UNIVERSITEIT GENT

Faculty of Medicine and Health Sciences

Speech perception outcomes

in cochlear implantees

Birgit Philips

Promotor Prof. dr. Ingeborg Dhooge

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Thesis submitted to fulfill the requirements for the degree of

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Table of contents

List of publications

This dissertation is based on the following articles accepted in or submitted to international peer-reviewed journals:

1. Philips B, Corthals P, De Raeve L, D'Haenens W, Maes L, Bockstael A, Keppler H, Swinnen F, De Vel E, Vinck BM, Dhooge I. Impact of newborn hearing screening: comparing outcomes in pediatric cochlear implant users. *The Laryngoscope*, 2009; 119(5): 974-979.

2. Philips B, Vinck B, De Vel E, Maes L, D'haenens W, Keppler H, Dhooge I. Characteristics and determinants of music appreciation in adult CI users. *European Archives of Otolaryngol*ogy, 2012, 269: 813-821.

3. Philips B, Maes L, Keppler H, Dhooge I. Cochlear implantation in children deafened by congenital cytomegalovirus and matched Connexin 26 peers. *Accepted for publication in International Journal of Pediatric Otolaryngology,* 2013.

4. Philips B, Baudonck N, Maes L, Keppler H, Dhooge I. Assessing and predicting speech perception outcome in early-deafened, late-implanted adults. *Submitted to Audiology and Neuro-otology*, 2013.

5. Philips B, Vanpoucke F, Maes L, Keppler H, Dhooge I. Spread of excitation measures in adult cochlear implant users: feasibility, long-term stability and correlation with speech performance. *Under review for Clinical Neurophysiology*, 2013.

List of abbreviations

A§E®	auditory speech sound evaluation
AABR	automated auditory brainstem response
AB	Advanced Bionics®
ADHD	attention deficit hyperactive disorder
ANOVA	one-way analysis of variance
ANSD	auditory neuropathy spectrum disorder
BICI	bilateral cochlear implants
BTE	behind-the-ear
C	consonant
CAP	categories of auditory performance
cCMV	congenital cytomegalovirus
CI	cochlear implant
CIS	continuous interleaved sampling
C-level	comfort-level
CNC	consonant-nucleus-consonant
CO	Cochlear®
CT	computertomography
CVC	consonant-vowel-consonant
cVEMP	colic vestibular evoked myogenic potentials
Cx26	connexin 26
dB HL	decibel hearing level
dB SPL	decibel sound pressure level
DR	dynamic range

e.g.	exempli gratia, for example
EABR	electrically evoked auditory brainstem responses
EAS	electroacoustical stimulation
ECAP FM	physiological forward masking
ECAP	electrically evoked compound action potentials
EFI	electrical field imaging
el	electrode
ErrSP	errors in speech production
ES	early screened
et al	et alia, and others
exp	experiment
F	female
FDA	federal drug administration
FDM	Freedom™
FM	frequency modulated
fMRI	functional magnetic resonance imaging
HA	hearing aid
HI	hearing impaired
HINT	hearing-in-noise-test
HL	hearing loss
HRQoL	health related quality of life
Hz	hertz
i.e.	id est
IT-MAIS	infant-toddler meaningful auditory integration scale
ITr	relative information transmitted
L	left
LINT	Leuven intelligibility number test
LiP	listening progress profile

LIST	Leuven intelligibility speech test
LS	late screened
M	male
m	months
MCL	most comfortable level
mm	millimeters
MMN	mismatch negativity
mo	month
MR	mental retardation
MRI	magnetic resonance imaging
ms	milliseconds
MTP	music training program
N	number
NCIQ	Nijmegen cochlear implant questionnaire
NH	normal hearing
NRT	neural response telemetry
NVA	'*Nederlandse vereniging voor audiologie'*
OAE	oto acoustic emissions
OTOF	otoferlin
PAP-V	pediatric auditory performance test for Flanders
PCA	principal component analysis
PDD	pervasive developmental disorder
PET	position emission tomography
PFM	psychophysical forward masking
progr	progressive
PTA	pure tone average
QoL	quality of life
R	right

rec	recording electrode
SAP	*'spraakaudiometrie met plaatjes'*
SAS	simultaneous analog stimulation
SD	standard deviation
SES	socioeconomic status
SIR	speech intelligibility rating
SNHL	sensorineural hearing loss
SNR	signal-to-noise ratio
SOE	spread of excitation
SOE-VM	spread of excitation, variable masker
SOE-VR	spread of excitation, variable recording
SPEAK	spectral peak
SPSS	statistical package for the social sciences
SRT	Speech Reception Threshold
SRT_n	speech reception threshold in noise
SRT_q	speech reception threshold in quiet
un	unknown
UNHS	universal newborn hearing screening
UNHSP	universal newborn hearing screening program
uni	unilateral
UZGent	'u*niversitair ziekenhuis Gent'*
WHO	World Health Organization
yr	year
µV	microvolt

Summary

Cochlear implants (CIs) provide bilaterally severely hearing impaired (HI) children and adults with important auditory cues resulting in equal or even better speech perception results compared to bilaterally severely HI hearing aid (HA) listeners. However, a large number of clinical studies showed that the outcome among implant recipients is highly variable. By understanding this variability, predictive accuracy on outcome after implantation will be improved and moreover, better counseling tools for future CI recipients will be available. Hence, clinicians, manufacturers, and researchers can create the best possible circumstances to optimize outcome after implantation.

The objective of this thesis was to assess the speech perception outcomes of a contemporary group of pediatric and adult CI recipients, and to investigate possible predictive variables.

Regarding the **pediatric** population, our study confirmed that early detection of hearing impairment due to the implementation of Universal Newborn Hearing Screening Program in Flanders results in early intervention and implantation of bilaterally severely HI children, and has a major positive impact on both auditory receptive skills and speech intelligibility.

As congenital cytomegalovirus (cCMV) and Connexin-26 (Cx26) are, respectively, the leading cause of non-genetic and genetic hearing impairment, these children represent a considerable part in the pediatric CI population. This study showed that on average, cCMV-CI children progress equally over a 5-yr period after CI compared to matched Cx26-CI children on speech perception and speech production outcomes. Former studies comparing implanted cCMV and non-cCMV children reported mixed results, possibly because these studies hardly controlled for confounding variables such as abnormal MRI results. Our study tentatively suggests that over a 5-yr follow-up period results of cCMV-CI children with abnormal MRI findings and their matched Cx26-CI counterparts show that these cCMV-CI children can catch up for speech perception, but lag behind for speech production, whereas cCMV-CI children with normal MRI results achieve comparable, or even slightly better,

speech perception and production results as their Cx26-CI peers. Although further research is needed, the inclusion of MRI results may assist in improved counseling of parents with cCMV deafened children seeking CI.

In an attempt to unravel the influence of channel interaction, we conducted a study to examine Electrically evoked Compound Action Potentials (ECAP) spatial patterns within **adult** CI users. Despite the use of a thorough speech perception test battery, neither statistically significant correlation was found between spatial ECAP patterns, nor the presence of peak displacements, and speech perception performance after implantation. These results suggest that ECAP derived spatial profiles with respect to width measurements and peak displacements should not be used to predict speech perception measurements within clinical settings.

Counseling early-deafened, late-implanted adults can be improved by implementing their speech production data. Our study showed that by means of a single factor, Errors in Speech Production, 78% of the variance of postimplantation speech perception (phonemescore at 70dBSPL, 12 m postop) can be explained, with participants having fewer errors in speech production achieving better scores in speech perception.

Adult CI recipients with residual hearing in their non-implanted ear, should be encouraged to wear a HA, because our study revealed that bimodal listeners have improved music appreciation. In addition, the implementation of a music training program in rehabilitation for all adult CI users, must be considered.

Samenvatting

Cochleaire implantaten (CIs) voorzien bilateraal ernstig gehoorgestoorde kinderen en volwassenen van belangrijke auditieve cues, die hen in staat stellen evenwaardige of zelfs betere spraakperceptiescores te behalen in vergelijking met bilateraal ernstig gehoorgestoorde hoortoestelgebruikers. Klinische studies tonen echter een aanzienlijke inter-subject variabiliteit in de bekomen resultaten na implantatie. Inzicht in de factoren die de resultaten na implantatie beïnvloeden, verhoogt de voorspelbaarheid van de postoperatieve resultaten en optimaliseert het counselen van toekomstige CI patiënten. Aldus kunnen therapeuten, fabrikanten en onderzoekers de best mogelijke omstandigheden creëren om de resultaten na implantatie te optimaliseren.

Het hoofddoel van dit doctoraat is het in kaart brengen van de spraakperceptieresultaten van een hedendaagse groep van pediatrische en volwassen CI-gebruikers en het onderzoeken van mogelijke voorspellende variabelen.

Onze studie omtrent de **pediatrische** CI populatie bevestigt dat vroegtijdige detectie van gehoorverlies na de invoering van de universele gehoorscreening in Vlaanderen, resulteert in vroegtijdige interventie en implantatie in bilateraal ernstig gehoorgestoorde kinderen. Bovendien heeft vroegtijdige detectie een aanzienlijke positieve impact op hun spraakperceptie en spraakproductie.

Congenitaal cytomegalovirus (cCMV) en Connexine 26 (Cx26) zijn, respectievelijk, de meest voorkomende niet-genetische en genetische oorzaak van gehoorverlies en zijn dus uitvoerig vertegenwoordigd bij de etiologische diagnostiek van de pediatrische CI-populatie. Onze studie toonde aan dat gemiddeld genomen, cCMV-CI kinderen eenzelfde vooruitgang boeken op spraakperceptie en spraakproductie als hun gematchte Cx26 leeftijdgenoten. Echter, cCMV-CI kinderen met afwijkende MRI resultaten lijken een vertragende ontwikkeling in hun spraakproductie te vertonen in vergelijking met hun Cx26-CI leeftijdsgenootjes. cCMV-CI kinderen met normale MRI resultaten bekomen vergelijkbare, of zelfs beperkt betere spraakperceptie en –productieresultaten in vergelijking met hun Cx26-

leeftijgsgenootjes. Hoewel verder onderzoek noodzakelijk is, kan het includeren van MRI gegevens overwogen worden in het counselinggesprek met ouders van toekomstige CI-gebruikers.

Om de voorspellende variabele kanaalinteractie beter in kaart te brengen, werd een studie uitgevoerd gebruik makend van spatiale patronen gebaseerd op elektrisch geëvokeerde actiepotentialen (ECAP – '*Electrially Evoked Action Potentials*') bij **volwassen** CI gebruikers. Ondanks het gebruik van een ruime spraakperceptie testbatterij, werd geen statistische significante correlatie gevonden tussen spatiale ECAP patronen, noch de aanwezigheid van piekverschuivingen en spraakperceptie na implantatie. Dit resultaat suggereert dat spatiale ECAP patronen die zich baseren op breedtemetingen en piekverschuivingen, in de klinische praktijk beter niet worden aangewend om spraakperceptie te voorspellen.

Het counselen van vroegtijdig-dove, laattijdig-geïmplanteerde volwassenen kan geoptimaliseerd worden door de implementatie van hun spraakproductiegegevens. Onze studie toonde aan dat gebruik makend van één enkele factor, nl. fouten in spraakproductie, 78% van de spraakperceptiescore post-implantatie (foneemscore op 70dB SPL, 12 m na implantatie) verklaard kan worden in die zin dat deelnemers met minder fouten in spraakproductie een beter spraakperceptiescore behalen.

Volwassen CI-gebruikers met restgehoor in hun niet-geïmplanteerde oor, dienen aangemoedigd te worden om een contralateraal hoortoestel te dragen, gezien onze studie aantoonde dat bimodale luisteraars een verbeterde muziekappreciatie kennen. Daarnaast dient de invoering van een muziektrainingsprogramma voor alle volwassenen CI-gebruikers binnen de revalidatie, overwogen te worden.

General introduction

Chapter 1

Hearing and deafness

1.1 The auditory system

Auditory processing begins when an acoustic stimulus arrives at the external ear and is conducted by structures in the middle ear to the sensory organ in the inner ear, or cochlea (**Figure 1.1**). Within the cochlea, acoustic stimuli are transformed into a train of impulses in the auditory nerve, which carries a neural representation of acoustic events to the brain (May, 2000).

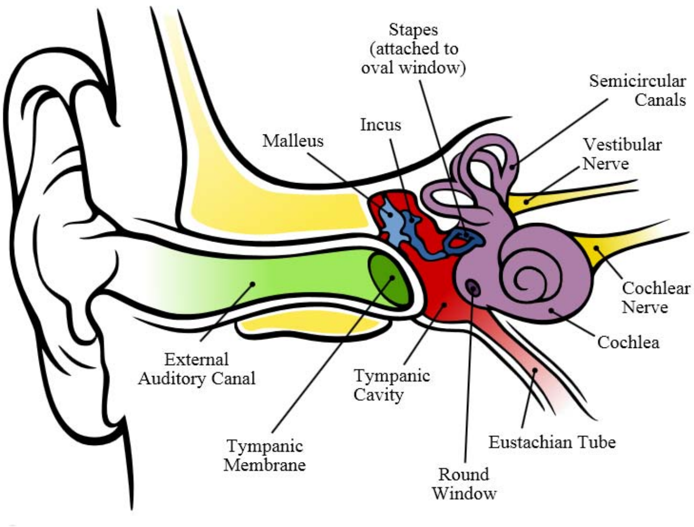


Figure 1.1 Outer, middle, and inner ear, Wikipedia.

The outer ear and its inherent resonances enhances auditory sensitivity by collecting and directing sound energy toward the relatively small surface of the tympanic membrane. Sound waves cause this membrane to oscillate, which sets up vibration in the three ossicles of the middle ear. Movement of the stapes footplate in the oval window transmits the sound vibrations from the middle ear to the inner ear. The middle ear functions as an impedance-matching device between air (outer ear) and fluid (inner ear). The inner ear, or cochlea, is a fluid filled snail-like structure with a total length of approximately 35 mm that houses the sensory organ (organ of Corti). The organ of Corti contains the inner and outer hair cells. Protruding from the hair cells are stereocilia. The motion of the basilar membrane causes the stereocilia of the inner hair cells to move back and forth starting the release of neurotransmitter towards the auditory nerve. If this release induces a sufficient depolarization of the nerve, action potentials are initiated and propagate towards the brain. The outer hair cells perform a mechanical amplification of the basilar membrane vibrations, sharpen up frequency tuning and provide dynamic range compression.

The patterns of excitation passed along the auditory pathways encode information about the spectrum, amplitude and frequency of the incoming sound. This representation is facilitated by the tonotopic organization of the cochlea and refers to the fact that sounds with different frequencies stimulate the organ of Corti in different areas along the basilar membrane. High frequency sounds stimulate the organ of Corti at the basal turn, whereas low frequency sounds cause maximum excitation after travelling some distance through the cochlea in the direction of the apex. This spectral analysis is preserved in the transmission of action potentials, because the different nerve fibres that constitute the cochlear nerve each correspond with a specific location in the cochlea (May, 2000, Cummings, 2004).

1.2 Hearing loss

Estimates by the World Health Organization (WHO, 2013) indicate that over 5% of the world's population - 360 million people - suffer from disabling hearing loss (HL). Disabling HL refers to HL greater than 40 dB HL in the better hearing ear in adults and a HL greater than 30 dB HL in the better hearing ear in children.

There are different **degrees** of HL. Based on the pure tone average (PTA) - the average pure tone threshold at 500, 1000 and 2000 Hz - the following categories are distinguished: mild (PTA 25-40 dB HL), moderate (40-55 dB HL), moderate-severe (55-70 dB HL), severe (70-90 dB HL) and profound (> 90 dB HL).

Besides degrees of HL, also the **type** of HL is taken into account. There are three basic types of HL: conductive HL, sensorineural hearing loss (SNHL) and mixed HL. Conductive HL occurs when sound is not conducted efficiently through the outer ear and/or middle ear. Pathologies such as otitis media or perforated eardrums can lead to conductive HL. The source of SNHL lies within the inner ear or cochlea. If a conductive HL occurs in combination with a SNHL, this HL is referred to as a mixed HL.

HL can be divided into congenital and acquired HL. Long-standing data on profound HL in infancy indicate that 50% of HL is thought to be due to **environmental** factors (e.g. congenital cytomegalovirus (cCMV) infection) and 50% is **hereditary** (e.g. GJB2 mutation) (**Figure 1.2**) (Paparella and Schachern, 1991).

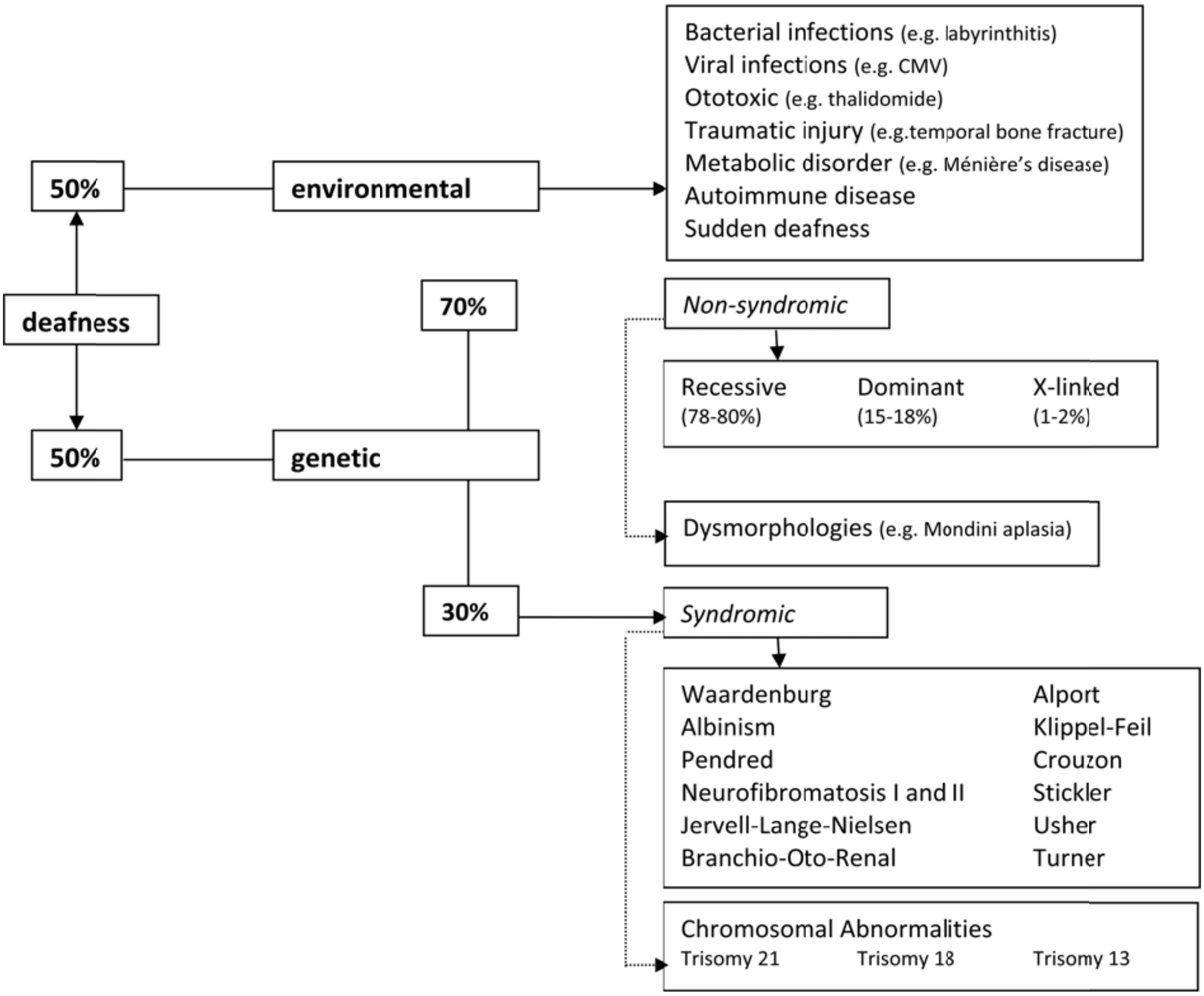


Figure 1.2 The distribution of HL etiology, adapted from Diefendorf, 2009

Furthermore, the **onset** of bilateral severe to profound SNHL is taken into account. Individuals acquiring bilateral severe to profound SNHL before the acquisition of oral language are prelingually deaf, whereas postlingually deafened subjects obtained their HL after developing oral language.

