Speech and sign perception in deaf children with cochlear implants
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1 INTRODUCTION

1.1 THE TOPIC AND GOAL OF THIS THESIS

As of January 2010, more than 150,000 patients with severe-to-profound sensorineural hearing loss worldwide had received a cochlear implant (CI), more than half of which were children (De Raeve et al., 2009). The majority of the operations were successful, restoring some sense of hearing. Each year, over 350 cochlear implant operations are performed in the Netherlands and it is estimated that at least between 70-75% of congenitally deaf children in the Netherlands use a CI (De Raeve et al., 2009).

For pre-lingually deaf children, one of the most important functions of a CI is to support spoken language development. As a result, research in pediatric cochlear implantation mainly focuses on spoken language outcomes (Thoutenhoofd et al., 2005). When the first pre-lingually deaf children received a CI in the 1980s, it was unknown whether they would be able to acquire a spoken language from the relatively poor auditory input provided by the implant (Svirsky, Robbins, Kirk, Pisoni, & Miyamoto, 2000). In the meantime, several studies have shown that some children with a CI show similar, or even faster, rates of spoken language development compared to children with normal hearing of the same age (Schauwers, Gillis, & Govaerts, 2005). That is, despite initial delays in language development, some children with a CI are able to catch up with their peers with normal hearing within a few years after implantation. However, individual outcomes are highly variable and many different factors affect the benefits a child will obtain from the CI (Geers, Nicholas, & Moog, 2007).

Within the domain of spoken language outcomes, speech perception has received much attention for several reasons. Firstly, compared to the human ear, auditory processing through a CI is characterized by relatively poor spectrotemporal resolution (Shannon, 2002), thus posing a challenge for language acquisition. Secondly, as an index of auditory functioning, speech perception tests are frequently used in the clinical assessment of children with a CI (Mendel, 2008). Thirdly, speech perception abilities have been found to be a strong predictor of their expressive and receptive language abilities (DesJardin, Ambrose, Martinez, & Eisenberg, 2009; Sarant, Blamey, Dowell, Clark, & Gibson, 2001). Speech perception outcomes also have been shown to depend on a range of factors including factors related to the hearing loss such as age at onset of hearing loss, degree of hearing loss and age at implantation, but also more general factors such as nonverbal IQ (Geers, Brenner, & Davidson, 2003a; Sarant et al., 2001; Wie, Falkenberg, Tvete, & Tomblin, 2007).
Most studies of speech perception in children with a CI have included standardized word and sentence recognition tests and have not focused on the underlying processes in speech perception. Nevertheless, understanding how the nature of the auditory input affects the linguistic and cognitive processes relevant to speech perception will contribute substantially to explaining why some children do particularly well with their CI, whereas others do not (Pisoni, 2000). The first goal of this thesis is to enhance the understanding of speech perception in children with a CI by examining underlying processes in their perception of sounds and words. More specifically, we will examine whether they use acoustic cues in consonant and vowel perception differently from children with normal hearing and how such differences, if found, affect word learning.

The second goal of this thesis is directly related to another, more controversial, topic in the pediatric cochlear implantation literature, namely the effect of signed input on spoken language development (Geers, 2006; Nicholas & Geers, 2003; Spencer & Tomblin, 2006; Yoshinaga-Itano, 2006). Given that the main function of a CI in pre-lingually deaf children is to support spoken language development, it is not surprising that the majority of time and effort in their rehabilitation and education is aimed at fostering spoken language abilities. However, a CI does not restore normal hearing and the child is again deaf if the device is switched off or not functioning properly. Moreover, as already mentioned, not all children benefit from the CI to the same extent. Signed input has been suggested to have positive effects on spoken language development (e.g. Connor, Hieber, Arts, & Zwolan, 2000; Delore, Robier, Bremond, Beutter, & Ployet, 1999; Yoshinaga-Itano, 2006) as well as negative effects (e.g. Cullington, Hodges, Butts, Dolan-Ash, & Balkany, 2000; Geers et al., 2002; Pisoni, Cleary, Geers, & Tobey, 1999; Svirsky et al., 2000). Most of these studies compared children in different educational settings (usually ‘Oral Communication’ versus ‘Total Communication’) and, as will become clear in §1.4.2, their findings are often difficult to interpret. Only a few studies have adopted the more valid approach of comparing spoken and signed language abilities in the same children (Cassandro, Nicastri, Chiarella, Genovese, & Gallo, 2003; Coerts, Mills, Van den Broek, & Brokx, 1994; De Raeve et al., 2009; Nordqvist & Nelfelt, 2004; Yoshinaga-Itano, 2006). This thesis will therefore provide more insight into the effects of signed input on spoken language abilities by examining speech perception and sign perception in the same sample of children.

In sum, this thesis contributes to understanding how children with a CI use the implant to perceive and learn spoken language as well as understanding their

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1 The term ‘signed input’ is preferred to ‘sign language’ since many children with a CI are exposed to artificial sign systems or sign-supported speech, but not to the natural sign language used in the deaf community.
potential strengths and weaknesses in perceiving language in different modalities. A group of 15 children with a CI, 02 age-matched children with normal hearing and 21 young adults with normal hearing participated in a series of experiments targeting pre-lexical and lexical perception in speech and sign. Before we turn to a discussion of the research questions (Chapter 2) and the research methodology (Chapter 3), the remaining part of this chapter provides an overview of the technology, process, and outcomes of (pediatric) cochlear implantation.

