuospatial working memory may further be particularly important in the acquisition of orthographic representations for children with CI. That is, visuospatial working memory may, speculatively, have a role in compensatory strategies to compensate for lower levels of phonological skills and phonological decoding ability.

A potential explanation for the relationships between visuospatial and phonological working memory and orthographic learning found in the children with NH may be that these two working memory factors alleviate the process of transferring visual and phonological representations of words into long-term memory, as suggested by the multi-component model of working memory (Repovš & Baddeley, 2006). Specifically, a high capacity to rehearse information in the phonological loop of the multi-component model of working memory has been suggested to improve the transfer of this type of information into long-term memory (Baddeley, 2000). The phonological loop in working memory is therefore considered to be a tool for vocabulary acquisition (Baddeley, Gathercole, & Papagno, 1998; Gathercole, 2006a). Similarly, efficient processing in the visuospatial sketchpad should be important for the transfer of orthographic representations into long-term memory. Such processes may have the potential to explain the relatively large differences between orthographic and phonological decoding skills (as compared to the children with NH) found in children with CI in study IV. The integration of information from different modalities is suggested to occur in the episodic buffer of working memory (Baddeley, 2003; Repovš & Baddeley, 2006), and this multimodal information may subsequently be transferred into long-term memory. In the case of orthographic learning, the visual and phonological representations of a particular word might be integrated into a multi-dimensional code in the episodic buffer of working memory before entering long-term memory.

Our results for the children with CI indicate that they may use orthographic reading strategies to a higher extent than their hearing peers. That is, they had larger differences in decoding performance for words compared to nonwords than the children with NH. Recent results (Asker-Árnason, in press; Ibertsson et al., 2009) indicating that children with CI have similar or higher spelling skills than children with NH may provide further support for the suggestion that children with CI have particularly distinct orthographic representations. Distinct orthographic representations would, in turn, allow these children to use orthographic reading strategies to a larger extent than hearing children. That is, a single internal orthographic lexicon is used for both reading and spelling (Conrad, 2008; Burt & Tate, 2002; Shahar-Yames & Share, 2008). The fact that the children with CI also had typical visuospatial working memory capacity may motivate the question whether children with CI use their visual working memory capacity to acquire orthographic representations in another way, and at an earlier stage of reading development than children with NH. The present studies did not investigate orthographic learning in children with CI but this area necessarily has to be addressed in future studies.

The use of orthographic reading strategies could be beneficial for reading skill in children with CI since it may allow for rapid and efficient reading despite low levels of phonological skill. However, skilled readers should be able to rapidly shift between orthographic and phonological reading strategies when necessary (Gustafson, 2000; Jackson & Doellinger, 2002), for example when orthographically unfamiliar words are encountered in a text passage, and when reading inflections of familiar words. Phonological decoding problems in those kinds of situations could hamper reading comprehension for skilled orthographic decoders and may be a factor behind the decreased reading comprehension skills found by Geers et al. (2008) in older children with CI.

Suggestions for future research

The present thesis intended to assess cognitive skill in children with CI. The fact that relatively little research has focused on these aspects of implantation in children, in combination with the characteristics of the population (as being small and heterogeneous), led us to adopt descriptive research strategies in the empirical studies. The suggestions for further research presented below are more specific research questions that have emerged along the research process, from the results and from experiences in the test situation.

One important issue concerns the nature of phonological representations in children with CI. The fact that children with CI often have short response times for correct responses may indicate that they have relatively distinct phonological representations for the corresponding words. In order to investigate this issue, it would be important to study the development of these representations over time, as well as the influence of semantic information on the relations between speed of lexical access and quality of phonological representations. It is also important to study possible effects of intervention in order to improve the quality of phonological representations in children.

The development of various aspects of working memory over time, i.e., specific components of storage and processing and to what extent the type of information to be stored and processed (semantic, linguistic vs. non-linguistic, visual, spatial etc.) has an impact on performance in this population, are questions that arise from the present findings and which deserve further research.

It would also be valuable to perform extensive studies on the development of phonological skills in children with CI, with a broad focus both at the level of discrimination and at a meta-level, involving the manipulation of phonemes and syllables in words and nonwords. The relations between these skills and other aspects of language, e.g., vocabulary, grammatical skills, et c., have not been assessed in the present studies but would be important to explore, as well as the benefits of audiovisual cues and lip-reading on cognitive performance.

The results from the present thesis may indicate that children with CI are more likely to use orthographic reading strategies than children with NH. This hypothesis needs to be tested in larger populations of children with CI and necessarily should include an assessment of orthographic learning in children with CI in order to find out more about their reading processes and strategies, which, obviously, are more efficient than we would expect based on their performance in tasks of working memory, phonological skills, and lexical access.

The above mentioned suggestions for further research are only a few examples of issues that need to be investigated in order to come closer to understanding the cognitive effects of cochlear implantation in children. We should, however, always remember that language development and other aspects of development in this population of children are not only dependent on cognitive factors. That is to say, communication and language develop in interaction with other people. The outcome of implantation is therefore, to a great extent, settled by those significant others, together with factors like self-confidence, that are important for all children, regardless of whether they have a hearing impairment or not.

Sammanfattning på svenska

Denna avhandling studerade kognitiva förmågor hos barn med grav hörselskada eller dövhet som fått cochleaimplantat (CI). Auditiv stimulering från CI i tidig ålder påverkar de flesta kognitiva funktioner som en följd av hjärnans plasticitet hos små barn. Det är viktigt att förstå de kognitiva konsekvenserna av auditiv stimulering från CI för att kunna ge dessa barn bästa möjliga stöd. Avhandlingen undersökte tre specifika aspekter av kognitiv förmåga (arbetsminne, fonologiska förmågor och lexikal aktivering), samt läsförmåga hos barn med CI, i jämförelse med barn med normal hörsel i samma åldrar. Relationerna mellan kognitiva förmågor och läsförmåga studerades också, liksom sambanden mellan demografiska faktorer (t ex implantationsålder och kommunikationssätt) och kognitiva förmågor samt läsfärdigheter. Barnen med CI hade generellt lägre prestationsnivå än barnen med normal hörsel i uppgifter som mäter fonologiskt och generellt arbetsminne, fonologiska förmågor och lexikal aktivering. De hade specifika problem i uppgifter som i hög grad belastar fonologiskt arbetsminne, medan deras visuospatiala arbetsminneskapacitet var jämförbar med den hos barnen med normal hörsel. Majoriteten av barnen med CI hade läsfärdigheter i nivå med normalhörande barn, för både avkodning och läsförståelse. Sambanden mellan demografiska faktorer och kognitiva förmågor och läsförmåga varierade mellan studierna. Resultatmönstren diskuteras utifrån teorier om arbetsminne, fonologiska färdigheter och lexikal aktivering.

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