1.3 The cochlear implant

If a patient suffers from a severe to profound bilateral SNHL and amplification by means of a classical hearing aid (HA) does not suffice, a cochlear implant (CI) should be considered. The CI is a sensory neurostimulating device that bypasses the malfunctioning peripheral ear and electronically stimulates the auditory nerve. All modern CIs consist of an internal and an external component and share a set of common functional components (**Figure 1.3**).

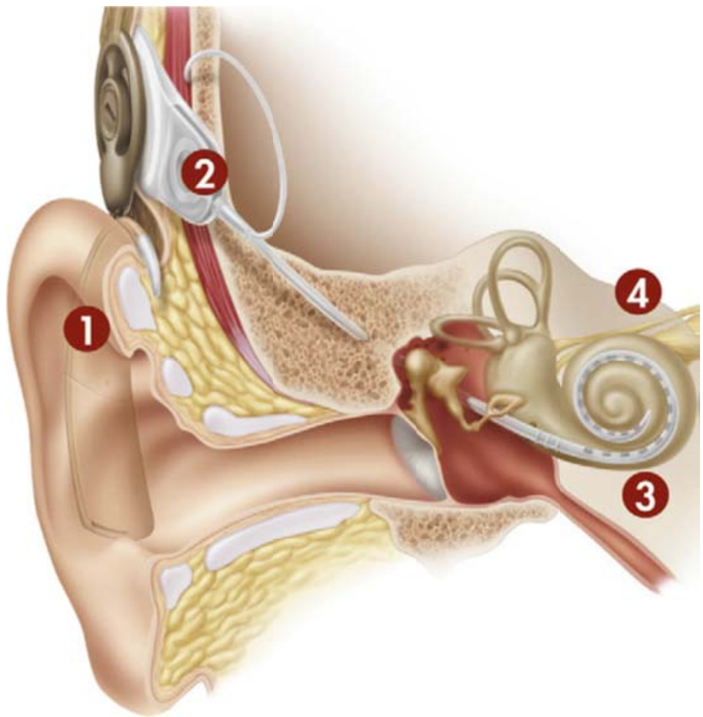


Figure 1.3 The Cochlear Implant (CI) (Printed with permission of Cochlear Americas, Ltd.)

The external part comprises three separate components: the microphone, the sound processor, and the transmitter. The microphone picks up the natural acoustic stimulus and converts the pressure variations from the sound stream into analog electrical variations that are subsequently sent to the behind-the-ear (BTE) sound processor (1). The sound processor in turn alters the signal to a format that is meaningful to the receiving spiral ganglion cells and central nervous system through the processes of amplification, compression, and filtering. This processed signal is encoded and transmitted by Frequency Modulated (FM)-waves through a transcutaneous link to the internal package (2). The internal part is surgically implanted and contains a receiver-stimulator and a magnet, placed behind the ear under the skin and an electrode array that is placed in the cochlea (usually the scala tympani). The receiver-stimulator decodes the signal and produces a pattern of electrical stimulus currents in the intra-cochlear electrodes (3), depending on the signal processing strategy used. The electrode array comprises several contacts (generally between 12 and 22 contacts) which are placed along the cochlea. Hence, different subpopulations of neurons depending on the frequency components of the incoming sound, are stimulated. Electrodes near the base of the cochlea are stimulated with high-frequency signals, while electrodes near the apex are stimulated with low-frequency signals. The CI is based on the idea that there are enough auditory nerve fibers left for stimulation in the vicinity of the electrodes (4). Once the nerve fibers are stimulated by these electrodes, they fire and propagate neural impulses to the brain (Loizou, 1999). In addition, the auditory brain interprets these impulses as sound (Wilson, 2000, Clark, 2006, Eisenberg, 2009).

1.4 Representing speech information with a cochlear implant

The purpose of the speech processor is to transform microphone input into patterns of electrical stimulation that convey the information content of speech and others sounds. The amount of information that can be presented and perceived with a CI is much less than that for someone with normal hearing listening to an unprocessed acoustic signal. For example, the number of electrodes that can be included in the scala tympani is far less than the number of ganglion cells and neurons in normal cochleas. Current implant systems have no more than 22 intracochlear electrodes, which for such closely spaced electrodes address highly overlapping populations of neurons because of the spread of the electric fields across electrode positions.

The design of implant systems can be viewed as a problem of squeezing speech through a narrow bottleneck, imposed by the lack of spatial specificity in stimulation and by limitations in perception. Early designs attempted to represent only the most important aspects of speech at the electrodes. Throughout the years, different speech coding strategies have been developed. Currently, the most widely used speech processing strategies are ACE (Advanced Combination Encoder), CIS (Continuous Interleaved Sampling), and HiRes (High Resolution). The general processing scheme is similar for these strategies. The incoming sound is passed through a pre-processing block, consisting of an optimal automatic gain control (AGC) and a pre-emphasis filter. The pre-emphasis filter attenuates frequency components below 1.2 kHz at 6 dB/octave in order to emphasize the low-energy high-frequent information that is important for speech perception, relative to the high-energy low-frequency portion of speech. The output of the pre-emphasis filter is directed to a bank of band-pass filters. Each filter corresponds to a stimulation channel or a pair of electrodes between which electrical currents can flow. The return electrode is either one or both extra-cochlear electrodes (monopolar stimulation) or one or more intra-cochlear electrodes (bipolar stimulation). In each channel the envelope of the signal is extracted through low-pass filtering and half-wave rectification. Next, compression is applied to map the acoustic signal into the smaller electrical dynamic range of the patient. Finally, the compressed envelope signals are used to modulate a train of biphasic electrical pulses. The output of each filter is mapped onto each of 12 to 22 stimulation channels, to mimic the tonotopic organization of the basilar membrane.

1.5 Amplitude, temporal, and spectral cues in cochlear implant speech processing

1.5.1 Amplitude cues

NH subjects have on average a 120 dB dynamic range and as many as 200 discriminable steps within this range. In contrast, a typical CI user has a 10 to 20 dB dynamic range and 20 discriminable steps (Nelson et al, 1996, Zeng et al, 1998, Zeng and Shannon, 1999). In acoustic hearing, loudness grows as a power function of intensity, whereas loudness tends to grow as an exponential function of electric currents in electric hearing (Zeng and Shannon, 1992). These differences are likely due to the loss of cochlear processing and have to be accounted for in both the design and fitting of the speech processor (Zeng, 2004).

1.5.2 Temporal cues

Temporal cues in speech can transmit information on envelope (< 50 Hz), periodicity (50-500 Hz), and fine structure (> 500 Hz) (Plomp, 1983, Rosen, 1992).

Shannon (1992) showed that CI users are able to follow temporal fluctuations in the 1-50 Hz range and concluded that their perception of envelope fluctuations is relatively normal as long as the loudness is mapped properly from acoustic to electrical domains; center or peak clipping would reduce the degree of amplitude fluctuations.

Temporal fluctuations found between 50 and 500 Hz provide periodicity information. NH listeners are able to perceive temporal fluctuations in this range purely in the temporal domain, without spectral analysis (e.g. Viemeister, 1979). CI recipients are also able to perceive and discriminate temporal information in this range (e.g. Shannon, 1983). Periodicty cues up to 300 Hz might provide information about voice pitch and intonation contours.

Generally, CI recipients are unable to detect temporal fluctuations faster than 300 to 500 Hz. However, some can discriminate these fluctuations up to 900 Hz (Kong, 2009). The ability to perceive temporal fine structure (> 500 Hz) may be a key difference in performance between good and poor users, because the frequency region between 500 and 1500 Hz is critical for speech perception, and CI users may not perform well if they are unable to discriminate information in this frequency range, either temporally or spectrally. CI users who can not access temporal information at frequencies higher than 500 Hz can only access information in this frequency range from the tonotopic place of the electrodes (spectral information).

1.5.3 Spectral cues

Frequency is likely to be encoded by both time and place mechanisms in NH listeners. The time code reflects the auditory nerve’s ability to phase lock to an acoustic stimulus for frequencies up to 5000 Hz, whereas the place code reflects the cochlear filter’s property to divide an acoustic stimulus into separate bands. The center frequency of these bands corresponds to different places along the cochlea and is preserved along the entire auditory pathway. In NH subjects, the number of largely independent filters is about 28 for the range of frequencies covered by speech sounds (Glasberg and Moore, 1990, Moore, 2003).

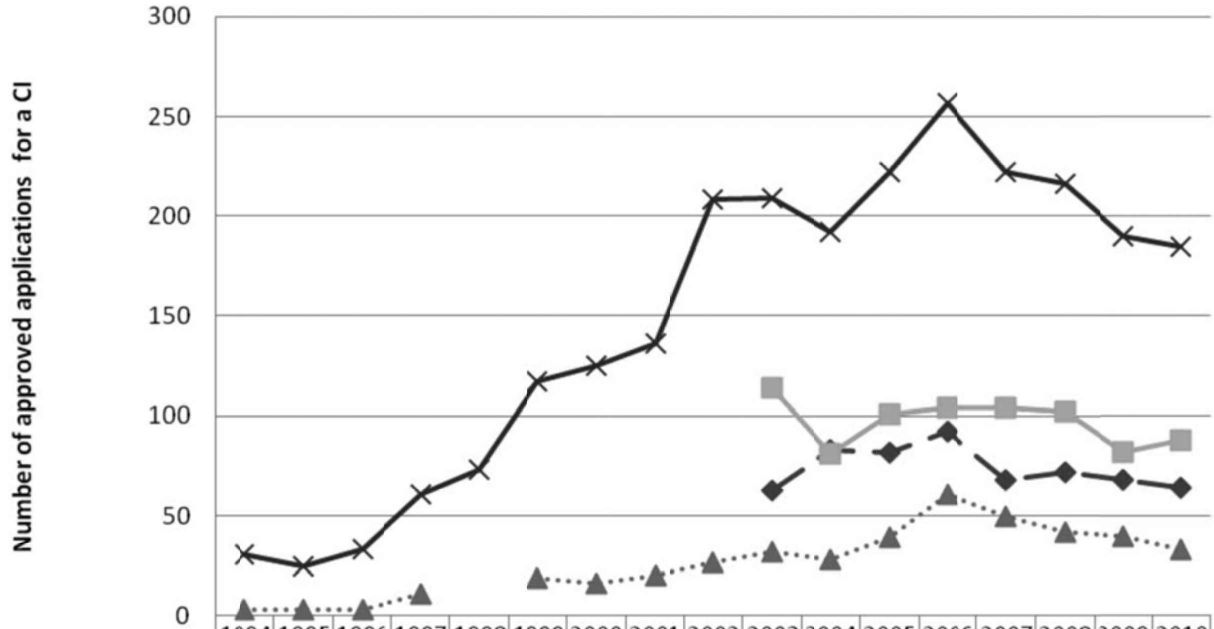
In CIs, the time coding is mimicked by varying the stimulation rate, whereas the place code is approximated by the electrode position. Psychophysical data have shown that this time code is limited to 300-500 Hz and the differences in electrode insertion depths and nerve survival patterns comprise the place code (Nelson et al, 1995, Collins et al, 1997, Zeng, 2002). At least four spectral channels are necessary for speech perception in quiet (Shannon et al, 1995), more than four spectral channels are necessary in noise (Fu et al, 1998), and at least 16 channels are needed for music (Smith et al, 2002). It should be noted that the number of effective or functional channels is not more than 4-8. Patients with low speech perception scores generally do not have more than four effective channels for any test, whereas patients with higher scores may have as many as eight or even slightly more channels, depending on the test (e.g. Friesen et al, 2001, Dorman and Spahr, 2006). As mentioned, contemporary CIs use between 12 and 22 intracochlear electrodes, so the number of electrodes exceeds the number of effective channels for practically all patients and for all current devices.

1.6 Cochlear implantation in Belgium

Belgium, and especially the northern part, Flanders, has been a center of expertise in CIs for many years. In 1998, as the first region in Europe and 2 years before the recommendations of the Joint Committee on Infant Hearing (JCIH, 2000) were published, the Flemish public child care organization ‘Kind en Gezin’ (Child and Family) started a Universal Neonatal Hearing Screening Programme (UNHSP) in Flanders (Verhaert et al, 2008).

In Belgium, CIs have been **reimbursed** for children and adults since October 1994 (Belgisch Staatsblad, 1994); initially only in patients with a bilateral total sensory deafness. In March 2006 (Belgisch Staatsblad, 2006), the reimbursement criteria were refined into (1) pure tone average thresholds of 85 dB HL or greater at 500, 1000, and 2000 Hz (best ear); (2) threshold of peak V in brainstem auditory evoked potentials at 90 dB HL or higher (best ear); (3) little or no benefit from hearing aids. In postlingually deafened subjects, results on a monosyllabic word test at 70 dB (phoneme score, with hearing aid) should be less than 30%. In case of mental retardation, psychological, or psychiatric problems, the family situation and the rehabilitation plan must be demonstrated in a psychological report. After implantation, long-term auditory/speech therapy and follow-up are required and reimbursed, until the age of 18 years for children and two years after CI for adults. This should be coordinated by a specialized multidisciplinary team.

Figure 1.4 gives an overview of the number of approved applications for a CI in Belgium from 1994 until 2010. Since 2003 detailed information with regard to the age of the CI candidate is available. It was found that between 2005 and 2010, on average, 68% of the pediatric CI candidates were younger than 2 years, 13% were children between 3 and 4 years, 9% were children between 5 and 7 years, and 11% were children between 8 and 12 years old (De Raeve and Wouters, 2013).



	1994	1995	1996	1997	1998	1999	2000	2001	2002	2003	2004	2005	2006	2007	2008	2009	2010
< 12 years										63	83	82	92	68	72	68	64
12-65 years										114	81	101	104	104	102	82	88
> 65 years	3	3	3	11		19	16	20	27	32	28	39	61	50	42	40	33
Total	31	25	33	61	73	117	125	136	208	209	192	222	257	222	216	190	185

Figure 1.4 Number of approved applications for a CI in Belgium, between 1994 and 2010 (copyright De Raeve and Wouters, 2013, printed with permission).

Since February 2010 (Belgisch Staatsblad, 2009) reimbursement for **bilateral** CI (sequential or simultaneous) has been provided for **children** (<12y). For children diagnosed with auditory neuropathy spectrum disorder (ANSD), bilateral CI can be foreseen between 12 months and 18 years. In case of meningitis bilateral CI is reimbursed until the age of 18 years. In 2010, 93% of severe-to-profound deaf preschool children (2.6-6yr) in Flanders received CIs and 25% is bilaterally implanted (De Raeve and Lichtert, 2012).

Information regarding the number of eligible **adults** receiving a CI is currently lacking. Furthermore, bilateral CI is not reimbursed in Belgium for adults. Adults considering bilateral CI must find financial support to reimburse implantation the contralateral ear. The exact number of bilaterally implanted adults in unknown, but is generally considered very low.

Some CI recipients continue to use their contralateral hearing aid (HA). This group is generally referred to as **bimodal** users (CI+HA).

The percentage of unilateral, bilateral, and bimodal pediatric and adult CI users at '*Universitair Ziekenhuis Gent'* (UZ Gent) is shown in Table 1.1.

Table 1.1
Percentage of unilateral, bilateral, and bimodal pediatric and adult CI users at Ghent University Hospital (2013)

	unilateral	bilateral	bimodal
Children (<18 yrs)	31%	42%	27%
Adults (>18 yrs)	64%	0%	36%

Chapter 2

Speech perception outcomes after cochlear implantation

2.1 Evaluation of speech perception outcomes after cochlear implantation

Determining the benefits of CI use in children and adults is essential to the clinical care of implant patients and to direct new research in the rehabilitation of deafness. The evaluation of speech perception abilities before and after implantation provides important clinical information about progress over time, about the need for adjusting the speech processor settings, about determining goals of aural rehabilitation, and, in the case of implanted children, about selecting appropriate educational programs. Therefore, clinical researchers must have a wide range of age-appropriate speech perception outcome measures available that allow them to evaluate a hierarchy of skills, ranging from detection and discrimination of vowel and consonant speech features through the comprehension of speech in complex listening environments (Iler Kirk, 2000).

Auditory assessment using speech stimuli has a long history in the evaluation of hearing. Konkle and Rintelmann (1983) defined speech audiometry as a procedure that uses speech stimuli to assess auditory functioning. Several materials can be used to assess speech perception such as monosyllabic words, sentences or even nonsense syllables.

The high level of auditory-only speech perception performance demonstrated by current CI subjects has necessitated a change in protocol for speech perception testing over the years.

This chapter describes the Dutch speech perception measures mostly used in Flanders (northern part of Belgium).

2.1.1 Evaluation of speech perception outcomes in children

2.1.1.1 Analytical tests in children

In this thesis analytical tests are tests which do not appeal onto semantical knowledge. Using analytical tests that include speech sounds bypasses a possible lack of language competency in young children and determines the basic levels of detection, discrimination, and identification. Both the Pediatric Auditory Performance Test for Flanders (PAP-V) (van Wieringen et al, 1998, van Wieringen and Wouters, 2005) and the Auditory Speech Sound Evaluation (A§E®) (Govaerts et al, 2006) can be applied. Detection implies being able to react to the presence of a sound. Discrimination requires the patient to detect a change in the acoustic stimulus, without the necessity to identify these stimuli. In addition, identification refers to the cognitive ability to attach a label to a stimulus (Govaerts et al, 2006, Thibodeau, 2009).

The advantages of these tests are that (1) analytical tests do not call on semantical knowledge and can be applied to very young children since word familiarity does not have to be taken into account, (2) results can be compared across languages, and (3) by using a closed-set task identification task, confusion matrices can be obtained to analyze the pattern of confusions of the child or adult (van Wieringen and Wouters, 2005).

2.1.1.2 Evaluation of speech perception outcomes in children using rating scales

Speech perception can be assessed using hierarchical **scales**. Currently, two scales are commonly used, namely the Listening Progress Profile (LiP) (Nikolopoulos et al, 2000a) and the Categories of Auditory Performance (CAP) (Archbold, 1995). These hierarchical scales can easily be understood by professionals and parents, and can be administered to a variety of age ranges (Archbold et al, 1995). However, it is a fairly global measure and the reduction of auditory performance to only a small number of levels implies little detail (Govaerts et al, 2002). In addition, speech perception can be assessed using speech audiometry. To date, Göttinger I/II (van Gompel and Vanhulle, 1979, Lambrechts, 1979) and NVA children (Wouters et al, 1994) are applied for assessing speech audiometry in Flemish speaking children.

2.1.1.2.1 Listening Progress Profile

The LiP is a profile devised to assess the early stage of auditory perception postimplantation to monitor the functioning of the device and the early development of speech and language (**Table 2.1**).

Table 2.1
Listening Progress Profile (LiP).

	Scoring
Response to environmental sounds	
Response to drum (elicited)	
Response to musical instrument (elicited)	
Response to voice-elicited	
Response to voice-spontaneous	
Discrimination between 2 different instruments	
Discrimination between loud and quit drum	
Discrimination between single and repeated drum	
Identification of environmental sounds	
Response to [oo, ah, ee, ss, mm]	
Discrimination between long and short speech sounds	
Single/repeated sounds	
Loud/quiet speech sounds	
Two of Ling five sounds	
All of Ling sounds	
Two family names of different syllable length	
Identification of own name in quiet	
	……/42

Scoring never= 0, sometimes= 1, always= 2

2.1.1.2.2 Categories of Auditory Performance

The CAP is a global outcome measure that assesses the auditory performance of deaf children. It comprises a nonlinear hierarchical scale of auditory receptive abilities. The lowest level describes no awareness of environmental sound and the highest level is represented by the ability to use the telephone with a known speaker (**Table 2.2**).