1.2 Cochlear Implantation

Both children and adults with severe-to-profound sensorineural hearing loss are candidates for a CI. Details of the implantation procedure are different for children and adults, but more importantly their outcomes can be different given that onset and duration of deafness are strong predictors of implant benefit (Dunham & Limb, 2007; Fallon, Irvine, & Shepherd, 2008; Peterson, Pisoni, & Miyamoto, 2010). More specifically, speech recognition performance achieved by children who lost their hearing before the age of four or five years (i.e., pre-lingually) is often quite good and comparable to that achieved by post-lingually deaf adults who received a CI at a later age (e.g. Dowell, Dettman, Blamey, Barker, & Clark, 2002). In contrast, pre-lingually deaf children and adults who have experienced a long period of auditory deprivation (i.e., more than eight years) before implantation typically perform poorly (e.g. Teoh, Pisoni, & Miyamoto, 2004a; Waltzman, Roland, & Cohen, 2002b). This is due to the lack of plasticity in auditory cortical structures after the long period of auditory deprivation (Fallon et al., 2008; Teoh, Pisoni, & Miyamoto, 2004b). Similarly, age-related differences in auditory cortical plasticity explain why age at implantation is an important predictor of spoken language outcomes in congenitally deaf children (see §1.4.1).

Pediatric cochlear implantation is a multifaceted and multidisciplinary topic covering areas of research as diverse as engineering, neurobiology, medicine, audiology, psychology, clinical linguistics, education and ethics. Here, two main topics will be addressed: cochlear implant technology (§1.2.1) and current practice in pediatric cochlear implantation (§1.2.2).

1.2.1 Cochlear Implant Technology

A CI is an electronic hearing prosthesis that consists of an external component and a surgically implanted internal component (see Figure 1.1). The external component consists of a microphone, a speech processor, a magnetic radio frequency transmitter
and a power source. The speech processor and power source are either integrated with the microphone in a behind-the-ear processor or worn on the body. Externally fitted or built-in electromagnets allow wireless coupling to other devices such as assistive listening devices, cell phones and MP3 players, which feed output directly into the speech processor. The surgically implanted internal component consists of a magnetic radio frequency receiver and an electrode array that is inserted in the cochlea. Sound is received by the microphone and relayed to the speech processor, where the input signal is digitized, compressed and filtered. The signal is then transmitted transcutaneously via radiofrequencies to the internal component, where the signals are converted into electrical discharges at assigned electrodes through induction of an electrical field between the assigned electrode and a reference electrode placed usually on the outside of the cochlea. The electrodes directly stimulate the auditory nerve.

Figure 1.1. External and internal components of a CI.

2 Some adults have a dysfunctional auditory nerve and some children are born without an auditory nerve. Cochlear implants do not provide benefit for these patients, but auditory brainstem implants or auditory midbrain implants might (Moore & Shannon, 2009). These implants bypass the cochlea and are inserted in the cochlea nucleus (auditory brainstem implant) or the inferior colliculus (auditory midbrain implant). Preliminary results have shown moderate success of the auditory brainstem implant in post-lingually deaf adults (Moore & Shannon, 2009) and also in pre-lingually deaf children implanted at three and a half years of age (Sennaroglu et al., 2009). Results with the auditory midbrain implant have not been very positive as yet.
There are three major manufacturers of implant devices: MED-EL®, Cochlear® and Advanced Bionics®. CIs can vary in design, implemented speech processing algorithm, electrode array and number of electrodes (Dunham & Limb, 2007). The functioning of a CI can depend on the placement of the array inside the cochlea (depth and proximity to spiral ganglion cells) and the number of electrodes that can be used effectively. Current CIs use between 12 and 24 electrodes that span the frequency range between approximately 100 and 8000 Hz. However, the number of available electrodes far exceeds the number that are actively used, which lies between four and eight for current electrode array designs and for current positioning of the electrode array within the cochlea. This limitation may be due to interference between the electric fields from adjacent stimulating electrodes (Wilson & Dorman, 2008a). Current CIs are flexible in the number of active electrodes and stimulation rate to allow adaptation to the needs and preferences of individual users or different listening situations.

CIs have different speech processing algorithms. The most frequently used algorithms are: continuous interleaved sampling (CIS), spectral peak (SPEAK), advanced combination encoder (ACE), n-of-m and HiResolution (HiRes) strategies. CIS is the default algorithm for MED-EL® devices, ACE for Cochlear® devices and HiRes for Advanced Bionics® devices (Wilson & Dorman, 2008b). However, manufacturers sometimes use different algorithms.

The CIS algorithm is widely used and compatible with devices from all three manufacturers. It filters speech and other sounds using frequency band filters. After compression of the dynamic range, the algorithm directs the output of each filter to a single electrode. High frequency bands are assigned to electrodes at the beginning of the cochlea (base) and low frequency bands to electrodes at the end of the cochlea (apex), following the frequency-to-place mapping in the normal cochlea. Signals are continuously sampled by rapidly presented pulses interleaved across electrodes. The HiRes algorithm resembles the CIS algorithm, but uses higher stimulation rates and cut-off frequencies for the frequency band filters.