Table 2.2
Categories of Auditory Performance (CAP).

Category	CAP Description
7	Use of telephone with a familiar talker.
6	Understand conversation without lip reading with a familiar listener.
5	Understand common phrases without lip reading.
4	Discrimination of at least two speech sounds.
3	Recognition of environmental sounds.
2	Response to speech sounds.
1	Awareness of environmental sounds.
0	No awareness of environmental sounds.

2.1.1.3 Evaluation of speech perception outcomes in children using speech perception tests

2.1.1.3.1 Göttinger I and Göttinger II

Besides Göttinger I and II, the '*Antwerpen-Nijmegen testbatterij*' (Beijnon et al, 1992) is available for assessing speech audiometry in young Flemish speaking children. To date, mostly Göttinger I and II are used.

Göttinger I and II were originally developed in the German language in the late 1970s ('*Der Göttinger Kinderspracheverständnistest*'), but were translated and adapted by van Gompel and Vanhulle (1979) and Lambrechts (1979). The Göttinger I is developed for 3 to 4-yr old children, while the Göttinger II is meant for 5-6 yr olds. Both tests consist of **monosyllabic nouns** presented in a four-alternative closed-response set in the same vowel context. These words are represented by a figure. Children are asked to point the figure that represents the word they heard during testing. Both lists contain 10 series of 10 target nouns. Scoring is done per correctly identified word. Göttinger I and II are normalized for normally hearing toddlers in quiet (van Wieringen and Wouters, 2005) and in noise (van Wieringen et al, 2008). In addition, normative data are available for toddlers with severe SNHL (van Wieringen et al, 2008).

2.1.1.3.2 NVA children

The '*Nederlandse Vereniging voor Audiologie'* (NVA) children test can be administered to children between 6 and 10 years. Older children can be tested with the standard NVA. The NVA children test consist of five lists, containing 12 **Consonant-Vowel-Consonant (CVC) words**, of which the first word is not scored. This test was originally developed by Bosman et al (1992), but a Flemish version was presented soon after (Wouters et al, 1994). Scoring is done per correctly identified phoneme, since children are asked to repeat the target word or, if not completely understood, to repeat as many phonemes as possible.

2.1.2 Evaluation of speech perception outcomes in adults

2.1.2.1 Analytical tests in adults

Based on Miller and Nicely (1955) and van Wieringen and Wouters (1999), closed-set identification of consonants (C) in a nonsense intervocalic /a/ context can be tested. Hence, consonant confusion matrices can be analyzed to examine the auditory possibilities and limitations of each subject. By means of these confusion matrices, insight can be gained into the distribution of errors among the off-diagonal cells. Therefore, stimuli are combined into groups in such a way that confusions within groups are more likely than confusions between groups.

In this doctoral dissertation 12 /aCa/ nonsense syllables with C being /m, n, p, b, k, s, z, f, v, t, d and ɣ/ and five consonant features (voicing, place, affrication, manner, and nasality, based on van Wieringen and Wouters, 1999) were implemented.

To examine the off-diagonal confusions, Miller and Nicely (1955) implemented an algorithm. For details of this algorithm, the interested reader is referred to the original paper of Miller and Nicely (1955) or a more recent paper of Christiansen and Greenberg (2011).

After the implementation of this algorithm, **Information transmission scores** (ITr) can be computed to examine how spectral, temporal and amplitude cues are perceived by the individual subjects. The relative ITr score lies between 0 and 1 (logarithmic).

2.1.2.2 Evaluation of speech perception outcomes in adults using questionnaires

2.1.2.2.1 APHAB

The Abbreviated Profile of Hearing Aid Benefit (APHAB) **questionnaire** is a 24-item self-assessment inventory in which patients report the amount of trouble they have with communication or noises in various everyday situations. Benefit is calculated by comparing the patient's reported difficulty in the unaided condition with their amount of difficulty using amplification. This questionnaire makes use of 4 subscales: ease of communication, reverberation, background noise, and aversiveness (Cox and Alexander, 1995).

2.1.2.3 Evaluation of speech perception outcome in adults using speech perception tests

2.1.2.3.1 NVA

The '*Nederlandse Vereniging voor Audiologie'* developed the validated NVA speech test. This speech perception task is an open-set test using Dutch CVC 3 phoneme **monosyllabic** words (Wouters et al, 1994). Fifteen lists consisting each of 12 CVC words uttered by a male speaker, are available. Scoring is done by correctly identified phoneme and thus, the first word is not scored.

2.1.2.3.2 BLU-list

Besides monosyllabic words, **spondees** can be used to gain insight in speech perception outcome. In Dutch, the BLU (Brugge-Leuven-Utrecht) list is the best-known spondaic word list, consisting of 15 lists of 10 words each. Every word is a compounded word and every syllable is a separate word. The BLU-list is suited to determine the SRT based on a word score (10% for each correctly repeated word) (Van de Calseyde et al, 1971).

2.1.2.3.3 LIST and LINT

The most well-known Dutch **sentence** material is that of Plomp and Mimpen (1979). However, because of its extensive use and because several words were outdated, van

Wieringen and Wouters (2008) developed and validated a new set of sentence material, the Leuven Intelligibility Speech Test (LIST). The LIST consists of 35 lists of 10 sentences of equal known difficulty uttered by a female speaker. Furthermore, a number test was also made available, the Leuven Intelligibility Number Test (LINT) (van Wieringen and Wouters, 2008). Because of its simplicity, closed response format (numbers 1-100 are included), and lack of learning effect this test can be applied to speakers of a foreign language, people with limited education or those with a mild mental handicap. The LINT consists of 400 numbers (number 1 to 100), uttered by two male and two female speakers. For both tests, normative values were determined at fixed signal-to-noise ratios (SNR).

2.2 Variables affecting speech perception outcomes after cochlear implantation

2.2.1 Introduction

Since the formal introduction of United States FDA approved single-channel CIs in 1984 for **adults**, significant improvements have been made in CI technology, speech coding strategies, and speech perception performance. With the first generation of CIs, speech perception goals were modest, that is, open-set speech recognition was not anticipated for the vast majority of implant users. In contrast, current CI patients are even outperforming many successful HA users with less severe HL (Dorman, 2007). In fact, relatively recent estimates of monosyllabic words recognition for adult postlingually deafened CI users ranges from 10% to 80% with a mean of approximately 50-60% correct (Helms et al, 1997, Parkinson et al, 2002, Firszt et al, 2004, Holden et al, 2013). (Gifford et al, 2008).

Since the Federal Drug Administration (FDA) approval for multichannel CIs in 1990 for **children**, (as young as 2 yrs) a comparable trend was seen. Early results mainly focused on the ability to improve speech reading or to obtain benefit in closed-set situations (Osberger et al, 1991). Afterwards, the focus shifted from postoperative performance on closed-set tests to the ability to perform on open-set measures (Waltzman, 2006 in Waltzman and Roland). Nowadays, monosyllabic word scores on tests developed for the pediatric population, show scores ranging from 0% to 100% at 1-yr postimplantation, with pediatric CI recipients continuing to outperform children with classical HAs (Meyer et al, 1998).

Because of an increased performance in profoundly deaf subjects, initially narrow candidacy criteria have expanded to include patients with multiple handicaps (e.g. Berrettini et al, 2008), abnormal cochlear anatomy (e.g. Dettman et al, 2011), residual low-frequency hearing (e.g. Turner et al, 2008) and even patients with unilateral deafness (e.g. Kamal et al, 2012). The majority of adult and pediatric implant recipients achieve open-set speech understanding in quiet, nevertheless, wide variation in individual outcomes has been reported at all implant centers around the world.

Understanding the factors that contribute to a favorable outcome after CI would potentially allow clinicians to predict the expected results for an individual CI patient. In addition, such knowledge could also allow adjustment of postimplantation rehabilitation to maximize the benefits of receiving a CI. Several studies have been undertaken to identify the factors that influence speech perception outcomes after cochlear implantation. The following chapter aims to highlight well-recognized variables known to affect CI performance in children and adults.

2.2.2 Factors affecting speech perception outcomes in pediatric CI users

Table 2.3 gives an overview of the studies conducted since 2000 that shed light on (several) variables possibly influencing speech perception outcome in pediatric CI users. Because of a large number of studies conducted within this field of research, this list is not exhaustive. A distinction was made between factors related to device characteristics and individual patient characteristics. Since in the pediatric population, most recent research focuses on patient characteristics, only patient related variables will be discussed in the following paragraph.

It is widely accepted that **age at implantation** is a major factor influencing outcome after CI. Some authors conclude that cochlear implantation should take place before the first birthday (Miyamoto et al, 2003, Dettman et al, 2007, Ertmer et al, 2007), whereas others state children should be implanted before their second (Svirsky et al, 2004, Tait et al, 2007, Hayes et al, 2009) or third birthday (Iler Kirk et al, 2002, Johnson and Goswami, 2010) (Boons et al, 2012). Vlastarakos and colleagues (2010) performed a systematic review and meta-analysis regarding auditory perception and speech production outcomes in infants implanted before the age of two. They state that evidence of these children’s outperformance regarding auditory perception and speech production is limited because wide-range comparisons between infant implantees and under-2-year old implanted children are sparse. Furthermore, the follow-up period included in recent studies is relatively short (mean follow-up duration between 6 and 12 months). Also, there is a need to develop and validate robust measures of monitoring implanted infants. Possibly, the effect of early implantation will become more evident in reading, writing or academic achievements. Such skills are typically tested in older children. Hence, a clear cut-off for the age at which children should be implanted, is still unknown and is subject of future research.

Because early implantation is depending on **early identification** of HI children, a newborn hearing screening program is essential (O'Donoghue et al, 2000, Richter et al, 2002). Since 1998 an integrated universal newborn hearing screening (UNHS) program based on automated auditory brainstem response (AABR) has been implemented in Flanders by the federal health care agency. By means of the Algo® Portable, which delivers 100 µs clicks with an intensity of 35 dB HL and has a frequency content ranging from 700 to 5000 Hz, 98% of all neonates are offered hearing screening at the age of 4 weeks. Data from the first 5 yrs of follow-up showed that 1.45‰ of the screened infants had a congenital HL (Van Kerschaver et al, 2007, Verhaert et al, 2008). Declau and colleagues (2008) investigated 170 consecutive records of neonates referred to a tertiary center after failure on UNHS. Bilateral HL was confirmed in 68 children and unilateral HL in 48 children, together accounting for a confirmed HL in 68% of referred children. Hence, the 2007 position statement of the Joint Committee of Infant Hearing indicating infants with significant congenital HL should be identified by 3 months of age is taken into account.

Communication mode has also been frequently reported to be a factor that contributes to speech perception outcome with oral-only communication producing speech perception results superior to those observed in children who use total communication (i.e. a combination of spoken language and sign language) or sign language. (O'Donoghue et al, 2000, Osberger et al 2002, Geers et al, 2003, Taitelbaum-Swead et al, 2005, Rotteveel et al, 2008, Percy-Smith et al, 2010, Dettman et al, 2013). Emerging literature suggests that when the child learns language visually (i.e. via sign language) the cortical connections are organized accordingly to the visual input. The application of an auditory device, such as a CI, at a later stage, would necessitate cortical reorganization to the auditory stimulus. Possibly, placing such cognitive demands on a child to learn (visually) and after CI, unlearn and relearn (auditory) does place lifetime restrictions (Dettman et al, 2013).

Another factor that accounts for a proportion of the variability is the presence of **additional disabilities.** Implanted children with additional disabilities tend to progress slower and even achieve lower scores on speech perception outcomes compared to implanted children without additional disabilities (e.g. Pyman et al, 2000, Berrettini et al, 2008). However, improved quality of life and increased social interactions are found within the majority of this special group of implanted children (e.g. Waltzman et al, 2000).

In addition, **etiology** of deafness affects speech perception in pediatric CI subjects (e.g. Hang et al, 2012). Children diagnosed with a Connexin 26 gene (Cx26) mutation generally perform equally (e.g. Dahl et al, 2003) or better than non-Cx26 peers (e.g. Wu et al, 2011), whereas children with congenital cytomegalovirus (cCMV) tend to lag behind (e.g. Ramirez-Inscoe et al, 2004, Ciorba et al, 2009, Yoshida et al, 2009). Children with cochlear and/or vestibular abnormalities and meningitis on average have higher incidences of partially inserted electrodes, which may lead to reduced results (e.g. Rotteveel et al, 2005, Dettman et al, 2013). Less clear are the outcomes in children diagnosed with auditory neuropathy spectrum disorder (ANSD) since their results vary greatly. Among their clinical characteristics reported are speech difficulties that are disproportionate to their hearing threshold levels (Starr et al, 1996) and difficulties understanding speech in noise (e.g. Kraus et al, 2000, Rance et al, 2007). Many ANSD-children achieve similar outcomes with matched non-ANSD peers, while others demonstrate a continued delay (e.g. Rance et al, 2009, Berlin et al, 2010, Teagle et al, 2010, Breneman et al, 2012). Roush et al (2011) conducted a systematic literature review and included 15 studies gaining insight on CI outcome in ANSD-children. It seems that children with cochlear nerve deficiency as the cause of ANSD achieve poorer results, although further research is needed. Especially, considering the heterogeneity of the ANSD-population and the varied etiologies associated with ANSD (e.g. prenatal, genetic (e.g. OTOF), and neurological), there is need for additional studies with more homogeneous grouping of ANSD-children to assess the impact of various comorbidities on auditory, speech, language, and hearing outcome.

Table 2.3
Literature overview of conducted studies predicting speech perception outcomes in pediatric cochlear implant users.

Author(s)	**N**	**Device related factors**	**Patient related factors**	**Results**
			2000	
Dawson et al	17	/	(1) electrode discrimination ability	(1) Electrode discrimination ability is the strongest factor in accounting for variance in the speech perception scores.
Loundon et al	40	/	(1) communication mode, (2) age at CI (mean 7 yrs), (3) audiologic status prior to CI	(1) Prognostic factors for good results are: good level of oral communication before implantation, residual hearing, progressive deafness and early implantation. (2) Poor prognostic factors are: presence of behavioral disorders, poor communication skills prior to CI.
Kuo and Gibson	45	/	(1) residual high-frequency hearing prior to CI, (2) age at CI (4 groups: 3-5yrs/6-9yrs/10-15yrs)	(1) Older recipients experience more difficulty than younger children during the initial periods of device use. (2) High-frequency hearing preoperative correlates with more rapid improvement.
O'Donoghue et al	40	/	(1) age at CI (mean 4,3 yrs), (2) number of inserted electrodes, (3) origin of deafness, (4) mode of communication, (5) socioeconomic group	(1) Young age at CI and oral communication mode are the most important determinants of speech perception
Nikolopoulos et al (b)	47	/	(1) preoperative prom-EABR	(1) Children without prom-EABR performed equally as those with clear promontory responses preoperative.
Pyman et al	75	/	(1) etiology, (2) central pathology	(1) Influence of motor and/or cognitive delays on development of speech perception skills. (2) No influence of etiology.
			2001	
Kileny et al	53	/	(1) age at CI (range 2,4-14,5yrs), (2) length of CI use	(1) Better performance with longer CI use and younger age at CI
Sarant et al	167	(1) speech processor	(1) etiology, (2) clinic, (3) onset of deafness, (4) ear implanted, (5)	(1) The independent variables accounted for 34% of variance in word perception scores.

			communication mode	
			2002	
Richter et al	106	/	(1) age at CI (mean 5,08 yrs), (2) additional disabilities, (3) speech production	(1) Age at CI is the most important prognostic factor
Dowell et al	25	/	(1) age at CI (range 8-18 yrs)	(1) Preoperative open-set sentence score, duration of profound HL and equivalent language age accounted for 66% of the variance.
Hammes et al	47	/	(1) age at CI < 18mo	(1) Better outcome if implanted before 18 months of age.
Iler Kirk et al	73	/	(1) age at CI (Ci before age 5 yrs)	(1) Better outcomes if implanted before age at 3 yrs. (2) Oral communication demonstrates more rapid gains in communication abilities.
			2003	
Pulsifer et al	40	/	(1) age at CI (mean age at CI: 50.72m (SD 27.66m)	(1) Young age at implantation (< 48m) correlates with better speech perception.
Oh et al	29	/	(1) metabolic status of brain cortices determined by means of 18F-fluorodeoxyglucose positron emission tomography	(1) Wider hypometabolic area correlates with better speech perception.
Geers et al	181	(1) number of active electrodes, (2) MAP dynamic range, (3) loudness growth, (4) loudness growth variability	(1) child and family characteristics, (2) educational characteristics	(1) Predictors of better speech perception are :greater nonverbal intelligence, smaller family size, longer use of the updated SPEAK/CIS processing strategy, a fully active electrode array, greater electrical dynamic range and greater growth of loudness with increasing stimulus intensity. (2) After controlling for these variables, oral-aural communication correlates with good speech perception.
Pisoni and Cleary	176	/	(1) forward digit span measures of verbal working memory	(1) Speed and efficiency with which phonological and lexical representations of spoken words are maintained in and retrieved from working memory account for 20% of variance in speech

				perception. (2) Working memory capacity accounts for 7% of variance.
			2004	
Kaplan-Neeman et al	10	(1) NRT-based mapping versus behavioural mapping	/	(1) No significant differences among NRT-based MAPs and behavioral-based MAPs.
Manrique et al	130	/	(1) age at CI (4 groups: 0-3/4-6/7-10/11-14yrs)	(1) Better improvement if implanted before 2 yrs of age.
Svirsky et al	75	/	(1) age at CI (3 groups: <2yrs/2-3,5 yrs/3,5-5yrs)	(1) Better speech perception results if implanted before 2 yrs of age.
Schauwers et al	10	/	(1) age at CI (range 5-20m)	(1) The earlier the implantation took place, the smaller the delay was in comparison with normally hearing children with regard to the onset of prelexical babbling and with regard to auditory performance as measured by CAP.
Zwolan et al	295	/	(1) age at CI (5 groups: 1–3 years, 3–5 years, 5–7 years, 7–9 years, and 9–11 years)	(1) Early implantation facilitates improved development of speech perception.
Arnoldner et al	6	/	(1) inner ear malformations	(1) No differences between groups with and without inner ear malformations.
Nikolopoulos et al	51	/	(1) age at CI (< 6yrs), (2) duration of deafness, (3) family and support services, (4) expectations, (5) children's cognitive abilities, (6) learning style	(1) Learning style explaining up to 29% of the variance. (2) Shorter duration of deafness, young age at implantation and family structure/support correlate with good speech perception.
			2005	
Gordon et al	23	/	(1) cortical waveforms postoperative	(1) Atypical types of responses correlate with poorer behavioral speech perception.
Rotteveel et al	25	(1) partial insertion	(1) meningitis	(1) Partial insertion correlates with poorer speech perception.
Taitelbaum-Swead et al	96	(1) commercially available speech processors	(1) age at CI (Nucleus 3,6yrs, AB 3,5 yrs, Medel 7,6 yrs), (2) mode of communication	(1) No differences in speech perception performance between implant devices when considering background variables. (2) Young age at CI and oral communication mode correlate with

				better speech performance.
Horn et al	47	/	(1) behavioural inhibition skills	(1) Correlation between delay task performance and speech perception.
Anderson et al	18	(1) short electrode array	/	(1) The short electrode children do not perform as well as the standard children initially, but do tend to catch-up at later test intervals.
Lee et al	11	/	(1) preoperative (18)F-FDG-PET	(1) Greater dependency on the visual function due to auditory deprivation may interfere with acquisition of auditory language after CI.
			2006	
Wu et al	28	/	(1) age at CI (2 groups: < 3 yrs/ > 3 yrs)	(1) Better speech perception if implanted before 3 yrs of age.
Edwards et al	32	/	(1) general development and cognitive functioning, (2) IT-MAIS, (3) age at switch-on (range 1.2-2.8 yrs), (4) aided HL pre-CI	(1) General development and cognitive functioning, accounting for around 40% of the variance in outcomes. (2) Significant developmental delay is predictive of poor outcomes, but children with a mild delay do make appreciable progress
			2007	
Uziel et al	82	/	(1) age at CI (mean 4,8 yrs), (2) additional disabilities, (3) education, (4) communication mode, (5) active electrodes	(1) Age at implantation is the most important factor influencing post-CI outcome.
Wolfe et al	12	/	(1) sequential BICI	(1) Bilateral CI allowed for better speech recognition in noise relative to unilateral performance. (2) Children receiving bilateral CIs younger than 4 yrs of age achieved better speech recognition in quiet performance for the later implanted ear as compared with children receiving their second CI after 4 yr of age.
Galvin et al	11	/	(1) sequential BICI	(1) Children over age 4 yr may gain significant additional benefit from a second implant, including improved speech perception in some noise contexts and functional advantages in daily life.