With SPEAK, n-of-m, and ACE algorithms, signals for the different channels are scanned before electrode stimulation. Stimulus pulses are delivered only to the electrodes that correspond to channels with the highest amplitudes. The number of chosen channels is fixed in n-of-m and ACE, but can vary in SPEAK, depending on the input signal. The main functions of channel selection in these strategies are to reduce interference between adjacent electrodes and to increase speech-to-noise ratios.

The development of new processing algorithms is largely directed at representing temporal fine structure information, that is, rapid high-frequency modulations in the acoustic signal (Wilson & Dorman, 2008b). Such information is especially important for speech perception under adverse conditions as well as for
the perception of melody in music. Current processing algorithms such as CIS, SPEAK and ACE were not designed to transmit fine structure information. Several recently developed algorithms aim to enhance the transmission of such information by, for instance, interlacing pulses for low-frequency channels and high-frequency channels or creating virtual electrodes by using multiple stimulation sites for each channel to represent intermediate frequencies. However, it is as yet unclear how much fine structure information is already provided by current algorithms and how much benefit can be gained from these new algorithms (Wilson & Dorman, 2008b).

Whatever future technological developments may bring, it is important to keep in mind that a CI does not restore normal hearing. Sound processing with a CI differs in several crucial ways from what the normal ear does, specifically in the spectral, temporal and amplitude information provided (Moore, 2003). Fortunately, speech recognition has been found to be fairly resistant to amplitude and temporal distortion in the speech signal (Dorman, Loizou, Spahr, & Maloff, 2002). However, a CI is also known to distort spectral information. This is due to a low pitch saturation limit, the limited number of effective channels and sometimes substantial shifts in frequency-to-place mapping in the cochlea (Moore, 2003). Unfortunately, spectral distortion has clear adverse effects on speech recognition (Dorman et al., 2002; Shannon, 2002; Xu & Pfingst, 2008). Furthermore, it should be noted that, although modern CIs allow fairly accurate speech recognition in quiet listening conditions, noisy environments or situations with multiple speakers talking at the same time continue to present a great challenge to users (e.g. Friesen, Shannon, Baskent, & Wang, 2001; Fu & Nogaki, 2005; Schafer & Thibodeau, 2006).

It is important to note that variation in CI technology impacts pediatric cochlear implantation practice and the study of spoken language outcomes. Depending on which manufacturer is preferred by CI centers, children will receive different CIs with different technical specifications and different implemented speech processing algorithms. However, all modern CIs have been shown to produce very good results in children and adults (Wilson & Dorman, 2008b). The role of these differences in explaining inter-individual variation in outcomes therefore appears to be limited (see e.g. Psarros et al., 2002; Skinner et al., 2002). Importantly, because all CI centers adopt the most recent techniques, it is difficult to compare children implanted today with children implanted five years ago - the so-called 'moving target' phenomenon (Geers, 2006).

1.2.2 CURRENT PRACTICE IN PEDIATRIC COCHLEAR IMPLANTATION

As a result of newborn hearing screening programs, the age of diagnosis in congenitally deaf children has decreased substantially in many countries (Gerrits, Brokx, & Rozier, 2005; Kennedy et al., 2006; Sarant, Holt, Dowell, Rickards, &
Reliable indices of auditory functioning in infants such as oto-acoustic emissions or auditory brainstem responses have made such a screening possible. Early diagnosis has paved the way for early intervention and support, including early cochlear implantation. Early diagnosis together with converging evidence for the positive effect of early implantation on spoken language development (see §1.4.1) has resulted in a sharp decrease in the age at which pre-lingually deaf children receive a CI. Implantation within the first year of life is becoming standard practice in many countries.

Cochlear implantation requires a multidisciplinary team to provide the most supportive environment for the child as well as for the parents and siblings (Archbold & O’Donoghue, 2009). If the hearing screening indicates severe to profound hearing loss, parents are referred to a CI centre where an audiological, medical, communication and psychological assessment is completed to determine whether a child is a potential candidate for a CI. In the meantime, acoustic hearing aids are fitted to determine the amount of benefit gained through means of acoustic amplification. The decision to proceed with implantation is shared between the parents and the CI team.

Currently, when the implant is surgically inserted, residual hearing is often lost during the operation. For profoundly deaf children with between 90 dB and 120 dB loss in the better ear, the benefit gained from the CI outweighs the loss of any residual hearing. For children with less severe hearing loss factors other than auditory thresholds, such as the amount of benefit obtained from acoustic amplification, may determine whether they receive a CI (Fitzpatrick et al., 2009). Another important issue to consider before implantation is whether the child suffers from additional disabilities, which concerns over 30% of the profoundly deaf children. Although the number of children with complex needs that are implanted has increased substantially in recent years, the presence of additional disabilities can strongly affect the outcomes and CI candidacy for these children has to be considered on a case-by-case basis (Edwards, 2007b).

The surgical procedure involved in cochlear implantation can safely be performed in young children, even within the first year of life (Eter & Balkany, 2009; Holt & Svirsky, 2008; Waltzman & Roland Jr., 2005). Approximately four weeks after surgical insertion of the internal component, the external component is fitted and the CI is activated. Following this first familiarization session, several more sessions will be necessary to program the individual electrodes. As the child

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3Cochlear implantation will only be considered when acoustic amplification provides insufficient benefit. In relation to this, it is important to note that in parallel with cochlear implant technology, digital hearing aid technology has also significantly advanced in the past decade (Edwards, 2007a). The rapid technological progress in both cochlear implant and digital hearing aid technology makes it difficult to establish strict criteria for CI candidacy, especially for children with hearing loss in the severe to profound range.
Chapter 1

gets used to the auditory input provided by the CI, re-programming is often required. The process of learning to interpret the signals from the CI takes time and stimulation by parents, teachers and speech therapists is important (Archbold & O'Donoghue, 2009).

Long-term electrical stimulation does not appear to have any negative consequences for the user (Waltzman, Cohen, Green, & Roland Jr., 2002a). However, Archbold and O'Donogue (2009) note that further operations during a child’s life-time will be likely, due to device failure or system upgrades, for instance to allow implementation of new processing strategies incompatible with the current internal component. Fortunately, re-implantation is usually accomplished without loss of functioning. The rates of pediatric revision surgery due to medical complications or device failure vary between 5% and 14% (Eter & Balkany, 2009).

Until a few years ago, implantation was performed only unilaterally and the non-implanted ear was sometimes fitted with an acoustic hearing aid to support some binaural hearing. However, bilateral implantation is rapidly becoming common practice, especially in children with no residual hearing. Reported benefits mainly concern localization and speech perception in noise, for which binaural hearing is important (Ching, Van Wanrooy, & Dillon, 2007; Firszt, Reeder, & Skinner, 2008; Johnston, Durieux-Smith, Angus, O'Connor, & Fitzpatrick, 2009; Schafer, Amlani, Seibold, & Shattuck, 2007). Much less is known about the benefits of pediatric bilateral implantation for more general spoken language outcomes (but see e.g. Nittrouer & Chapman, 2009; Scherf et al., 2009). Similar to the advantage for early implantation of the first CI, there appears to be an advantage for early implantation of the second CI, both in absolute terms and relative to the first implant (Gordon & Papsin, 2009; Papsin & Gordon, 2008).

For children with residual hearing in the non-implanted ear, it is as yet unknown under what circumstances a second CI will provide greater benefit than an acoustic hearing aid (Ching et al., 2007; Nittrouer & Chapman, 2009). The development of the so-called hybrid CIs might be helpful in this respect. These CIs combine acoustic amplification and electrical stimulation in a single device. They are not fully inserted in the cochlea and preserve residual hearing in the low-frequency range for acoustic amplification. As of yet, only limited data is available on the effectiveness of these CIs (but see e.g. Turner, Reiss, & Gantz, 2008). They might provide benefit to those with severe sensorineural hearing loss that have too much residual hearing in the low-frequency range to be a candidate for a traditional CI.

4 Budgetary constraints form one of the major obstacles for bilateral cochlear implantation; health agencies do not always reimburse the second CI (as is currently the case in the Netherlands, for instance). However, as more evidence concerning the benefits of bilateral implantation relative to unilateral implantation is gathered, more health agencies will probably decide to also reimburse the second implant as was the case in 2010 in Flanders.
Thus, children with a CI not only vary in the type of CI and implemented speech processing algorithm, but also age at diagnosis and implantation, the presence of additional disabilities and the use of hearing aids or bilateral CIs. The complex interplay of these and other variables contributes to the difficulty of predicting spoken language outcomes in children with a CI. Moreover, as already mentioned in §1.2.1, due to continuously changing technology and progressive lowering of age at implantation, children with a CI represent a “moving target” and research is therefore rapidly outdated. In the next two sections we will nevertheless try to evaluate the outcomes of pediatric cochlear implantation (§1.3) and discuss in detail two factors that have been found to affect these outcomes, namely age at implantation and communication modality (§1.4).

1.3 OUTCOMES OF PEDIATRIC COCHLEAR IMPLANTATION

1.3.1 EFFECTS ON SPOKEN LANGUAGE ABILITIES

Most studies on spoken language outcomes report scores on standardized clinical assessment measures of speech perception, speech production, and expressive and receptive vocabulary and language abilities (Schauwers et al., 2005). Studies have also elicited spontaneous speech samples to analyze speech production (e.g. Ertmer et al., 2002; Flipsen Jr. & Parker, 2008 for English; Schauwers, Gillis, Daemers, De Beukelaer, & Govaerts, 2004 for Dutch) and morphosyntactic abilities (e.g. Nicholas & Geers, 2007; Spencer, Tye-Murray, & Tomblin, 1998; Svirsky, Stallings, Lento, Ying, & Leonard, 2002 for English; Szagun, 2000 for German). Furthermore, a few studies have addressed narrative and conversational abilities in children with a CI (e.g. Crosson & Geers, 2001 for English; Ibertsson, Hansson, Maki-Torkko, Willstedt-Svensson, & Sahlen, 2009 for Swedish). Importantly, the rapid decrease in age at implantation has resulted in a need for reliable clinical assessment measures that are suitable for use with very young children (Eisenberg, Martinez, & Boothroyd, 2007; Mendel, 2008; Nikolopoulos, Archbold, & Gregory, 2005).