			2008	
Rotteveel et al	67	/	(1) age at CI (3 groups <4yrs/4-6yrs/>6 yrs), (2) duration of deafness, (3) communication mode	(1) Duration of deafness correlates to speech perception performance. (2) Mode of communication is also a major factor.
Francis et al	188	/	(1) percent active channels	(1) Activation of the entire electrode array is associated with better early auditory outcomes.
Berrettini et al	23	/	(1) additional disabilities	(1) Variable, but overall, positive results in CI children with additional disabilities.
Profant et al	30	/	(1) age at the time of diagnosis, (2) age at the time of the first HA fitting, (3) age at CI (ES 26,4 m vs LS 32,6 m)	(1) Early screened and diagnosed children have better outcomes.
Wu et al	67	/	(1) genetic diagnosis, (2) imaging	(1) Better outcome if SLC26A4 or GJB2 mutations compared to narrow internal auditory canal as etiology of deafness.
Durisin et al	60	/	(1) meningitis, (2) duration of deafness	(1) Better auditory performance if implanted within 6 months after meningitis.
			2009	
Edwards et al	127	/	(1) communication skills, (2) cognitive abilities, (3) family structure and support, (4) use of HAs pre-CI	(1) Scores on tests of fluid reasoning skills (sequencing) correlates to speech perception.
Artieres et al	74	/	(1) age at CI (4 groups <2yrs, 2-3 yrs/3-4yrs/4-5 yrs), (2) duration of CI use, (3) preoperative hearing levels, (4) age of HA fitting, (5) chronological age	(1) Positive impact of early implantation, duration of CI use and preoperative hearing status on receptive language.
Scherf et al	35	/	(1) sequential bilateral CI	(1) Better outcomes in BICI children compared to uni-CI children even if CI2 received after age 6 yr. (2) Longer adjustment period needed for older children to gain benefit.

			2010	
Tajudeen et al	117	/	(1) age at CI (3 groups: <1yrs/<2yrs/<3yrs)	(1) The sensitive period for developing word identification seems to extend at least until age 3 yr.
Gerard et al	89	/	(1) etiology, (2) additional disabilities, (3) residual HL prior to CI	(1) Communication ability scores increases with high socioeconomic level, presence of residual hearing, younger patients when no residual hearing, connexin-26 mutation related deafness, and absence of associated disabilities.
Sininger et al	44	/	(1) age at HA fitting, (2) degree of HL in the better ear, (3) CI status, (4) intensity of oral education, (5) parent-child interaction, (6) number of languages spoken at home	(1) Better outcomes if early amplified.
Percy-Smith et al	155	/	(1) hearing age, (2) age at CI (mean 3 yrs), (3) gender, (4) educational placement, (5) ear of implantation, (6) CI center, (7) communication mode	(1) Better outcomes if communication mode at home is spoken language, as opposed to a mixture of spoken language/sign language or purely sign language.
			2011	
Dettman et al	48	/	(1) cochlear and/or vestibular abnormality	(1) Correlation between cerebrospinal fluid gusher and partial electrode insertion, fewer active electrodes and poorer sentence understanding.
			2012	
Moon et al	177		(1) MRI abnormalities preoperative	(1) Slower progress and reduced performance if MRI abnormalities.
Zhou et al	44	(1) surgical technique	/	(1) No differences in outcome between implantation techniques.
			2013	
Janeschik et al	163	/	(1) etiology	(1) Better performance if connexin-26 mutation, poorer if Usher and CHARGE-syndrome. (2) Post-meningitic and post-septic children develope slower but reache same levels later.

Leigh et al	35	/	(1) age at CI (2 groups: 6-12m/13-24m)	(1) Better outcome if implanted before 12 mo of age.
Dettman et al	39	/	(1) communication mode	(1) Better results if emphasis on oral/aural input.
Caldwell and Nittrouer	54	/	(1) phonological awareness, (2) general language, (3) cognitive skills	(1) Noise effects were consistent across NH and CI groups. (2) Scores on independent variables included did not explain any group differences in speech recognition.

yr= year, CI=cochlear implant, NRT= Neural response telemetry, NH=normal hearing, MRI= magnetic resonance imaging, HL=hearing loss, HA=hearing aid, uni=unilateral, BICI= bilateral cochlear implants, IT-MAIS= Infant-Toddler Meaningful Auditory Integration Scale, PET= position emission tomography, CAP= Categories of Auditory Performance, EABR= electrically evoked auditory brainstem responses, SPEAK=spectral peak, CIS= continuous interleaved sampling.

2.2.3 Factors affecting speech perception outcomes in adult CI users

In the past decade, data on postoperative outcomes following CI in adults have identified a wide spectrum of variables known to affect postimplantation performance. Studies that investigated these factors since 2000 are listed in **Table 2.4**. As for the pediatric population, these variables relate to device characteristics such as electrode design and speech processing strategies on the one hand and individual patient characteristics such as duration of deafness and etiology on the other hand.

Since the introduction of CI, advances in hardware, software, and **speech processing technology** have directly affected performance, successively improving postimplantation speech perception with each significant technologic advance. Upgrades to spectral peak (SPEAK) from multipeak (MPEAK), and from SPEAK to continuous interleaved sampling (CIS) saw concomitant improvement in postoperative speech perception. Research on novel processing strategies is still ongoing, the goal being to improve speech perception in noise, music perception and speech perception of tonal languages. During the past several years, increasing attention has been devoted to representing 'fine structure' information with CIs (e.g. Smith et al, 2002, Nie et al, 2005, Wilson et al, 2005, Zeng et al, 2005, Hochmair et al, 2006, Arnoldner et al 2007, Berenstein et al, 2008, Litvak et al, 2008). One of these new speech processing strategies is HiRes Fidelity 120 ® (HiRes 120®) which makes use of virtual channels. By varying the proportion of current delivered to each electrode of an electrode pair, 'current steering' can increase the number of spectral (virtual) channels beyond the number of physical electrodes provided by the HiRes 90K® CI array. Hence, 16 active electrodes (and 15 electrode pairs) can be used to create eight additional stimulation sites, resulting in up to 120 virtual channels. Furthermore, multiple methodologies attempting to minimize current interactions between electrodes have been investigated, including the use of monopolar, tripolar and even quadrupolar techniques for virtual channel creation (e.g. Srinivasan et al, 2013). However, significant clinical correlation between these newly investigational processing strategies has yet to be definitively demonstrated.

One of the clearest device-related factors that positively influences speech perception outcome is the implementation of **multi-channel implants**. It is not surprising that relatively few patients could obtain open-set speech understanding with single-channel implants given

the limited spectral information (Loizou, 1998). However, the exact number of electrodes that has to be applied in multi-channel CIs to obtain high levels of speech understanding is still subject to debate (e.g. Dorman and Loizou, 1997). Research has shown that understanding in quiet requires only four spectral channels (e.g. Shannon et al, 2004), whereas speech perception in a more difficult listening environment requires more channels. For example, 8 to 10 spectral channels are needed for speech perception in noise (e.g. Friesen et al, 2001). Although modern CIs have between 12 and 22 physical contacts, CI listeners perform as if they are only receiving between 4 and 8 independent channels of information (Friesen et al, 2001). This limited spectral information is thought to be due to **channel interaction** across stimulated electrodes, which arise from overlapping populations of activated neurons (e.g. Fu et al, 1998, Fu and Nogaki, 2005). Reducing the spread of excitation from a stimulated electrode would narrow the population of activated neurons and would therefore presumably reduce channel interaction. Possibly, reduced spread of excitation might enhance speech perception (e.g. Bierer et al, 2007, Srinivasan et al, 2013).
In addition, an inverse relationship is observed for the percentage of **electrodes** in the scala vestibuli as opposed to the scala tympani and **depth of insertion** of the electrode array and speech perception performance (Finley et al, 2008, Holden et al, 2013). Holden and colleagues (2013) showed that electrode location in the scala vestibuli occurred in all six outcome groups (with group 1 being the lowest performers and group 6 being the highest performers 2-yrs postimplantation on a speech perception task), but occurred more often and with greater degree as group performance declined. Furthermore, length of insertion along the array trajectory and the angular position of the basal-most electrode were significantly and inversely related to speech perception outcome. Deeper insertions occurred more frequently and to a greater degree in lower-performing subjects. Together, length of insertion and angular position, explained 92% of the variance on a speech perception task 2-yrs postimplantation.

Also patient-related factors have to be taken into account. **Duration of deafness** has been found to have a negative effect on speech perception (e.g. Blamey et al, 1996, Gomaa et al, 2003, UK Study Group, 2004, Leung et al, 2005, Green et al, 2007, Holden et al, 2013). Although length of deafness is inarguably an important factor related to CI outcome, age at CI as a factor predictive of outcome in postlingually deafened adults is divisive among studies. Some authors state that there are no differences between younger and older CI recipients (e.g. Shin et al, 2000, Pasanisi et al, 2003, Haensel et al, 2005, Hiraumi et al, 2007, Williamson et al, 2009), whereas others do find that younger CI recipients perform better in comparison to older peers (e.g. Chatelin et al, 2004, Lalwani et al, 2009, Friedland et al, 2010, Budenz et al, 2011, Lin et al, 2012, Holden et al, 2012). In addition, **onset of deafness** is a clear predictor of speech perception outcome since prelingually deafened adults on average perform worse than postlingually deafened recipients (e.g. Santarelli et al, 2008, Caposecco et al, 2012).

In recent years, more research has been conducted regarding **central auditory pathways and cortical function** (e.g. Giraud and Lee, 2007, Strelnikov et al, 2010, Lazard et al, 2013). Heydebrand and co-workers (2007) demonstrated that verbal learning, a specific cognitive skill, predicted 42% of the variance in 6 months postimplant monosyllabic words. Moreover, when combined, verbal learning, baseline monosyllabic word performance (measured at 2 weeks postinitial CI switch-on), and lipreading ability accounted for 82% of the variance in 6 month word scores. Lazard and colleagues (2010) acquired functional Magnetic Resonance Imaging (fMRI) data during a phonological task prior to CI surgery and related these results to 6 months postoperative word recognition scores. They identified two neurofunctional traits that prospectively distinguish good and poor CI performers based on their approach to written language. While the maintenance of a normal 'dorsal' phonological circuit in the course of deafness predicts a good speech perception outcome after CI, the switch to an alternative lexico-semantic 'ventral' route and the recruitment of the right supramarginal gyrus to compensate for deficient phonological processing suggests a poor speech perception outcome. Blamey and co-workers (2013) gained insight in factors affecting speech perception outcome postimplantation. Based on results of 2251 adult CI recipients, they stated that known physiological processes (e.g. malformation of the cochlea) are still relevant, however, plasticity changes and degeneration of central auditory processing play a much more important role than peripheral factors (e.g. number of surviving spiral ganglion cells).

Table 2.4
Literature overview of conducted studies predicting speech perception outcomes in adult cochlear implant users.

Author(s)	N	Device related factors	Patient related factors	Results
2000				
Kiefer et al	13	(1) stimulation rate, (2) number of channels	/	(1) Lower stimulation rate results in lower performance. (2) Number of active channels below 7 or 8 decreases performance.
Shin et al	27	/	(1) age at implantation>60yr	(1) Equal results obtained in older and younger adult CI users
Fu et al	6	(1) stimulation rate	/	(1) Stimulation rates lower than 150 pulses/s/electrode produce poorer performance.
2001				
Groenen et al	9	/	(1) speech-evoked cortical potentials	(1) P300 amplitude and magnitude and /i/-/a/ contrasts and behavioural speech recognition are related
Stollwerck et al	55	(1) speech processing strategy	/	(1) 25% of subjects preferred SAS and 75% CIS. (2) Subjects performe better in their strategy of choice.
2002				
Shannon	review	(1) temporal cues, (2) spectral cues	/	(1) Only four spectral channels can produce good speech understanding, but more channels are required for difficult listening situations.
Firszt et al	11	/	(1) electrically evoked potentials	(1) The inability to follow changes in the temporal characteristics is associated with poor speech perception. (2) Variability in speech perception scores relates to neurophysiologic responses at higher cortical levels of the auditory pathway.
Skinner et al	26	(1) insertion depth	/	(1) Insertion depth is correlated with speech perception. (2) Lower scores are related to low spiral ganglion cell survival.
Fu	9	/	(1) modulation detection threshold	(1) Phoneme recognition scores and mean modulation thresholds are correlated
2003				
Hamzavi et al	66	(1) number of electrodes, (2) speech processor, (3) insertion depth, (4) reimplantation	(1) duration of deafness, (2) etiology	(1) No correlation between independent variables and outcome.

Gomaa et al	60	/	(1) duration of deafness, (2) preoperative sentence recognition	(1) Shorter duration of deafness implies better speech perception. (2) Better preoperative performance implies better speech perception.
Pasanisi et al	16	/	(1) age at implantation>65yr	(1) Older patients obtain significant benefits from CI
			2004	
UK CI Study group	480	/	(1) preoperative speech perception score aided	(1) Effectiveness was not strongly associated with age at the time of implantation. (2) CI is least effective when preoperative audiological status of the better-hearing ear is good and duration of deafness of implanted ear is long.
Chatelin et al	65	/	(1) age at implantation > 70yr	(1) Older patients perform less well than younger patients
Marrinan et al	28	(1) degree of modiolar coiling of electrode array	/	(1) No significant correlations between the degree of modiolar coiling of the electrode array and speech perception.
Collison et al	15	/	(1) linguistic abilities, (2) cognitive abilities	(1) No correlation between cognitive and linguistic abilities and speech perception.
			2005	
Leung et al	749	/	(1) duration of deafness, (2) age at CI	(1) Duration of profound deafness and residual speech recognition carry higher predictive value than the age at CI.
Haensel et al	26	/	(1) age at implantation > 65yr	(1) No significant differences between older and younger patients.
Francis et al	43	/	(1) residual hearing in the nonimplanted ear	(1) Presence of residual hearing in one/both ears is associated with higher postoperative speech perception compared with participants with bilateral profound HL. (2) If similar amounts of residual hearing in the non-implanted ear occur, there is no difference in speech recognition between those with profound and those with severe HL in the implanted ear.
Kelly et al	12	/	(1) duration of deafness, (2) auditory evoked potentials	(1) Duration of deafness correlates with speech scores with poor scores reflecting greater years of deafness. (2) MMN was absent or degraded in CI subjects with poor speech scores.
Pfingst et al	17	/	psychophysical measures (1) maximum comfortable loudness levels, (2) dynamic ranges	(1) Subjects with high mean C levels and large mean DRs have better speech recognition.

		2006		
Fayad et al	14	/	(1) cochlear neuronal survival	(1) No correlation between performance variability and cochlear neuronal survival.
Hong and Turner		/	(1) auditory stream seggregation	(1) Better stream segregation is associated with better understanding of speech in noise.
Stickney et al	8	(1) electrode array, (2) electrical field interactions	/	(1) Electrical-field interaction accounts for 70% of the variance in speech recognition.
Orabi et al	38	/	(1) age at implantation >65yr	(1) No correlation between age at CI and speech performance.
Umat et al	13	/	(1) intensity on pitch	(1) Spectral smearing or underlying cognitive abilities might affect speech perception.
		2007		
Green et al	117	(1) implant type, (2) speech processor strategy	(1) age at CI, (2) sex, (3) duration of HL, (4) etiology of HL, (5) residual hearing, (6) number of active electrodes inserted	(1) Duration of deafness prior to implantation is an independent predictor of implant outcome.
Hiraumi et al	109	/	(1) age at the operation, (2) duration of deafness, (3) number of electrodes outside the cochlea	(1) No correlation between cochlear obstruction and speech performance. (2) No impact of onset of deafness (progressive vs sudden) on speech performance.
Heydebrand et al	55	/	(1) cognitive ability preoperative	(1) General cognitive ability nor processing speed is correlated with outcome. (2) Verbal learning scores and lip-reading skill account for 72% of the speech perception variance.
Morris et al	101	/	(1) ear of implantation	(1) No differences between left-ear- and right-ear-implanted patients in improvement on speech perception.
Arnoldner et al	18	(1) electrode design	/	(1) Higher distance between contacts in the apical part show improved speech performance.
Bierer	9	(1) electrode stimulation	/	(1) Speech perception correlates with channel-to-channel variability in tripolar threshold: greater variability relates to poorer performance.

			2008	
Helbig et al	9	/	(1) EAS	(1) Better performance in noise with EAS.
Finley et al	14	(1) electrode placement	(1) subject demographic descriptors, (2) apical pitch discrimination	(1) Lower outcome if greater insertion depth and greater number of contacts being located in scala vestibule.
Buss et al	26	/	(1) simultaneous BICI	(1) Benefit of BICI when speech and masker are spatially coincident and when they are spatially separated.
Santarelli et al	18	/	(1) onset of deafness	(1) Preoperative auditory input associated with auditory-oral therapy and a good level of education may positively influence the outcome in prelingually deafened adults.
Adunka et al	29	/	(1) preoperative residual hearing	(1) Substantial residual hearing before implantation does not provide a measurable performance advantage after CI.
			2009	
Williamson et al	28	/	(1) age at CI>70yr	(1) Patients older than 80 yrs obtain similar benefit, although auditory performance is less robust.
Litovsky et al	17	/	(1) bilateral CI	(1) Benefits with BICI when spatial cues are used to segregate speech and competing noise.
Lalwani et al	71	/	(1) GJB2-related deafness, (2) length of deafness, (3) age at CI, (4) length of device usage	(1) GJB2-related deafness and increased age at implantation are associated with poorer outcome.
Roditi et al	55		(1) duration of HL, (2) pre-HINT in quiet, (3) duration of HL either ear	(1) The regression analysis resulted in a model that accounted for 60% of the variance in postoperative CNC scores, including duration of HL in CI ear, pre-HINT in quiet, and duration of HL either ear.
			2010	
Friedland et al	78	/	(1) age at CI	(1) Older CI subjects achieve lower speech perception scores.
Stacey et al	11	/	(1) computerd-based self-administered training	(1) Significant improvements for consonant discrimination, but not on sentence tests or vowel discrimination.
Rotteveel et al	53	/	(1) otosclerosis	(1) Better performance relates to less severe signs of otosclerosis on CT scan, full insertion of the electrode array, little or no facial nerve stimulation and little or no need to switch off electrodes.