Studies on spoken language outcomes report considerable inter-individual variation, varying from performance at age-appropriate levels within a few years after implantation to short- and long-term delays (for reviews, see e.g. Belzner & Seal, 2009; Bond et al., 2009; Peterson et al., 2010; Schauwers et al., 2005; Thoutenhoofd et al., 2005). The percentages of children that perform age-appropriately vary substantially between studies and language domains. For instance, Schorr, Roth and Fox (2008) compared 39 American children with a CI between five and fourteen years of age with age-matched children with normal
hearing on a range of language measures. They found that, as a group, the children with a CI performed within one standard deviation or less of age-equivalent scores (i.e., age-appropriately) on tests of speech production, expressive and receptive vocabulary, morphology and syntax, and phonological processing, but not auditory memory and meta-semantics. However, on all tests except speech production, the group scored significantly lower than the children with normal hearing. On average, 85% obtained age-appropriate scores for speech production, 51% for receptive vocabulary, 66% for expressive vocabulary, 36% for morphology and syntax, 26% for phonological processing and 13% for meta-semantics.

Geers et al. (2009) reported scores for 153 American 6-year old children with a CI on expressive and receptive vocabulary and language abilities, as well as verbal intelligence. 50% of the children obtained age-appropriate scores for receptive vocabulary, 58% for expressive vocabulary, 47% for receptive language, 39% for expressive language and 46% for verbal intelligence. A longitudinal follow-up study of 85 children from the same sample showed that, from eight to nine years of age until 15-18 years of age, the percentage of children that obtained age-appropriate scores for verbal intelligence increased from 60% to 77% (Geers, Tobey, Moog, & Brenner, 2008).

Niparko et al. (2010) compared expressive and receptive language development in 188 American children with a CI to 97 age-matched children with normal hearing in 6-month intervals until three years post-implantation. Through the years all children with a CI showed greater improvement than expected from their pre-implantation scores, but did not reach age-appropriate levels even three years after implantation. Importantly, individual developmental trajectories differed widely among the children with a CI in comparison to the children with normal hearing.

A range of factors has been found to predict spoken language outcomes, including age at implantation, age at onset of hearing loss, pre-implant auditory thresholds, family support, communication modality, nonverbal IQ and implant characteristics (Geers et al., 2007). The most consistent predictors appear to be age at implantation, communication modality and pre-implant residual hearing (Peterson et al., 2010). That is, positive spoken language outcomes in children with a CI are associated with earlier implantation, oral communication approaches and lower pre-implantation auditory thresholds. Even when taking all these factors into account, however, typically only slightly more than 50% of the variation in outcomes is explained (e.g. Geers, 2002), precluding reliable predictions of outcomes (Peterson et al., 2010). In an attempt to help clinicians evaluate the spoken language abilities of children with a CI, Nicholas and Geers (2008) provide expected test scores according to age at implantation for two commonly administered formal language tests in English and a widely used parent-report instrument based on a relatively homogenous sample of 76 children with a CI. These scores can be used to compare against the performance of other children with a CI at 3.5 years of age (parent-
report) and 4.5 years of age (parent-report as well as formal language tests). Such comparisons can help in determining the child’s progress and in making informed educational decisions.

Reading outcomes in children with a CI have also been found to be extremely variable. Some studies report substantial delays for most children, whereas other studies report age-appropriate scores (Marschark, Rhoten, & Fabich, 2007). Unsurprisingly, the same demographic variables that affect spoken language outcomes have been found to affect reading outcomes (Connor & Zwolan, 2004). In addition, whether delays or age-appropriate scores are observed may be dependent on the skill measured (Vermeulen, Van Bon, Schreuder, Knoors, & Snik, 2007). Interestingly, Geers et al. (2008) report reading scores at eight to nine and 15-18 years of age. In this period, the percentage of children that obtained age-appropriate scores on a standardized test for reading recognition and comprehension actually decreased from 56% to 44%, whereas for verbal intelligence it increased from 60% to 77%, suggesting a discrepancy between long-term spoken language and reading outcomes.

It is important to note that standardized spoken language tests administered in clinical settings do not fully reflect communicative functioning (Lin et al., 2007; Lin et al., 2008). Furthermore, speech recognition with a CI might reach age-appropriate levels in quiet listening conditions, but be substantially poorer in the presence of noise (e.g. Schafer & Thibodeau, 2006). Unfortunately, many real-life situations are noisy, which present a particular challenge to hearing-impaired listeners (Shinn-Cunningham & Best, 2008).

In addition, a problem with most standardized spoken language tests is that they only provide the end result of a range of sensory, perceptual, cognitive and linguistic processes (Pisoni, 2000). This makes it difficult to interpret the results from such tests and determine the underlying cause of relatively good or poor performance. In an attempt to tackle this problem, several recent studies of spoken language outcomes in children with a CI have included more processing-oriented measures such as lexical access (e.g. Wass et al., 2008), non-word repetition (e.g. Burkholder-Juhasz, Levi, Dillon, & Pisoni, 2007), novel word learning (e.g. Houston, Carter, Pisoni, Kirk, & Ying, 2005) and verbal working memory (e.g. Burkholder & Pisoni, 2003; Pisoni & Cleary, 2003). Inter-individual variation in cognitive processes underlying spoken language acquisition and processing might help to explain variation in children’s outcomes (Pisoni, 2000; Pisoni et al., 2008). The present thesis contributes to this “processing approach” because it examines the use of acoustic cues in sound perception and the relation between sound perception and word learning in children with a CI, topics that have not yet received much attention.
1.3.2 Effects on Other Cognitive Abilities

Pisoni and colleagues, besides introducing the above-mentioned processing approach, have also drawn attention to the interaction between spoken language abilities and other cognitive abilities, such as sensorimotor and visuospatial abilities, working memory abilities and executive functioning (see Pisoni et al., 2008 for a review). For instance, Pisoni et al. (1999) reported strong correlations between verbal working memory abilities and scores on standardized tests of speech production and perception as well as language comprehension in English. Furthermore, more recent studies found, for instance, slower verbal working memory processing in children with a CI compared to children with normal hearing (Burkholder & Pisoni, 2006), delays in the development of sustained visual attention (Horn, Davis, Pisoni, & Miyamoto, 2005; but see also Tharpe, Ashmead, & Rothpletz, 2002) and delays in the development of visual-motor integration abilities (Horn, Fagan, Dillon, Pisoni, & Miyamoto, 2007a).

Fagan, Pisoni, Horn and Dillon (2007) related scores on subtests of sensorimotor and visuospatial processing from a developmental neuropsychological assessment battery to scores on receptive vocabulary, reading and digit span measures in children with a CI. They found that visuospatial, but not sensorimotor abilities correlated positively with these measures. More generally, delays in the development of sustained visual attention may have consequences for early social-cognitive and communicative development, for instance, in the development of joint visual attention, an important precursor of social learning (Corina & Singleton, 2009). In addition, children with a CI might be more prone to visual distraction in the classroom, which raises important concerns regarding the provision of optimal learning environments for these children (Dye, Hauser, & Bavelier, 2008).

The relationship between spoken language and other cognitive abilities in children with a CI is further exemplified by studies that report positive effects of cochlear implantation on, for instance, the development of visual attention abilities (Quittner et al., 2007), behavioral regulation (Edwards, Khan, Broxholme, & Langdon, 2006), social abilities (Bat-Chava, Martin, & Koseiw, 2005) and social well-being (Percy-Smith, Caye-Thomasen, Gudman, Hedegard-Jensen, & Thomsen, 2008; Schorr, Roth, & Fox, 2009).

Together, these findings underline that spoken language development is only one aspect of development affected by cochlear implantation. Many neural and cognitive

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5 For instance, subvocal rehearsal and serial scanning appear to operate more slowly in children with a CI, resulting in poorer performance on not only auditory, but also visual measures of verbal working memory (Burkholder & Pisoni, 2003; Cleary, Pisoni, & Geers, 2001; Dawson, Busby, McKay, & Clark, 2002). Children with a CI perform similarly to children with normal hearing, however, when the stimuli in the memory tasks are unlikely to be encoded verbally (Burkholder & Pisoni, 2006; Dawson et al., 2002).
changes as a consequence of auditory deprivation will already have taken place before implantation and reduced hearing abilities will continue to affect neural and cognitive development after implantation (Fagan & Pisoni, 2009). Deaf infants by necessity will be more focused on visual information in their surroundings (Fagan & Pisoni, 2009; Mitchell & Maslin, 2007), but the effects of such sensory processing biases on, for instance, the development of early speech perception abilities are not yet known (Gerrits, 2006; Houston, 2005). More generally, several decades of research have revealed both similarities and differences in cognitive processing between deaf and hearing children, adolescents and adults (Marschark & Hauser, 2008). It remains to be seen to what extent these findings also apply to children, adolescents and adults with a CI.

1.3.3 LONG-TERM OUTCOMES

Studies on the long-term outcomes of cochlear implantation have only recently become available (see e.g. Geers, Strube, Tobey, Pisoni, & Moog, 2011 and other papers in that issue). These studies suggest that spoken language abilities continue to improve well into adolescence (Geers et al., 2008) and that only a small percentage of those implanted in the countries studied no longer use their CI (Archbold, Nikolopoulos, & Lloyd-Richmond, 2009; Beadle et al., 2005; Spencer, Gantz, & Knutson, 2004; Waltzman et al., 2002a). Even less is known about the academic-occupational achievements of young adults who received a CI when they were young, but the few studies that have been published report good achievements (Beadle et al., 2005; Spencer et al., 2004). More long-term outcome studies in different countries are much needed, especially longitudinal ones with large representative samples that include outcomes for reading and academic achievements as well as for spoken language (Marschark et al., 2007).

1.4 EFFECTS OF AGE AT IMPLANTATION AND COMMUNICATION MODALITY ON SPOKEN LANGUAGE

1.4.1 AGE AT IMPLANTATION

Of all the factors that influence the outcomes of pediatric cochlear implantation, age at implantation seems to be one of the most robust factors, especially in relation to spoken language acquisition. The development of brain and behavior in humans and animals is characterized by multiple sensitive periods (Bischof, 2007; Knudsen,
2004; Thomas & Johnson, 2008), periods during which “having a certain kind of experience at one point in development has a profoundly different impact on future behavior than having that same experience at any other point in development” (Bruer, 2001, p.4). Language development has long been associated with one of these sensitive periods on the basis of different types of evidence: children reared in social isolation (Bortfeld & Whitehurst, 2001), deaf adults who had been raised and educated orally and systematically exposed to sign language only as adults (Mayberry & Lock, 2003), adult second language learning (Birdsong, 2006; Hakuta, 2001), and infant native language attunement (Kuhl, Conboy, Padden, Nelson, & Pruitt, 2005; Werker & Tees, 2005).