Lazard et al	16		(1) cognitive factors based on fMRI preoperative	(1) Good performers postoperative make use of a ‘normal’ dorsal route seen on fMRI preoperative, whereas the switch to an alternative lexico-semantic ‘ventral’ route and the recruitment of right supramarginal gyrus to compensate for deficient phonological processing suggests a poor outcome.
			2011	
Budenz et al	108	/	(1) age at CI	(1) Differences in speech outcomes between <70yr and >70yr depend on duration of deafness. (2) Better performance for older subjects if implanted at right side.
Park et al	161	/	(1) age at CI, (2) HA prior to CI	(1) Similar results for younger and older CI recipients. (2) No influence of prior HA use and its location in relation to the CI on speech perception improvement.
Lenarz et al	1005	(1) electrode design, (2) speech processing strategies	/	(1) Improvement in speech perception, especially in noise, with recent electrode designs and speech-processing strategies.
Buckley and Tobey	22	/	(1) cross-modal visual and auditory plasticity, (2) onset of HL	(1) Cross-modal plasticity influences perception performance. (2) Influence of cross-modal plasticity is higher for prelingually deafened adults.
			2012	
Moon et al	61	/	(1) etiology, (2) duration of deafness prior to CI	(1) Poorest speech perception if deafened by meningitis.
Lin et al	83	/	(1) age at implantation>60yr	(1) Older adult CI candidates who are younger at implantation and with higher pre-operative speech scores obtain highest speech performance.
			2013	
Holden et al	114	/	(1) biograph/audiologic information, (2) electrode position in the cochlea, (3) cognitive abilities	(1) Better performance if younger in age at CI, shorter duration of HL, lower CI sound-field threshold levels. (2) Electrode position and depth of insertion of the electrode array are inversely related to speech performance. (3) Better performance if electrode array is positioned closer to modiolar wall. (4) After controlling for age, cognition is no longer a factor affecting outcomes.

Blamey et al	2215	/	(1) duration of severe HL, (2) age at CI, (3) age at onset of severe HL, (4) etiology, (5) CI experience	Compared to the results of the study in 1996 (N=800), results revealed (1) less important negative effect of long duration of HL, (2) effect of age at CI and age at onset of HL were delayed until older ages, (3) effect of CI experience was greater with a steeper learning curve.

N= number, yr= year, CI= cochlear implant, fMRI= functional magnetic resonance imaging, HA= hearing aid, CT= computertomography, BICI= bilateral cochlear implants, HL= hearing loss, HINT= hearing-in-noise-test, EAS= electroacoustical stimulation, CNC= consonant-nucleus-consonant, C-level= comfort-level, DR= dynamic range, MMN= mismatch negativity, SAS= simultaneous analog stimulation, CIS= continuous interleaved sampler.

2.3 Defining success in speech perception outcome after cochlear implantation

The range of speech perception outcomes after CI is very wide: some CI recipients demonstrate substantial open-set speech perception in noisy conditions, whereas others may not consistently discriminate certain speech features presented in a closed-set.

A common yardstick for speech perception performance is the **performance of the 'average' patient.** Recently, Holden and colleagues (2013) obtained several monosyllabic speech perception scores of 114 postlingually deafened adults at numerous test intervals from 2 weeks until 2 years after the initial fitting. The final average speech perception score was 61.5% (median 65.4%, SD 20.7, range 2.9-89.3%). Based on percentile ranking of speech perception results (e.g. CNC words in Holden et al, 2013), outcome groups can be defined. Hence, these 114 CI recipients were addressed to one out of six outcome groups, with group 1 being the lowest and group 6 being the highest performers. Their top 10% of perfomers obtained on average 87.3% (median 87.4%, SD 1.4, range 85.3-89.3%), whereas average scores for their bottom 10% performers were 17.7% (median 22.2%, SD 10, range 2.9-28.8%).

Speech perception outcomes are also **depending on the test performed**. Dorman and Spahr (2006) defined above-average performers as those obtaining at least 80% correct on a CNC word recognition test in quiet. They revealed that four aspects of these CI recipients performance were notable, relative to that of recipients in the average group (having 58% correct on a CNC word recognition test). First, scores on a sentence test in quiet (CUNY, clearly articulated sentences) did not separate average and above-average recipients because of a ceiling effect. Second, performance on sentences produced in a casual speaking style in quiet was very high (90%), demonstrating that better patients can function at a very high level in a quiet environment even when multiple speakers are not trying to speak clearly and predictably. Third, at a SNR of +5 dB, performance of the above-average recipients fell to 52% correct (average patients obtained 27%), suggesting that even the better patients have difficulty understanding speech in a noisy environment. Finally, performance on a test of within-speaker gender recognition (70%) and melody recognition (56%) was, although superior to that of the average recipients (resp. 65% and 33%) poor. As a result of their findings, Dorman and Spahr (2006) defined the *statistically average postlingually-deafened* *patient* as follows: ‘This CI recipient can, in a quiet environment, understand carefully articulated, conversational speech with a very high degree of accuracy. In many common environments speech understanding remains relatively poor. In a quiet environment, only about three quarters of the words in sentences that are spoken casually by multiple speakers are recognized. Speech understanding is poor in any environment with background noise.’ They also defined the *statistically above-average patient*: ‘This patient enjoys near perfect speech understanding in quiet, even when sentences are spoken in a conversational style and when multiple speakers speak in turn. In modest amounts of noise, performance falls and is relatively poor at a SNR ratio found in busy work environments (Dorman and Spahr, 2006, p. 202).

Chapter 3

Research aims

Nevertheless selection criteria for candidacy vary per country, in general they have relaxed over the years and in general an improvement in speech perception performance after cochlear implantation is expected in the vast majority of patients. However, considerable inter-individual variability in speech perception performance remains incompletely understood. Moreover, because of expanding candidacy criteria, speech perception outcome after CI has become more difficult to predict since factors that may influence this performance have become increasingly more complex.

Within this thesis speech perception outcomes of a contemporary group of pediatric and adult CI users were obtained. Because research focusing at the extremes of speech perception performance is sparse, specific attention was given to recipients at the lower and upper part of the speech perception continuum. Furthermore, possible predictive variables that could administer a CI recipient's speech perception outcome, were investigated. Knowledge of variables influencing speech perception outcomes postimplantation will improve clinician's predictive accuracy and will enhance counseling. In addition, understanding and possibly manipulating the causes of variation might facilitate clinicians creating the best possible circumstances to optimize speech perception outcomes after implantation.

Consequently, five studies were conducted.

In **Chapter 4**, the impact of a newborn hearing screening program on the management and outcome of deaf children is evaluated and underlying factors responsible for the differences between high and low performing implanted children are identified by means of a retrospective cohort study performed on 391 implanted children.

The long-term outcome of implanted children deafened by congenital cytomegalovirus (cCMV) compared to a matched group of Connexin 26 (Cx26) children is discussed in **Chapter 5**. In addition, the possible influence of abnormal MRI results in cCMV-CI children is investigated.

Recently, researchers tend to focus on the electrode-neural bottleneck because insufficient independent channels might be one main cause responsible for the large individual variability. Therefore, a study was conducted to examine spatial spread of excitation (SOE) measurements performed in adult CI users. The first purpose of this study was to compare two Electrically evoked Compound Action Potentials (ECAP) based methods within a group of adult CI recipients. The second objective was to perform SOE measurements over repeated trials and hence to explore ECAP long-term stability. The third purpose was to gain insight in the relationship between ECAP spatial forward masking patterns and speech perception performance to determine how channel interaction relates to speech performance. Results of this study are presented in **Chapter 6**.

Chapter 7 is dedicated to a particular group of adult CI users, namely early-deafened, late-implanted adults. Commonly, their outcomes postimplantation are poor. In this study, a thorough test battery that permits evaluating a hierarchy of auditory skills ranging from sound detection through comprehension of sentences in noise is used to gain insight in speech perception postimplantation in 19 early-deafened, late-implanted adults. Moreover, errors in speech production are applied as a predictive factor for speech perception postimplantation.

While a CI is successful in delivering speech in quiet in the majority of adult CI users, its performance in delivering music is less than ideal. In **Chapter 8,** the associations between self-reported listening habits and perception of music and speech perception outcomes are assessed in quiet and noise for both unilateral adult CI users and bimodal (CI in one ear, HA in the opposite ear) adult users.

Research

Part I

Cochlear implantation in children

Chapter 4

Impact of newborn hearing screening: comparing outcomes in pediatric CI users

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De Raeve L
D'Haenens W
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Bockstael A
Keppler H
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Vinck BM
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Based on Laryngoscope
2009
119(5): 974-979

Abstract

Objectives

To evaluate the impact of a newborn hearing screening program on the management and outcome of deaf children and to identify underlying factors that may be responsible for the differences between high and low performing implanted children.

Methods

First, implanted children were sorted into two groups on account of screening age (early screened, N = 195 against late screened, N = 196). Both groups were compared with respect to several variables. Second, outcome of cochlear implantation was measured in terms of the child's speech perception and production skills (N = 355). A subgroup of high performing CI users was compared with low performing CI users with regard to several variables.

Results

Early screened children differ significantly from late screened children with respect to age of HL detection and age at cochlear implantation. Furthermore, early screening and implantation is associated with better auditory receptive skills and speech intelligibility. Additional impairments negatively influence both receptive and productive skills. In addition, children who communicate orally and wear bilateral CIs perform better on speech production, whereas a better speech perception was found in children who became progressively deaf as opposed to congenitally deaf children.

Conclusions

The results of this extensive study of profoundly deaf children with CIs in Flanders indicate that a newborn hearing screening program results in earlier intervention in deaf children which beneficially influences the auditory receptive skills and speech intelligibility.

4.1 Introduction

Since 1992, cochlear implantation is the standard treatment for profoundly deaf children in Flanders (Belgium). In 1998, the Flemish government implemented a Universal Newborn Hearing Screening (UNHS) program. By means of automated measurements of auditory brainstem evoked potentials (AABR, Algo® Portable Neonatal Hearing Screener (Natus Medical Inc., San Diego)), 97.91% of all newborns are offered hearing screening at the age of 4 weeks (Van Kerschaver et al, 2007). This screening test has a large sensitivity (99.95%) and specificity (99.98%), resulting in early detection of hearing impairment for the majority of HI children. Since the implementation of the UNHS program, referral for CI assessment is usually accomplished before the age of 6 months and, if appropriate, most children receive a CI by their first birthday. Before 1998, hearing screening in children was done by means of the Ewing behavioural test at the age of 9 months. The first aim of our study is to compare the population of children screened before 1998 (late screened children, LS) with children screened after 1998 (early screened children, ES) with regard to several variables.

Various studies have shown that CIs not only give deaf children access to sound, but may also provide beneficial effects on speech perception, speech production, language, and reading skills (Kishon-Rabin et al, 2002, Spencer et al, 2008). However, few studies have included large numbers of implanted children although this is advisable given the large inter-subject variability. Also, studies comparing results of high versus low performing implanted children are sparse. The second aim of our study is to compare outcome measures of a subgroup of high performing CI users with low performing CI users in an attempt to identify underlying factors that may be responsible for the variance of outcome in implanted children.

4.2 Methods and materials

4.2.1 Patients

This study included all profoundly deaf children (n = 391) under the age of 18 yrs who have been implanted in Flanders since 1992 and have received rehabilitation in one of the seven participating rehabilitation centers. The data were gathered under the guidance of the CORA-CI consortium, which encompasses these seven rehabilitation centers. All children had a bilateral HL of at least 85 dB HL before implantation and received their CI between 1992 and 2007. There was no case selection and, consequently, children were drawn from the full range of social, educational, and communication environments. Not only congenitally deaf children (73%), but also children with a progressive (17%) or sudden HL (10%) were included. The study was conducted in accordance with the ethical standard stipulated in the Helsinki declaration (2000) for research involving human subjects.

4.2.2 Early versus late screened children

Children were divided into two groups according to the screening method used. The late screened (LS) children (n = 196) were children screened before 1998, by means of the behavioral Ewing test. At the time of the study, their mean age was 13.7 yr (SD 2.5yr, range 9.4-18.0yr). The early screened (ES) group (n = 195) consists of children screened after 1998, by means of the Algo® test. Their mean age was 5.5 yr (SD 2.3yr, range 1.1-12.5yr) at the time of this study. Both groups were comparable with respect to device systems used. ES and LS groups were compared with respect to several variables. Variables concerning age were age at HL detection (in months) and age at implantation (in months). Age at HL detection was defined as the age at which HL was formally confirmed. Variables regarding etiology were cause of deafness (environmental, genetic or unknown), syndromal nature of deafness, congenital cytomegalovirus (cCMV), Connexin 26 (Cx26) and onset of deafness (congenital, progressive or sudden). Also the use of a device in the opposite ear (CI, HA or none) as well as variables with respect to additional impairments and disabilities were taken into consideration. For additional impairments (e.g. visual impairment) and disabilities (e.g. dyslexia) the World Health Organization definition was applied (WHO, 1980). Variables describing the family situation were hearing status of the parents (both hearing, one or both HI, unknown) and current communication mode of the parents (oral, total communication or sign language). Finally, variables concerning the educational setting at the time of the study were: current communication mode of the child (oral, total communication or sign language) and current school type (mainstream school, special school (mainly school for the deaf), part-time mainstream/special school or does not go to school).

4.2.3 High versus low performing children

Outcome of cochlear implantation was measured by means of the child's speech perception and production skills. These abilities were assessed with the Categories of Auditory Performance (CAP) and the Speech Intelligibility Rating (SIR) respectively. The CAP consists of a nonlinear hierarchical scale of auditory receptive (**Table 4.1**) (Archbold et al, 1995). The SIR, on the other hand, is a five-point hierarchical scale ranking a child's spontaneous speech (**Table 4.2**) (Allen et al, 1998). CAP and SIR reflect everyday performance of implanted children and can be applied to large groups. They are time-effective, can be used longitudinally and their inter-observer reliability has been formally confirmed (Archbold et al, 1998, Allen et al, 2001).

Table 4.1
Categories of Auditory Performance (CAP).

Category	CAP Description
7	Use of telephone with a familiar talker.
6	Understand conversation without lip reading with a familiar listener.
5	Understand common phrases without lip reading.
4	Discrimination of at least two speech sounds.
3	Recognition of environmental sounds.
2	Response to speech sounds.
1	Awareness of environmental sounds.
0	No awareness of environmental sounds.

Table 4.2
Speech Intelligibility Rating (SIR).

Category	SIR Description
5	Connected speech is intelligible to all listeners; the child is understood in easily everyday contexts.
4	Connected speech is intelligible to a listener who has little experience of a deaf person's speech.
3	Connected speech is intelligible to a listener who concentrates and lip reads.
2	Connected speech is unintelligible; intelligible speech is developing in single words when context and lip reading cues are available.
1	Connected speech is unintelligible; pre-recognizable words in spoken language, primary mode of communication may be manual.

CAP and SIR scores of 355 children were obtained at the time of the study. First, CAP and SIR scores were compared across age groups. Second, CAP as well as SIR scores were used to categorize children into high and low performing groups. Children were sorted into performance groups on the basis of their auditory receptive abilities (CAP): children with a CAP score below level 4 were assigned to the low performing CAP group (n = 64), children achieving the highest CAP level were categorized as being in the high performing CAP group (n = 60). Secondly, children were sorted into performance groups based on their speech intelligibility (SIR): children who achieved a SIR score below level 3 were counted as being low SIR performers (n = 92), children who achieved the highest SIR level were counted as being high SIR performers (n = 125).

Following variables were compared between high and low performers: additional impairments (none, 1, 2, 3 or 4), communication mode of the child at the time of the study (oral, total communication or sign language), type of device in the opposite ear (CI, HA or none) and onset of deafness (congenital, progressive or sudden).

4.2.4 Data analysis

Data were analyzed using SPSS version 15 (Chicago, IL). For the comparison of early versus late screened children, a one-way analysis of variance (ANOVA) was used for continuous variables and a Pearson χ^2 analysis for categorical variables. Furthermore, Mann-Whitney-U tests were performed to compare current CAP and SIR scores with respect to the age groups. For comparison of high versus low performing children, a Pearson χ^2 analysis was performed to determine the impact of 4 independent categorical factors on 2 dependent categorical outcome measures. Where appropriate, Bonferroni adjustments were made to control for type I errors. For all analyses the level of significance was set at 0.05.

4.3 Results

4.3.1 Comparison of early versus late screened children

Significant differences between age of HL detection ($p = .000$) and age of implantation ($p = .000$) could be established between both groups (**Table 4.3**). In early screened children (ES) HL was detected at a mean age of 5 months as opposed to late screened children (LS), whose HL was detected at a mean age of 14 months (**Figure 4.1**). Furthermore, mean age of implantation for ES children was 22 months as opposed to 71 months for LS children, implying that LS children were implanted on average 49 months later than ES children (**Figure 4.2**). Both figures show outliers, indicating a relatively high age at detection and/or implantation for some of the ES and LS children. These were cases of progressive or sudden childhood HL instead of congenital hearing losses.

Table 4.3
Results of statistical analysis by means of one-way ANOVA or Pearson χ^2 test.

		One-way ANOVA	**Pearson χ^2 test**	**P-value**
Age				
	Detection	$F(1,310) = 38.3$		**.000**
	Implantation	$F(1,381) = 207.4$		**.000**
Etiology				
	Cause of deafness		$\chi^2(2) = 9.21$	**.010**
	Syndromal		$\chi^2(1) = 0.47$	.493
	cCMV		$\chi^2(1) = 2.80$	.094
	Cx26		$\chi^2(1) = 11.76$	**.001**
	Onset		$\chi^2(3) = 1.61$	.658
Device opposite ear			$\chi^2(2) = 3.77$	.152
Additional				
	Impairment(s)		$\chi^2(4) = 3.68$	.461
	Disability(ies)		$\chi^2(3) = 27.55$	**.000**
Family				
	Hearing status parents		$\chi^2(3) = 4.443$	.197
	Current communication mode parents		$\chi^2(2) = 5.25$	.072
Education				
	Current communication mode child		$\chi^2(2) = .537$	.799
	Current school type		$\chi^2(3) = 16.78$	**.001**

Values given in bold indicate significant difference between early and late screened children based on one-way analysis of variance (ANOVA) or Pearson χ^2 test ($p<.05$). cCMV =congenital cytomegalovirus; Cx26 = connexin 26.

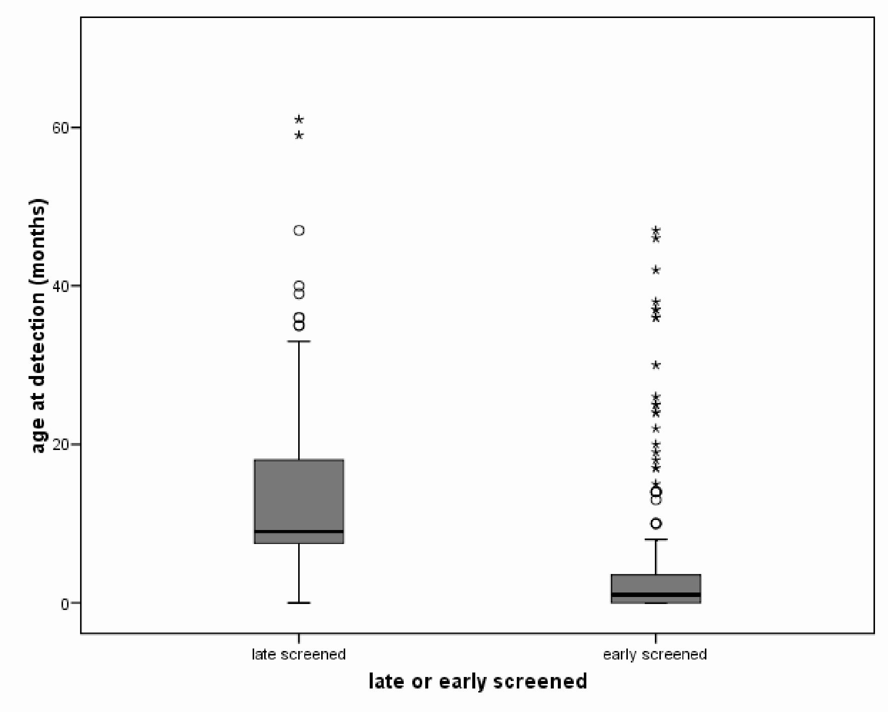

Figure 4.1 Box plot of the age at detection (in months) for early and late screened children. Boxes represent the median (thick horizontal line), lower and upper quartiles (ends of boxes), minimum and maximum values (ends of whiskers), outliers (values between 1,5 and 3 box lengths from either end of the box - circles) and extreme values (values more than 3 box lengths from either end of the box - asterisks).

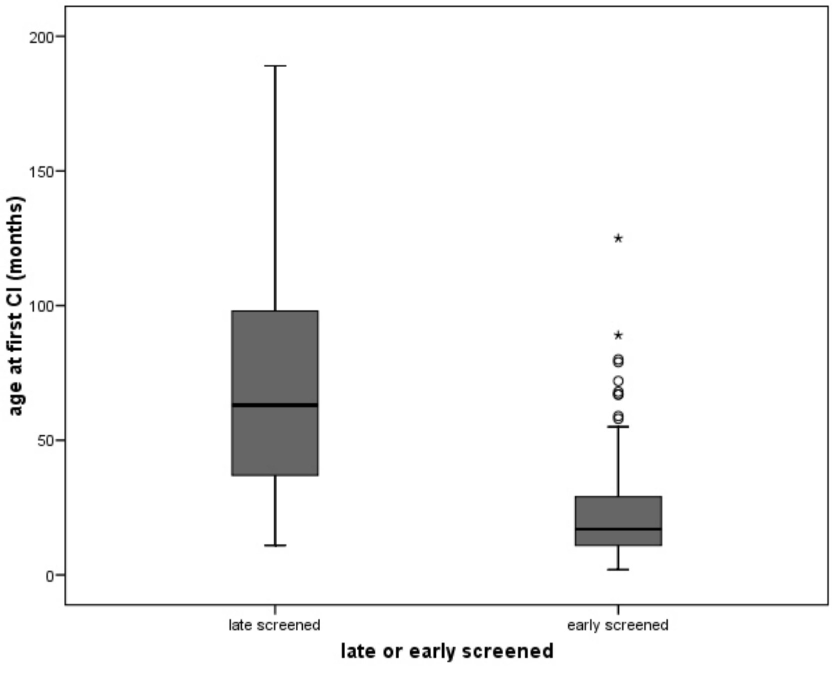

Figure 4.2 Box plot of the age at cochlear implant (CI) (in months) for early and late screened children. Boxes represent the median (thick horizontal line), lower and upper quartiles (ends of boxes), minimum and maximum values (ends of whiskers) outliers (values between 1,5 and 3 box lengths from either end of the box - circles) and extreme values (values more than 3 box lengths from either end of the box - asterisks).

Concerning the etiology of deafness, significant differences between the ES and LS groups were found (p = .010). Significant differences between the number of cases with a genetic as compared to an unknown etiology were established by means of post hoc Bonferroni analysis (p = .003). In ES children the proportion of unknown etiologies (41%) was smaller than in LS children (58%), indicating that the proportion of children with an unknown etiology has decreased in recent years. This difference is attributable in large part to Cx26 diagnostics. Nowadays, more children are diagnosed with a Cx26 mutation (19% in ES against 7% in LS). With regard to the number of children diagnosed with syndromic HL (e.g. Waardenburg syndrome) (p = .493), cCMV (p = .094) and the onset of deafness (p = .658), no significant differences could be found between both groups. Approximately the same amount of children was identified in each group with a syndromic HL (7% in ES against 5% in LS), with cCMV (11% in ES against 6% in LS) or with a congenital HL (72% in ES against 74% in LS). In relation to additional impairments, no significant difference was found between early and late screened children (38% ES against 35% LS with at least one additional impairment) (p = .461), whereas for additional disabilities fewer ES children (14%) were diagnosed with one or more additional disability(ies) compared to LS children (35%) (p = .000). Parents in both groups did not differ significantly with respect to hearing status (p = .197) nor communication mode (p = .072). The majority of parents are not HI (95% in ES and 97% in LS) and communicate orally with their HI child (51% in ES and 59% in LS). Finally, the ES and LS groups were compared with regard to educational setting at the time of the study and significant differences were found (p = .001). As to the early screened children, 24% attended mainstream school, 70% went to a school for the deaf (5% was too young to attend school) as opposed to the late screened children, of whom 37% attended mainstream school and 60% went to a school for the deaf. There were no significant differences between both groups concerning the communication mode used by the child (p = .799).

4.3.2 Comparison of high versus low performing children

Figures 4.3 and **4.4** show the current CAP and SIR scores for all children divided into age groups. These figures indicate that children between 0 and 12 years achieve higher CAP and SIR scores by increasing age, respectively. In contrast, children between 12 and 14 years obtain a significantly lower CAP (p = .014) and SIR (p = .006) compared to children between

10 and 12 years. Children older than 12 years at the time of the study are late screened children.

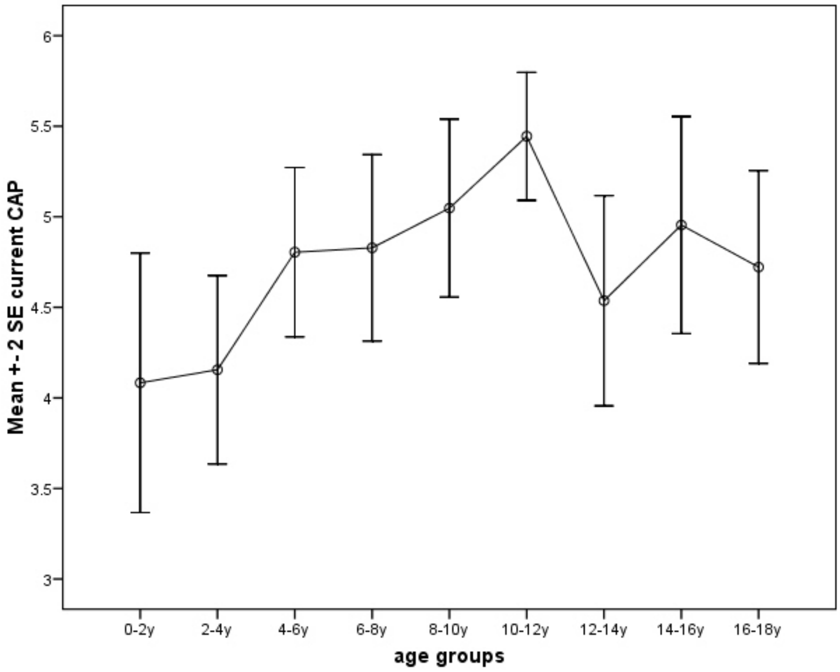

Figure 4.3 Error bar chart for current Categories of Auditory Performance (CAP) score and the current age of the children (by means of age groups: 0-2y, 2-4y, 4-6y, 6-8y, 8-10y, 10-12y, 12-14y, 14-16y and 16-18y). These graphs represent the mean score (circles) and standard error of the mean (SE, multiplier 2, ends of bars).

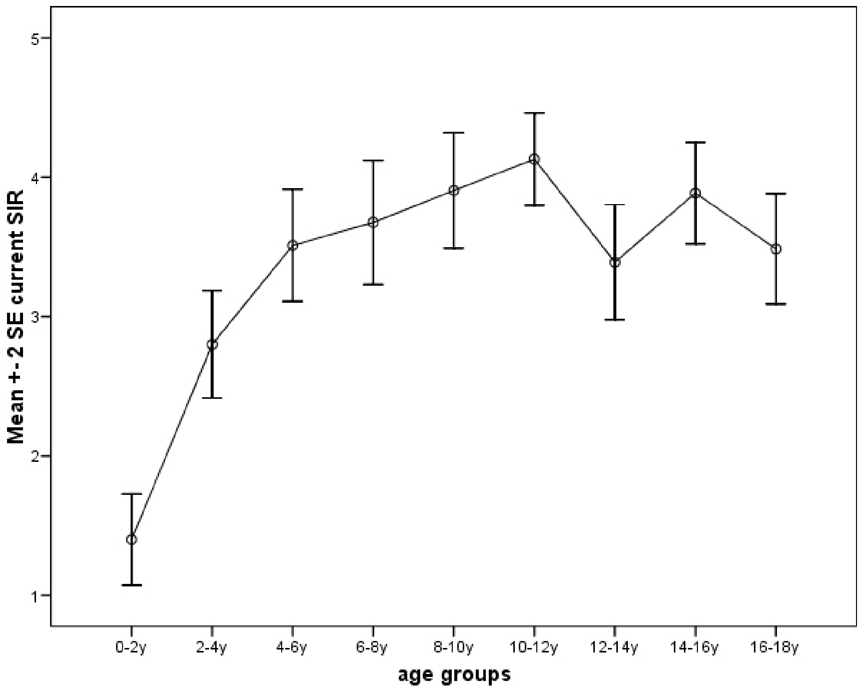

Figure 4.4 Error bar chart for current Speech Intelligibility Rating (SIR) score and the current age of the children (by means of age groups: 0-2y, 2-4y, 4-6y, 6-8y, 8-10y, 10-12y, 12-14y, 14-16y and 16-18y). These graphs represent the mean score (circles) and standard error of the mean (SE, multiplier 2, ends of bars).

Second, children were divided into high and low performing groups. There were no significant age differences between high and low performing groups. Four categorical factors were treated as explanatory variables for levels of auditory receptive skills (CAP) and speech intelligibility (SIR) (**Table 4.4**). Children without any additional impairment performed significantly better on CAP (p = .002) and SIR (p = .000). If current communication mode of the child was oral, speech intelligibility was significantly better compared to children who communicated through total communication or sign language (p = .000), but differences in auditory receptive skills did not reach significance levels (p = .096). Bilaterally implanted children scored significantly better on speech intelligibility than unilaterally implanted children, regardless of whether they used a HA at the opposite ear or not (p = .000). Again, no significant differences in auditory receptive skills could be established between the latter subgroups (p = .398). Finally, children who became deaf because of a progressive etiology, demonstrated significantly better auditory receptive skills than children who were born profoundly deaf (p = .016), but no significant speech intelligibility differences could be found between both subgroups (p = .549).

Table 4.4

Univariate analysis Pearson χ^2 test.

	Additional impairments	**Current comm. mode. child**	**Device opposite ear**	**Onset of deafness**
CAP	$\chi^2(4) = 16.47$ p = **.002**	$\chi^2(2) = 4.67$ p = .096	$\chi^2(2) = 2.06$ p = .398	$\chi^2(3) = 9.73$ p = **.016**
SIR	$\chi^2(4) = 60.84$ p = **.000**	$\chi^2(2) = 27.43$ p = **.000**	$\chi^2(2) = 24.68$ p = **.000**	$\chi^2(3) = 2.14$ p = .549

Values given in bold indicate significant difference between high and low performing children based on Pearson χ^2 test (p < .05). Current comm. mode child = current communication mode of the child.

4.4 Discussion

4.4.1 Comparison of early versus late screened children

This study compared 196 children of the pre-AABR era (LS children) with 195 children of the Algo® era (ES children). As expected, the age of HL detection loss was significantly lower in the group of children were the neonatal hearing screening program was implemented. On average, hearing impairment nowadays is identified within the first 5 months of life. When including only the congenitally deaf children, the mean age of HL detection diminished from 5 months to 3 months for the ES children and from 14 months to 12 months for the LS children. The former results are within the range of the stated goals for UNHS by the Joint Committee of Infant Hearing and American Academics of Pediatrics (2000). This study also shows that early detection of HL results in earlier implantation. ES children were implanted at the age of 22 months on average (19 months for the congenitally deaf children), whereas for LS children age at implantation was 70 months on average (67 months for the congenitally deaf children). Thus, UNHS has a major impact on early detection and intervention of hearing impairment in children.

Variables regarding the etiology of deafness revealed that the number of unknown etiologies diminishes, probably because diagnostic techniques are evolving. This is in accordance with the results of Declau and colleagues who studied the etiologic factors in 170 referred neonates who failed UNHS (Declau et al, 2008).

The result concerning additional impairments revealed no significant differences, although additional disabilities were diagnosed in a higher number of LS children. Two conceivable explanations are the following. First, as a result of early screening, children enter in a multidisciplinary rehabilitation setting in an early stage of their life. As a result of this early multidisciplinary intervention, additional disabilities (e.g. dyslexia) probably are early identified and treated. Second, as the group of ES children is younger, it is possible that additional disabilities will still develop. Therefore, all implanted children should be followed up longitudinally to gain insight in their future cognitive and emotional development.

Currently, less ES children attend mainstream schools compared to LS children. It should be born in mind that the ES children are on average 7 years younger than the LS children which may explain the fact that the majority of ES children still attend the schools for the deaf. These results are in accordance with the results of Verhaert and colleagues (2008), indicating that almost 60% of the young implanted children attend special education until primary school.

4.4.2 Comparison of high versus low performing children

CIs do not always provide recipients with major benefits. The second aim of our study was to examine factors that might contribute to their speech perception and production performances. Our results indicated that children who were screened early and, consequently, received early intervention and implantation, reached better CAP and SIR scores. Results are in accordance with results of several other authors who noticed an improvement in receptive and expressive skills as a result of earlier implantation (O'Neill et al, 2002, Svirsky et al, 2004).

Having one or more additional impairments negatively influences both receptive and productive skills. This finding supports those of previous investigators (Nikolopoulos et al, 2006). Despite poor outcomes, children with special needs may achieve some benefits from cochlear implantation, resulting from a better access to the environment provided by the CI. Possibly, specific tests and parental questionnaires are suited better for evaluations of overall postimplant benefits in daily life (Berrettini et al, 2008). Because a number of additional disabilities and impairments may be identifiable only after implantation, the need for ongoing tests and follow-up care by a multi-professional team is emphasized.

A third factor taken into account was the type of device worn in the opposite ear. Currently, about 20% of the LS children and 29% of the ES children are bilaterally implanted. Some studies have reported advantages in speech recognition of bimodal (CI in one ear and HA in opposite ear) and bilateral (BICI, two CIs) fittings, compared to single CIs. Therefore, specific tests are required to investigate binaural advantages, such as binaural redundancy, head shadow effects and binaural squelch. The CAP scale used in this study, is not recommended as a measure for binaural advantages. In the future, tests should be developed, aimed specifically at measuring binaural advantages in young children. In SIR scores, a significant difference was found, indicating that children fitted with bilateral implants performed better with respect to speech intelligibility than bimodally or unilaterally fitted children, which is in accordance with the study of Boons and colleagues (2012). However, it should be noted that the majority of the bilaterally implanted children received their implant because the participated in a national study that funded the second implant, or because their parents had sufficient financial resources to pay the second implant. Possibly, these bilaterally implanted children achieved better speech production scores compared to unilaterally implanted children irrespective of their unilateral/bilateral CI status. Chang and co-workers (2010) showed that fewer children from families with a low socio-economic status (SES) are bilaterally implanted compared to children for families with a high SES. This possible confounding factor can be taken into account in future studies, because bilateral CI is reimbursed for children (< 12 years) in Belgium since 2010. Hence, also children from families with fewer financial resources can benefit form bilateral CI.

Further, the impact of the current communication mode of the child on CAP and SIR was examined. Our results revealed no statistical significant difference with respect to the receptive skills between oral and total communication users. This is in contrast with findings of other researchers. Iler Kirk and colleagues (2000) found a significant influence on receptive performance skills of communication mode in favor of children who used oral communication. It should be noted that speech recognition tests used in this study not always reflect ability levels in daily life. The CAP scale, on the other hand, is designed to measure real-life auditory receptive skills. Our SIR results, i.e. outcomes for productive abilities, were in accordance with the results of several researchers (Tobey et al, 2004, Uziel et al, 2007). These studies confirmed that mean speech intelligibility score was higher for children who were enrolled in an oral program than for those who were enrolled in a total communication program.

Finally, the onset of deafness was taken into account. Few researchers have investigated outcome differences between children with congenital, progressive or sudden hearing losses. Nikolopoulos et al (2006) investigated long-term speech perception abilities by means of the CAP scale within comparable groups of congenitally deaf children and children who became suddenly deaf caused by meningitis. Both groups showed improvement over time, suggesting that the onset of deafness has little influence on receptive outcome, provided that children are implanted early. Because of the small number of deaf children with meningitis causing deafness in our study, it was not possible to assess outcomes for this particular group. However, with respect to children with a congenital and a progressive HL, a difference in speech perception skills was found in favor of children with a progressive HL.

4.5 Conclusions

In conclusion, this report on 391 children fitted with CIs of which 196 were late screened and 195 were early screened, confirms that early detection of hearing impairment due to the implementation of UNHS program results in early intervention and implantation of deaf children which has a major positive impact on both auditory receptive skills and speech intelligibility.

Chapter 5

Outcome after cochlear implantation in children deafened by congenital cytomegalovirus matched with children deafened by Connexin 26 peers

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Abstract

Objectives

To compare the long-term speech perception and production outcome after cochlear implantation (CI) in children deafened by congenital cytomegalovirus (cCMV) with a matched group of Cx26-CI children by controlling for chronological age and magnetic resonance imaging (MRI) findings.

Methods

Retrospective review of 12 cCMV-CI children and matched Cx26-CI children for speech perception and speech production.

Results

Two trends were seen in our data. First, cCMV children with normal MRI scans perform equally or even slightly better on speech perception tests compared to their Cx26 peers during the first three years. The majority of cCMV children with normal MRI scans (5 out of 7), suffered from a delayed-onset SNHL. Their mean age at first implantation (2yr9m, range 15m-82m) was higher compared to their matched Cx26 peers (9m, range 7m-12m). Before being implanted, the majority of these delayed-onset hearing impaired children had benefitted from a certain period of normal hearing (with or without amplification of a hearing aid). Possibly, this input might have led to an advantage the first three years after CI. Second, results between cCMV-CI children with and cCMV-CI children without MRI abnormalities and their matched Cx26-CI counterparts tentatively suggest that, over a 5-yr follow-up period, cCMV-CI children with abnormal MRI scans catch up for speech perception, but lag behind for speech production.

Conclusion

cCMV children with normal MRI scans perform equally or even slightly better on speech perception tests compared to their Cx26 peers during the first three years, whereas results between cCMV-CI children with and cCMV-CI children without MRI abnormalities and their matched Cx26-CI counterparts tentatively suggest that, over a 5-yr follow-up period, cCMV-CI children with abnormal MRI scans catch up for speech perception, but lag behind for

speech production. In future, the inclusion of MRI results may assist in improved counseling of parents with cCMV deafened children seeking CI.

5.1 Introduction

cCMV is the leading cause of non-genetic congenital sensorineural hearing loss (SNHL) in early childhood and an important cause of neuro-developmental delay. In Flanders (Belgium), 0.53% of all live born infants are affected (Foulon et al, 2008). Approximately 10 % of cCMV children is symptomatic at birth. Their outcome is usually poor: there is substantial mortality (10-20%) and most survivors suffer from severe neurologic sequelae (55%) (Dobbins et al, 1992). Seven to twenty-one percent of clinically asymptomatic patients develop late sequelae including SNHL, neurologic deficits, and behavioral problems (Dahle et al, 2000, Foulon et al, 2008). In addition, cCMV related SNHL can be uni- or bilateral, mild to profound, late-onset, fluctuating or progressive (Williamson et al, 1992, Fowler et al, 1997, Dahle et al, 2000, Fowler and Boppana, 2006, Grosse et al, 2008).