Studies examining the effects of early implantation in congenitally deaf children are a recent contribution to the literature on sensitive periods in language development (Tomblin, Barker, & Hubbs, 2007). Sensory deprivation, such as hearing loss or sight loss, during sensitive periods in development can have profound and permanent effects on the development of cortical connections within the brain (Bavelier & Neville, 2002; Pascual-Leone, Amedi, Fregni, & Merabet, 2005). One such effect is cross-modal reorganization, when cortical areas that usually respond to input from the deprived sensory organ (e.g., the ear) start responding to input from another sensory organ (e.g., the eye). Several studies have provided evidence for cross-modal cortical reorganization in children and adults with CIs (for reviews, see Peterson et al., 2010; Sharma, Nash, & Dorman, 2009). If a CI is inserted and activated after cortical reorganization has taken place, benefit of the implant is limited because necessary neural connections within the auditory cortex can no longer be established (Kral & Eggermont, 2007). Age cut-offs reported in neurophysiological studies suggest a sensitive period for the development of normal (sub)cortical auditory processing that lasts until three to four years of age (e.g. Sharma, Gilley, Dorman, & Baldwin, 2007).

In support of these neurophysiological studies, behavioral studies have also found strong effects of early versus late implantation on spoken language outcomes. In fact, many behavioral studies report advantages for early implantation in children younger than three years (Marco-Algarra et al., 2009). That is, implantation before two years of age seems to lead to significantly better spoken language outcomes than at a later age (e.g. Anderson et al., 2004; Artieres, Vieu, Mondain, Uziel, & Venail, 2009; Chin, Svirsky, & Jester, 2007; Geers, Nicholas, & Sedey, 2003b; Kirk, Miyamoto, Ying, Perdew, & Zuganelis, 2003; Svirsky, Teoh, & Neuburger, 2004; Zwolan et al., 2004). A few recent studies even suggest that implantation before 12 months of age is advantageous (e.g. Coene, Schauwers, Gillis, Rooryck, & Govaerts, in press; Colletti, 2009; Dettman, Pinder, Briggs, Dowell, & Leigh, 2007; Holt & Svirsky, 2008; Waltzman & Roland Jr., 2005).

However, not all studies report advantages for early implantation (e.g. Geers, 2004). It has been suggested that age at implantation might differentially affect
language domains (Geers et al., 2009; Holt & Svirsky, 2008) as well as abilities within one language domain (Harrison, Gordon, & Mount, 2005). Additionally, other variables such as length of CI use, age at diagnosis, pre-implant hearing thresholds or demographic variables such as ethnicity and social-economic status can confound with age at implantation. The effect, or lack thereof, of age at implantation may thus be indirect the result of uncontrolled variables. Early implantation does not guarantee successful spoken language outcomes, but it makes them likely.

1.4.2 COMMUNICATION MODALITY

The deaf community was long opposed to pediatric cochlear implantation, but this opposition has slowly waned (Christiansen & Leigh, 2004). It has now become a standard procedure in many countries, with the majority of congenitally deaf children receiving a CI. Around 90% of the congenitally deaf children have two hearing parents, which in part explains why so many deaf children receive a CI.

Ideally, after the diagnosis of hearing loss in their child, parents receive objective, balanced information on all available options such as acoustic hearing aids, CIs, sign language and the deaf community, so that they can make a well-founded decision (Archbold, Sach, O'Neill, Lutman, & Gregory, 2006). Unfortunately, this is not always the case. Parents in different countries often report that information about educational and communicative options was conflicting and biased towards or against cochlear implantation (Berg, Ip, Hurst, & Herb, 2007; Christiansen & Leigh, 2004; Sach & Whynes, 2005; Sorkin & Zwolan, 2008; Wever, 2002). Nevertheless, few parents regret having taken the decision to have their child implanted (Sach & Whynes, 2005) and most children, when they are older, appreciate the decision their parents took on their behalf (Wheeler, Archbold, Gregory, & Skipp, 2007). This is supported by studies that have examined their quality of life through parent report (e.g. Archbold, Sach, O'Neill, Lutman, & Gregory, 2008; Sach & Whynes, 2005; Stacey, Fortnum, Barton, & Summerfield, 2006) and through the children themselves (e.g. Leigh, Maxwell-McCaw, Bat-Chava, & Christiansen, 2008; Preisler, Tvingstedt, & Ahlstrom, 2005; Schorr et al., 2009).