In children with bilateral severe to profound HL, uni- or bilateral cochlear implantation (CI) is the treatment of choice. However, studies determining speech perception and production outcomes in cCMV-CI children are scarce. Seven studies compared the outcomes of cCMV-CI children with a group of non-cCMV-deafened implanted children (Ramirez-Inscoe, 2004, Yoshida et al, 2009, Ciorba et al, 2009, Iwasaki et al, 2009, Malik et al, 2011, Matsui et al, 2012, Yamazaki et al, 2012). Ciorba et al (2009), Matsui et al (2012), and Yamazaki et al (2012) matched their cCMV-CI children to a group of Connexin-26 (Cx26) CI children, but they did not control for chronological age nor age at implantation. Ramirez-Inscoe et al (2004), Malik et al(2012) and Iwasaki et al (2009) controlled their mean group data for age at implantation, however, etiology of deafness within their control group was not specified. Overall, improved performance after CI was reported, nevertheless, on the long-term, outcome after CI in cCMV-children was quite variable. None of the abovementioned studies controlled for neurological deficits as a possible confounding variable, although previous studies have shown that cCMV infants are at risk for additional neurological deficits (Boppana et al, 1992, Meinzen-Derr et al, 2009, Birman et al, 2012).

Resulting from the shortcomings in the literature, the objective of the current study is to compare the long-term speech perception and production outcomes of cCMV-CI children with a matched group of Cx26-CI children by controlling for chronological age and magnetic resonance imaging (MRI) findings. We purposefully selected an etiology that is known to have a low comorbidity with additional disabilities. Therefore, Cx26-CI children serve as matched control group. It is widely known that Cx26-CI children have good spoken language outcomes postimplantation, possibly because the pathologic changes do not affect the spiral ganglion cells which are the neural elements stimulated by the CI (Sinnathuray et al, 2004). Therefore, we hypothesized that Cx26-CI children would obtain better results on speech perception and production scores compared to their cCMV-CI matched peers.

5.2 Methods

5.2.1 Children deafened by congenital cytomegalovirus

Twelve children (6 boys, 6 girls) deafened by cCMV were included in the current study. The diagnosis of cCMV was made by polymerase chain reaction (PCR)-based assay using dried blood spots taken within the first week after birth (Guthrie card, N=8) or by CMV isolation in neonatal urine (N=4). Within our study, asymptomatic infants are those without apparent clinical manifestations at birth, such as petechiae, jaundice, hepatosplenomegaly or microcephaly, whereas symptomatic children exhibit one or more of these clinical manifestations. One child (cCMV-3) had clinical features at birth (microcephaly, dysmaturation), the others were asymptomatic. In addition, neuroimaging by means of MRI showed abnormalities in 4 additional children (cCMV-4, cCMV-5, cCMV-8, cCMV-11) (**Table 5.1**). In addition, some cCMV children suffered from additional disabilities determined at their last test moment (**Table 5.2**).

Table 5.1
MRI abnormalities.

Subject	MRI abnormalities
cCMV-3	polymicrogyria
cCMV-4	periventricular leucomalacia
cCMV-5	white matter lesions bifrontal
cCMV-8	white matter hyperintensities
cCMV-11	polymicrogyria, periventricular leucomalacia

cCMV=congenital cytomegalovirus, MRI=magnetic resonance imaging

Table 5.2
Additional disabilities.

Subject	Additional disabilities
cCMV-2	hypotonia
cCMV-4	severly motot impairment, severly cognitive impairment
cCMV-5	delayed motor skills
cCMV-6	cognitive impairment, autism spectrum disorder
cCMV-11	motor impairment, cognitive impairment
cCMV-12	delayed motor skills

cCMV=congenital cytomegalovirus

Three children (cCMV-8, cCMV-11 and cCMV-12) received intravenous ganciclovir for 6 weeks during the first months of their life.

Five children (cCMV-1, cCMV-4, cCMV-8, cCMV-11, cCMV-12) were diagnosed as having a bilateral profound SNHL at the universal newborn hearing screening (ALGO®) which was performed before the age of 4 weeks. The symptomatic child (cCMV-3) was also diagnosed as having a bilateral profound SNHL during his stay at the NICU. The remaining six children had a delayed-onset HL with an average age at detection of HL at 2y1m (range 10m-5y3m). In all children diagnosis of bilateral profound HL was confirmed by means of Auditory Brainstem Responses (ABR, thresholds > 90dB nHL), absent Oto Acoustic Emissions (OAEs) and age-appropriate behavioral audiometry.

Approximately 8 months (range 4m-14m) after the diagnosis of bilateral profound SNHL, all 12 cCMV children were implanted (Nucleus™). Six children received a unilateral CI, the remaining were bilaterally implanted of whom one was simultaneously implanted (cCMV-2) (**Table 5.3**).

5.2.2 Children deafened by Connexin 26

A carefully matched control group was drawn from the Ghent University Hospital CI Center database. This control group consists of twelve Cx26-CI children, matched for chronological age ($t(23) = .395$, $p > .05$) and cochlear implantation (unilaterally or bilaterally implanted). Hence twelve pairs were included in the study.

All Cx26 children were diagnosed as bilaterally hearing impaired by ALGO ® newborn hearing screening and confirmed by ABR testing (thresholds > 90dB nHL), absent OAEs and age-appropriate behavioral audiometry. Cochlear implantation was performed on average 9 months (range 6-38 m) after the diagnosis of bilateral profound SNHL. Six children were unilaterally implanted, the remaining six received sequential bilateral CIs (**Table 5.3**). These children were implanted with a Nucleus™ device, except for subject Cx-10, who received a HiRes90K™ (Advance Bionics Corporation®). None of these matched Cx26 children had abnormal MRI results, nor apparent additional disabilities at their last test moment.

Table 5.3
Patient CI characteristics.

Subject	Gender	Delayed onset SNHL	CI	Age CI left (yr,mo)	Type CI left	Age CI right (yr,mo)	Type CI right	Chronological age (yr,m)
cCMV-1	F	N	BI	1yr2mo	CI512	10mo	CI512	3yr8m
cCMV-2	M	Y	BI	1yr6mo	CI512	1yr6mo	CI512	3yr11m
cCMV-3	M	N	UNI	10mo	24RECA	/	/	4yr10m
cCMV-4	F	N	BI	1yr5mo	24RECA	10mo	24RECA	4yr11m
cCMV-5	M	Y	UNI	/	/	3yr2mo	CI512	4yr11m
cCMV-6	F	Y	BI	1yr3mo	24RECA	6yr0mo	24RECA	6yr8m
cCMV-7	F	Y	BI	2yr11mo	24RECA	4yr5mo	CI512	6yr9m
cCMV-8	M	N	UNI	2yr3mo	24RECA	/	/	7yr11m
cCMV-9	M	Y	BI	6yr10mo	CI512	7yr7mo	24RECA	8yr5m
cCMV-10	F	Y	UNI	1yr4mo	24RECA	/	/	9yr0m
cCMV-11	F	N	UNI	/	/	1yr0mo	24RCA	8yr9m
cCMV-12	M	N	UNI	7mo	24RECA	/	/	1yr4m
Cx-1	F	N	BI	10mo	CI512	1yr3mo	CI512	3yr7m
Cx-2	F	N	BI	1yr5mo	CI512	7mo	CI24RECA	3yr9m
Cx-3	M	N	UNI	10mo	CI24RECA	1yr5mo	CI24RECA	2yr0m
Cx-4	F	N	BI	1yr0mo	CI512	9mo	CI512	4yr0m
Cx-5	M	N	UNI	/	/	3yr2mo	24RECA	3yr7m
Cx-6	F	N	BI	4yr0mo	CI512	9mo	24RECA	6yr8m
Cx-7	M	N	BI	1yr7mo	24RECA	7mo	24RECA	6yr7m
Cx-8	M	N	UNI	/	/	7mo	24RECA	6yr11m
Cx-9	F	N	BI	9mo	24RCA	6yr9mo	CI512	9yr0m
Cx-10	M	N	UNI	/	/	1yr0mo	HiRes90K	8yr10m
Cx-11	M	N	UNI	9yr8mo	24RECA	1yr 0mo	24RCA	9yr10m
Cx-12	F	N	UNI	10mo	24RECA	/	/	1yr3m

CMV= cytomegalovirus, Cx=Connexin 26, F=female, M=male, SNHL= sensorineural hearing loss, Y=yes, N=no, BI= bilaterally implanted, UNI= unilaterally implanted, yr=year, mo=months.

5.2.3 Speech perception and production

Outcomes after CI were measured by means of the child's speech perception and production skills. Because of the wide range in patient age and speech development, these abilities were assessed with the Categories of Auditory Performance (CAP) (**Table 5.4**) and the Speech Intelligibility Rating (SIR) respectively (**Table 5.5**). The CAP consists of a nonlinear eight-point hierarchical scale of auditory receptive abilities (Archbold et al, 1995). The SIR is a five-point hierarchical scale ranking a child's spontaneous speech (Allen et al, 1998). CAP and SIR reflect everyday performance of implanted children. They are time-effective, can be used longitudinally and their inter-observer reliability has been formally confirmed (Archbold et al, 1998, Allen et al, 2001).

Table 5.4
Categories of Auditory Performance (CAP).

Category	CAP Description
7	Use of telephone with a familiar talker.
6	Understand conversation without lip reading with a familiar listener.
5	Understand common phrases without lip reading.
4	Discrimination of at least two speech sounds.
3	Recognition of environmental sounds.
2	Response to speech sounds.
1	Awareness of environmental sounds.
0	No awareness of environmental sounds.

Table 5.5
Speech Intelligibility Rating (SIR).

Category	SIR Description
5	Connected speech is intelligible to all listeners; the child is understood in easily everyday contexts.
4	Connected speech is intelligible to a listener who has little experience of a deaf person's speech.
3	Connected speech is intelligible to a listener who concentrates and lip reads.
2	Connected speech is unintelligible; intelligible speech is developing in single words when context and lip reading cues are available.
1	Connected speech is unintelligible; pre-recognizable words in spoken language, primary mode of communication may be manual.

5.2.4 Longitudinal follow-up

At the last test moment, mean average chronological age for the cCMV-CI children was 5y11m (range 1y4m-9y0m) and 5y6m (1y3m-9y10m) for the Cx26-CI children. The cCMV-CI children had on average 3y11m (9m-7y9m) of experience since their first CI and the Cx26-CI children had on average 4y6m (5m-8y10m) of CI experience. Since a wide range in chronological age between pairs was seen, not all pairs were followed-up over a 5-yr period (**Table 5.6**).

Table 5.6
Follow-up period per pair.

Pair	Follow-up period (yr)
pair 1	2yr
pair 2	2yr
pair 3	1yr
pair 4	2yr
pair 5	1yr
pair 6	5yr
pair 7	4yr
pair 8	5yr
pair 9	3yr
pair 10	5yr
pair 11	5yr
pair 12	1yr

yr=year

5.2.5 Statistical analyses

Statistical analyses were performed using SPSS version 19.0. Wilcoxon signed-rank tests were used to gain insight in yearly CAP and SIR scores for Cx26-CI and cCMV-CI pairs. A second series of Wilcoxon signed-rank tests was applied to yearly CAP and SIR scores with normal MRI scores for both cCMV-CI and Cx26-CI children (7 pairs included) and abnormal MRI scores for the cCMV-CI and normal MRI scores for their matched Cx26-CI peers (5 pairs included). Bonferroni corrections were applied to adjust for multiple tests. Level of significance was set at .05.

Testing was approved by the UZ Ghent Medical Ethical Committee and was in accordance with the Declaration of Helsinki (2000).

5.3 Results

Based on Wilcoxon signed-rank test, and after applying for Bonferroni corrections, no statistical significant differences were found between cCMV-CI and matched Cx26-CI pairs for speech perception nor speech production (**Table 5.7**).

Table 5.7
Wilcoxon signed-rank test.
All cCMV-CI and Cx26-CI pairs included (12 pairs).

Speech perception					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=3.00	Mdn=5.00	Mdn=5.50	Mdn=5.00	Mdn=5.50
Cx26	Mdn=4.00	Mdn=5.00	Mdn=5.00	Mdn=5.00	Mdn=5.50
p-value	p>.05	p>.05	p>.05	p>.05	p>.05
Speech production					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=2.00	Mdn=3.00	Mdn=4.50	Mdn=4.00	Mdn=4.00
Cx26	Mdn=4.00	Mdn=3.00	Mdn=4.00	Mdn=4.00	Mdn=5.00
p-value	p>.05	p>.05	p>.05	p>.05	p>.05

cCMV= congenital cytomegalovirus, Cx26= Connexin 26, MRI= magnetic resonance imaging, CI= cochlear implant, yr= year, Mdn= median

Figures 5.1 and **5.2** show the differences in yearly CAP and SIR scores for the seven pairs having normal MRIs. A positive result implies that, averaged over the included pairs, Cx26-CI children obtain better results on speech perception and speech production compared to their matched cCMV-CI peers and vice versa. The Wilcoxon signed-rank test could not reveal statistical significant differences between these groups for speech perception nor production (**Table 5.8**). However, results of the first 3 years tentatively suggest that cCMV-CI children with normal MRI results, perform better on speech perception scores compared to their Cx26-CI peers.

Table 5.8
Wilcoxon signed-rank test.
cCMV-CI and Cx26-CI children with normal MRI results (7 pairs).

Speech perception					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=4.00	Mdn=5.50	Mdn=6.00	Mdn=6.00	Mdn=6.00
Cx26	Mdn=4.00	Mdn=5.00	Mdn=5.00	Mdn=5.00	Mdn=6.00
p-value	p>.05	p>.05	p>.05	p>.05	p>.05
Speech production					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=2.00	Mdn=4.00	Mdn=5.00	Mdn=5.00	Mdn=5.00
Cx26	Mdn=2.00	Mdn=3.00	Mdn=4.00	Mdn=5.00	Mdn=5.00
p-value	p>.05	p>.05	p>.05	p>.05	p>.05

cCMV= congenital cytomegalovirus, Cx26= Connexin 26, MRI= magnetic resonance imaging, CI= cochlear implant, yr= year, Mdn= median

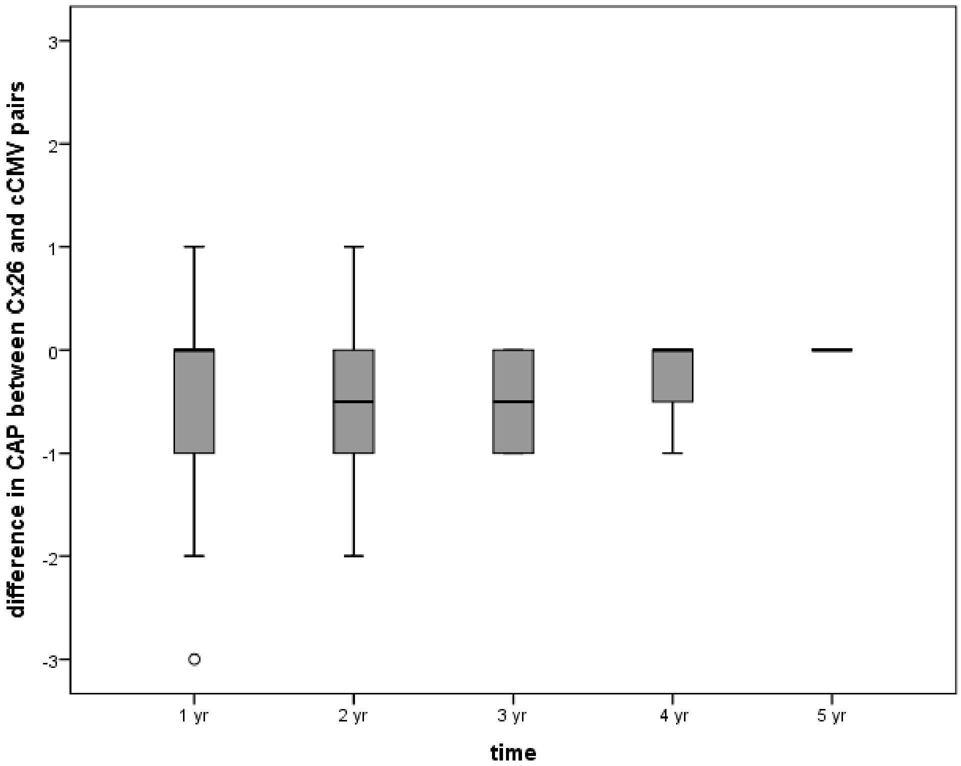

Figure 5.1 shows box plots of differences in speech perception results (CAP) over a 5-yr period for cCMV and matched Cx26 children with normal MRI results.
CAP= Categories of Auditory Performance, yr= year.

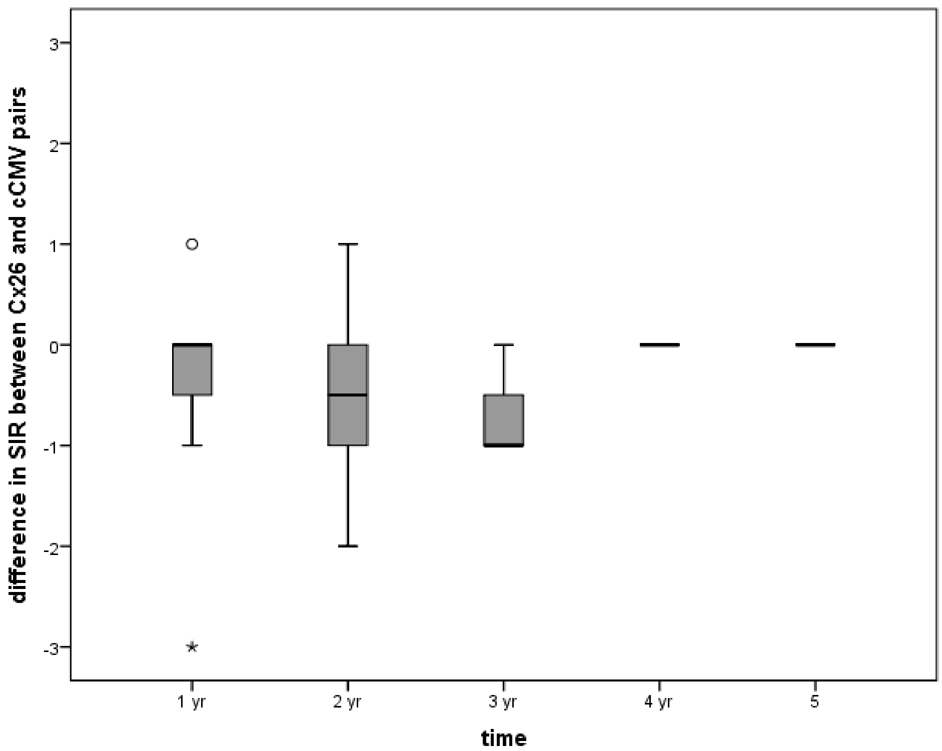

Figure 5.2 shows box plots of differences in speech production results (SIR) over a 5-yr period for cCMV and matched Cx26 children with normal MRI results.
SIR= Speech Intelligibility Rating, yr= year.

Figures 5.3 and **5.4** show the differences in yearly CAP and SIR scores for the five pairs of whom the cCMV-CI children had abnormal MRI results. For speech perception, cCMV-CI children with abnormal MRI results tend to catch up with their Cx26-CI matched peers after a 5-yr period. Although not statistically significant (Wilcoxon signed-rank test, **Table 5.9**) differences in speech production tend to arise by increasing chronological age, with the Cx26-CI children obtaining better speech production scores.

Table 5.9
Wilcoxon signed-rank test.
cCMV-CI with normal and Cx26-CI children with abnormal MRI results (5 pairs).