Notwithstanding the wide-spread acceptance of cochlear implantation, a much discussed topic remains the role of signed input in the raising and education of children with a CI (e.g. Delore et al., 1999; Leigh, 2008; Marschark, 2007; Papsin & Gordon, 2007). Parents may decide to include signs in the child’s language input at home, often after taking advice from pediatricians, educational psychologists and other parents (e.g. Christiansen & Leigh, 2004; Watson, Hardie, Archbold, & Wheeler, 2008; Wever, 2002). The type of language input at school is a more
complex question, however (e.g. Knoors, 2007; Leigh, 2008; Marschark, 2007). Countries can differ in the extent to which they stimulate mainstreaming of children with special needs. Furthermore, the role of a sign language in the educational setting differs between countries and within countries between states, provinces and districts. Schools that embrace an auditory-verbal approach do not include any sign language. Other schools might adopt a simultaneous communication approach, with sign-supported speech as the main mode of communication, or a bilingual-bicultural approach, with instruction in a spoken language as well as a sign language. Depending on national, state and local laws, parents might not be entirely free in the decision they make regarding the educational placement of their child. Adding to this complexity, children with a CI often make a transition from one educational setting to another, for instance from special education to mainstream or from a more bilingually oriented program to a more sign-supported oriented program (e.g. Watson, Archbold, & Nikolopoulos, 2006).

When they are older, children with a CI will mainly decide themselves whether they will speak or sign, possibly even depending on the communicative situation and interlocutor, and whether they switch off the implant at time or even stop using it (Wheeler et al., 2007). Parents report that changes in how they communicate with their children at home are most often driven by the preferences of the children themselves (Preisler, Tvingstedt, & Ahlstrom, 2001; Watson et al., 2008). That is, cochlear implantation entails a communication journey for both the parents and the children (Wever, 2002; Wheeler, Archbold, Hardie, & Watson, 2009). Unfortunately, it is not without obstacles due to opposition from professionals or restrictions imposed by local or national educational policies (Sach & Whynes, 2005; Sorkin & Zwolan, 2008; Wever, 2002; Wheeler et al., 2009).

Fueling the discussion on the role of signed input for children with a CI are reports of its negative effects on spoken language (Geers, 2006). Available studies mostly compared “Oral Communication” (OC) settings, where only spoken language is used, to “Total Communication” (TC) settings, where both spoken language and some form of signed communication are used, mainly in the United States or the United Kingdom. The majority of these studies report an OC advantage (Archbold et al., 2000; Geers et al., 2000; Kirk et al., 2003; Svirsky et al., 2000). However, other studies found no effect of educational setting (e.g. McConkey Robbins, Svirsky, & Kirk, 1997; Svirsky et al., 2004) or even a TC advantage (e.g. Connor et al., 2000; McConkey Robbins, Bollard, & Green, 1999). One explanation for these variable results may lie in the nature of Total Communication, which is an educational philosophy that incorporates a variety of practices that are often not described in research studies (Spencer & Tomblin, 2006). It may also be that children who show less than expected progress are the ones who start in a TC setting, remain in this setting for a longer time or transition from an OC setting to a TC setting. Another explanation may come from the fact that some studies only used
the spoken modality to administer tests, whereas others used the modality preferred by the child (for discussion, see Geers, 2006).

Regarding the effects of signed input, positive results have been explained by the suggestion that signed vocabulary acquired pre-implantation might bootstrap spoken vocabulary development (Yoshinaga-Itano, 2006). Additionally, it might provide important early language stimulation (Connor et al., 2000). In contrast, Pisoni et al. (1999) suggested that the efficiency of auditory short-term memory processes such as encoding and rehearsal might benefit from increased exposure to speech and therefore signed input may have negative effects. Moreover, simultaneously attending to two visual sources of information (i.e., manual-visual and audiovisual) might create competition for limited processing resources (Bergeson, Pisoni, & Davis, 2005). Finally, using sign language before implantation might stimulate cross-modal reorganization of the auditory cortex, which may negatively impact speech processing (Giraud & Lee, 2007).

Unfortunately, only a few studies have compared spoken and signed language abilities in the same sample of children with a CI, and most of these are case studies (but see De Raeve et al., 2009). A within-subject approach allows a more direct examination of the effects of signed input on spoken language development and controls for most confounds that may affect studies comparing children in OC and TC settings. In addition, the question of whether children with a CI benefit from seeing signs at the same time when they lip-read spoken words or whether the two sources of visual information compete has yet to be empirically tested. This thesis contributes to filling these two gaps in the literature by examining the relationship between speech and sign perception in children with a CI who varied in the amount of signed input they received at home and school.

1.5 THE ORGANIZATION OF THIS THESIS

This introductory chapter presented the goals of this thesis, namely to provide insight into the underlying processes in the perception of speech sounds and words by children with a CI, as well as the relationship between sign and speech perception abilities. In addition, it provided an introduction to pediatric cochlear implantation that will help in understanding and evaluating the research presented in this thesis. The remainder of this thesis is organized as follows.

Chapter 2 discusses the research questions of this thesis in more detail. Chapter 3 introduces the tasks that were designed to answer the research questions, as well as the different groups of participants that provided the answers. In Chapters 4 to 7, the results of the different experiments are presented. More specifically, Chapter 4 focuses on the use of acoustic cues in consonant and vowel perception. Chapter 5
Chapter 1 examines the relation between perceiving speech sounds and learning novel words. Chapter 6 presents data on pre-lexical and lexical perception in the signed modality and addresses the relationship between perception in the signed and the spoken modality. Chapter 7 discusses the interaction between both language modalities during language perception. Finally, Chapter 8 summarizes the findings and presents the overall conclusions.