Speech perception					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=2.00	Mdn=4.00	Mdn=4.00	Mdn=4.50	Mdn=4.50
Cx26	Mdn=4.00	Mdn=4.00	Mdn=4.50	Mdn=5.00	Mdn=5.00
p-value	p>.05	p>.05	p=	p>.05	p>.05
Speech production					
	1yr	2yr	3yr	4yr	5yr
cCMV	Mdn=1.00	Mdn=2.00	Mdn=2.00	Mdn=2.00	Mdn=2.00
Cx26	Mdn=2.00	Mdn=3.00	Mdn=3.50	Mdn=4.00	Mdn=4.50
p-value	p>.05	p>.05	p>.05	p>.05	p>.05

cCMV= congenital cytomegalovirus, Cx26= Connexin 26, MRI= magnetic resonance imaging,
CI= cochlear implant, yr= year, Mdn= median

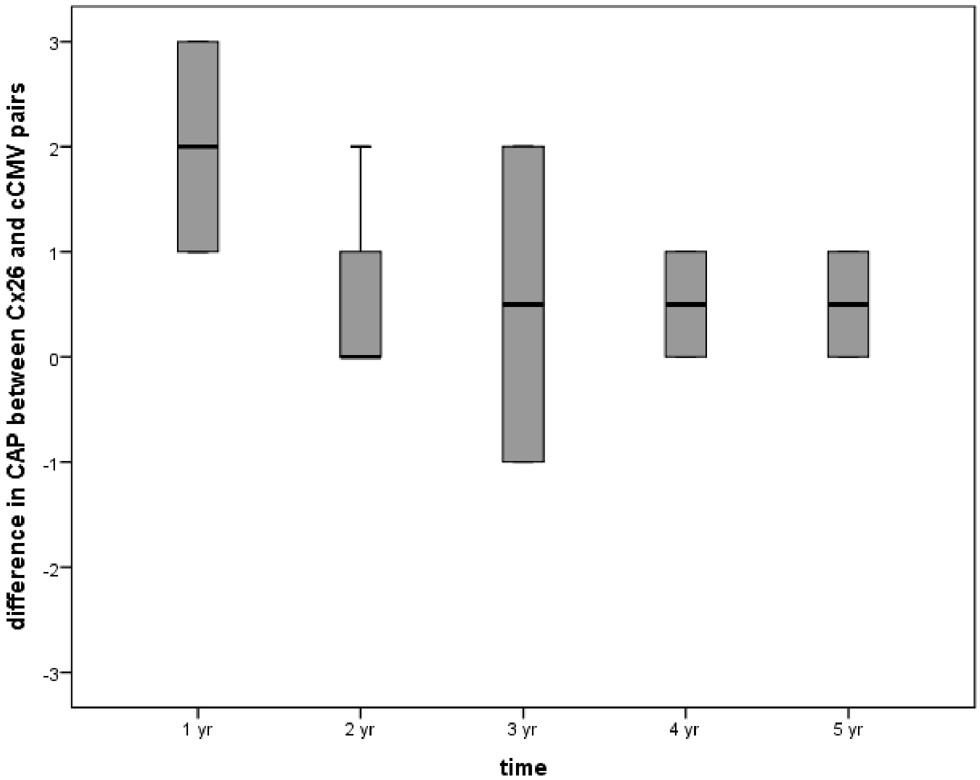

Figure 5.3 shows box plots of differences in speech perception results (CAP) over a 5-yr period for cCMV children with abnormal MRI results and matched Cx26 children with normal MRI results.
CAP= Categories of Auditory Performance, yr= year.

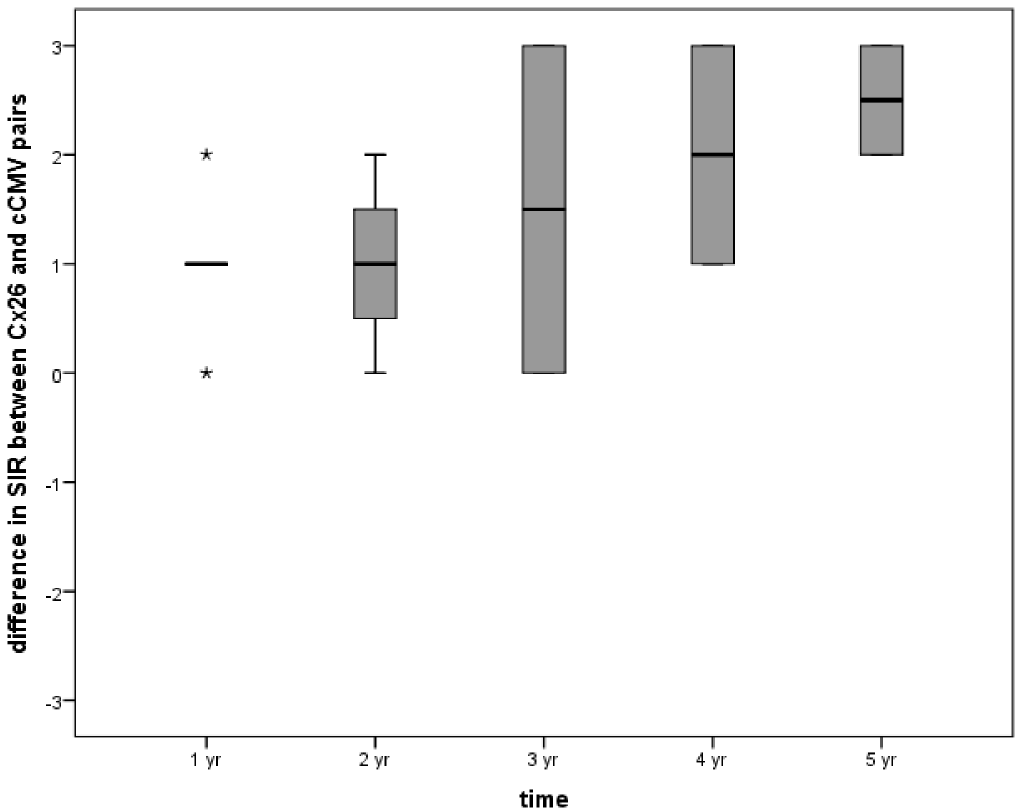

Figure 5.4 shows box plots of differences in speech production results (SIR) over a 5-yr period for cCMV children with abnormal MRI results and matched Cx26 children with normal MRI results.
SIR= Speech Intelligbility Rating, yr= year.

5.4 Discussion

This study investigated speech perception and production data of 12 cCMV-CI children, matched with 12 Cx26-CI children, over a 5-yr period. On average, no statistically significant differences were found between both groups with respect to CAP and SIR.

Other researchers have also gained insight in speech perception and production outcomes of cCMV-CI children. Matsui and colleagues (2012) compared speech and language skills of 5 cCMV-CI and 7 Cx26-CI children over a 4-yr period. Results between both groups were equivalent, although not confirmed by statistical analysis. Outcomes of 4 cCMV-CI children were examined in the study of Yoshida et al (2009) and comparisons were made with a non-cCMV group (N=17). The mean speech production scores within the first year after CI showed a statistically significant difference, implying lower scores for the cCMV-CI group. Beyond the first year, no difference was established. Viccaro and colleagues (2012) evaluated retrospectively language development of 6 cCMV-CI children 10 years postoperatively. They showed an improvement in language perception and production, although, by comparing chronological and language age, all children had a language delay. However, no comparisons were made with a matched control group. Other studies reporting outcome after implantation did not include long-term follow-up data (Malik et al, 2011) or included a small number of children (Iwasaki et al, 2009).

Although not statistically significant, two trends were seen in our data. First, cCMV-CI children with normal MRI scans perform equally or even slightly better on speech perception tests compared to their Cx26-CI peers during the first three years. The majority of cCMV-CI children with normal MRI scans (5 out of 7), suffered from a delayed-onset SNHL. Their mean age at first implantation (2yr9m, range 15m-82m) was higher compared to their matched Cx26-CI peers (9m, range 7m-12m). Before being implanted, the majority of these delayed-onset hearing impaired children had benefitted from a certain period of acoustic hearing (with or without amplification of a hearing aid). Possibly, this input might have led to an advantage the first three years after CI. Second, results between cCMV-CI children with and cCMV-CI children without MRI abnormalities and their matched Cx26-CI counterparts tentatively suggest that, over a 5-yr follow-up period, cCMV-CI children with abnormal MRI scans catch up for speech perception, but lag behind for speech production. Research has shown that infants with cCMV are more likely to have coexisting abnormalities on neuroimaging compared to non-cCMV-infants (Steinlin et al, 1996, Weiss et al, 2012). The impact of MRI abnormalities on CI outcome has never been verified before. Ciorba and colleagues (2009) compared 16 cCMV children, of whom 14 were implanted, with 8 Cx26-CI children, matched for age and preimplant linguistic category. Long-term results of language development over a 3-yr period showed that both groups improved, however, progression was slower in the cCMV-CI group. Since their cCMV-CI group included 10 symptomatic and 4 asymptomatic children, results were attributed to concomitant cognitive impairment, but no details were given upon MRI status. Another study reported results of 6 cCMV-CI infants 10 years postimplantation, including 4 children with MRI calcifications who demonstrated a language delay (Viccaro et al, 2012). However, this possible confounding factor was not taken into account in the statistical analysis. Malik and colleagues (2011) showed that asymptomatic cCMV-CI children (N=14) with learning difficulties obtained statistically significantly lower speech perception and production scores two years after implantation compared to a non-cCMV-CI control group (N=45) matched for sex and age at CI. Children with significant learning disabilities only developed environmental awareness and limited understanding of language. By contrast, children with no or mild learning disabilities achieved good speech perception and production scores. None of these asymptomatic cCMV-CI children had abnormal MRI results. In our study, the majority of included children was too young to investigate possible learning difficulties. In the future, it might be interesting to succeed this follow-up study to explore possible learning difficulties in our matched cCMV-Cx26-CI group, especially for cCMV-CI children having normal MRI results.

Because a wide range in speech perception and speech production abilities was found in our CI recipients, two rating scales (CAP and SIR) were implemented, in accordance with other researchers examining speech perception and production outcomes in CI recipients with additional needs (Amirsalari et al, 2012, Birman et al, 2012). One of the disadvantages of the usage of a rating scale is that these instruments can suffer from ceiling effects. Second, obtained results give an indication of speech perception and production outcome, but detailed information is not included.

To date, there is a lack of objective, reliable and validated tools with regard to assessing the long-term progression of CI children with additional disabilities, such as (a subgroup of) cCMV-CI children. Research has shown that the number of implanted children with additional disabilities has grown up to 45% (Meinzen-Derr, 2009, Birman et al, 2012). Hence, there is an emerging need to develop such instruments at the earliest possible time (Vlastarakos et al, 2009).

5.5 Conclusions

This study showed that on average, cCMV-CI children progress equally over a 5-yr period after CI compared to matched Cx26-CI children on CAP and SIR. Former studies comparing implanted cCMV and non-cCMV children reported mixed results, possibly because these studies hardly controlled for confounding variables such as abnormal MRI results (Shin et al, 2011). Our study tentatively suggests that over a 5-yr follow-up period results of cCMV-CI children with abnormal MRI findings and their matched Cx26-CI counterparts show that these cCMV-CI children can catch up for speech perception, but lag behind for speech production, whereas cCMV-CI children with normal MRI results achieve comparable, or even slightly better, speech perception and production results as their Cx26-CI peers. Although further research is needed, the inclusion of MRI results may assist in improved counseling of parents with cCMV deafened children seeking CI.

Part II

Cochlear implantation in adults

Chapter 6

Spatial spread of excitation measurements within adult cochlear implant users: feasibility, long-term stability and correlation with speech performance

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Abstract

Objectives

The first purpose was to compare two Electrically evoked Compound Action Potential (ECAP) based methods (Spread Of Excitation, Variable Recording (SOE-VR) and Spread of Excitation Variable Masker (SOE-VM)) within a group of adult CI recipients. The second objective was to perform SOE-VM patterns over repeated trials and hence to explore ECAP long-term stability. The third purpose was to gain insight in the relationship between ECAP spatial forward masking patterns and speech perception to determine how channel interaction relates to speech performance.

Methods

Data were collected from 24 postlingually deafened unilaterally implanted adults.

Results

Experiment 1 revealed that SOE-VR measurements were statistically significantly broader compared to SOE-VM measurements. Within experiment 2, no significant differences could be found in width variations over repeated trials, implying a good long-term stability. Furthermore, results revealed no statistical significant results which could correlate ECAP derived spatial profiles with three speech perception tests.

Conclusions

Normalized SOE-VR functions were broader than normalized SOE-VM functions. Furthermore, long-term stability of SOE-VM patterns for three probe electrode locations over repeated trials showed that SOE-VM measures were highly stable across repeated trials over a one-yr time period. This suggests that physiological spatial forward masking measurements can be re-used when conducting studies that investigate possible relationships between these types of ECAP measures and psychophysical measurements. Furthermore, no significant correlation was found between ECAP derived spatial profiles and speech perception nor relative information transmitted. Moreover, the presence of peak displacements could not account for poorer speech perception results or lower relative information transmitted.

6.1 Introduction

The majority of patients with profound to severe SNHL have improved listening capabilities after cochlear implantation and average performance with CIs has increased during the last decades through improvements in patient indications, technology and sound processing techniques. In spite of the success, speech perception, especially on complex listening tasks, remains limited and a large inter-individual variability is seen (Skinner et al, 2002). Only a small portion of this variability can be accounted for by factors such as etiology and duration of deafness (Gfeller et al, 2008; Blamey et al, 2013). Recently, researchers tend to focus on the electrode-neural bottleneck because insufficient independent channels might be one main cause responsible for the large individual variability (Shannon et al, 2002). In CI recipients, insight in the electrode-neural interface can be gained by performing psychophysical and physiological measurements. Since psychophysical measures, such as forward masking and gap detection, rely on perceptual judgments, results are influenced by central processes and therefore do not directly reflect peripheral spread of neural excitation (Cohen et al, 2003). Furthermore, retrieving psychophysical data requires the cooperation of the CI recipient and is time-consuming. Therefore, psychophysical methods will not be used in this study.

Several methods have been developed to assess spatial spread profiles physiologically by means of the electrical evoked compound action potential (ECAP) through the implant telemetry system by presenting masker and probe stimuli to electrodes along the intra-cochlear array (Cohen et al, 2003; Abbas et al, 2004; Eisen and Franck, 2005; Hughes and Abbas, 2006b; Busby et al, 2008; Hughes and Stille, 2008, 2010). Within this study, two methods to retrieve ECAP profiles will be used. The first method (termed 'Spread Of Excitation -Variable Recording' (SOE-VR)) makes use of a stimulation on a fixed electrode contact and measures the evoked response on all other contacts along the array (Frijns et al 2002, Cohen et al 2004) (**Figure 6.1**). In the second method (termed 'SOE-Variable Masker' (SOE-VM)) probe and recording electrodes are fixed and the masker electrode is variable (**Figure 6.2**). Within this second measurement, the resulting ECAP amplitude pattern indicates the amount of spatial overlap between neural populations stimulated by masker and probe electrodes. It is hypothesized that recipients with smaller spatial profiles achieve better speech perception scores, however, results from earlier studies comparing spatial forward masking profiles and speech perception have been mixed. Hughes and Abbas (2006b) examined the relationship between ECAP channel interaction measures and the ability to discriminate pitch between electrodes in a psychophysical pitch-ranking task. Furthermore, speech perception performance (phonemes, words and sentences in quiet) was compared to pitch-ranking ability and width of the ECAP measurements. No significant correlation between speech perception results and either pitch-ranking or width of the ECAP measurements was found. Hughes and Stille (2008) investigated the relationship between both psychophysical (PFM) and physiological (ECAP FM) spatial forward masking patterns and speech perception. Their results showed no significant correlation between speech perception (phonemes and words in quiet, sentences in noise) and PFM or ECAP FM. Nevertheless, CI recipients whose ECAP FM correlated well with PFM generally had the poorest speech perception whereas recipients with the poorest correlation had the best speech perception performance. Recently, van der Beek et al (2012) used linear mixed models to investigate a possible relationship between SOE-VM and speech. They used the width of SOE-VM scores to predict monosyllabic speech perception scores in quiet, but no significant predictive effect was found.

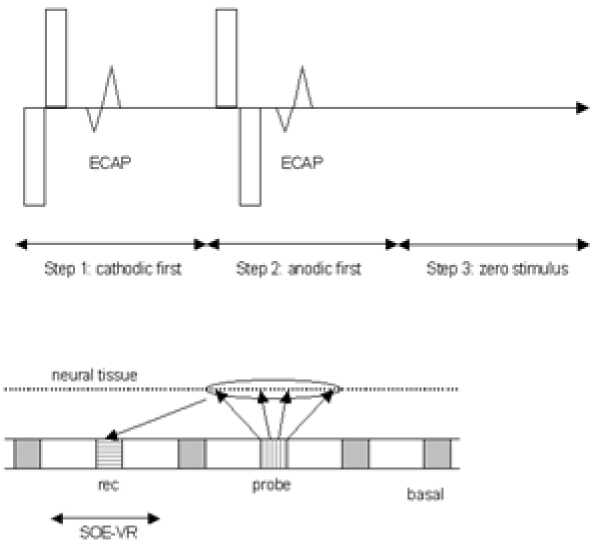

Figure 6.1 Schematic representation illustrating Spread Of Excitation Variable Recording
ECAP=Electrically Evoked Compound Action Potential, SOE-VR=Spread Of Excitation, Variable Recording, rec=recording electrode

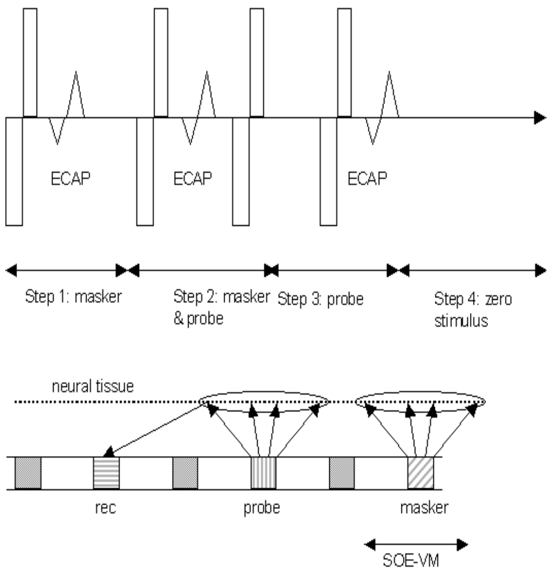

Figure 6.2 Schematic representation illustrating Spread Of Excitation Variable Masker ECAP=Electrically Evoked Compound Action Potential, SOE-VM=Spread Of Excitation, Variable Masker, rec=recording electrode

In contrast, some researchers did find a correlation between spatial measurements and speech perception performance. Throckmorton and Collins (1999) explored the relationship between psychophysical forward masking, electrode discrimination and speech perception tests. They found that forward masking and electrode discrimination were strongly correlated with measures requiring consonant and phoneme recognition, respectively. Furthermore, Boëx et al (2003) found a significant negative correlation between psychophysical forward masking and percentage correct on a medial consonant test. Kim et al (2011) revealed that the slope of the ECAP growth function was significantly correlated with performance in quiet and noise.

However, the fundamental understanding of neural channel interaction in particular and its relationship to speech performance in general remains unclear. Earlier studies exploring this possible relationship never included a variety of speech perception material that makes use of both lower and higher-level auditory mechanisms. Mostly, speech perception in quiet, measured at one intensity level was used. This study addresses this lack in various speech tests and the possible correlation with ECAP derived spatial profiles.

The first purpose of this study was to compare two ECAP based methods (SOE-VR and SOE-VM) within a group of adult CI recipients. Based on previously performed studies, it is hypothesized that SOE-VR measurements have broader excitation profiles compared to SOE-VM profiles (Abbas et al (2004), Cohen et al (2004), Hughes and Stille (2010) and van der Beek et al (2012)). The second objective was to perform SOE-VM patterns over repeated trials and hence to explore ECAP long-term stability. It is hypothesized that SOE-VM patterns are stable over a 1-year period. The third purpose was to gain insight in the relationship between ECAP spatial forward masking patterns and speech perception performance to determine how channel interaction relates to speech performance. It is hypothesized that recipients with less channel interaction would obtain better results on speech perception tests because of improved spectral resolution and vice versa.

6.2 Methods

6.2.1 Subjects

Data were collected from 24 postlingually deafened adults who were unilaterally implanted at the Ghent University Hospital. Eleven subjects were implanted with a HiRes90K CI system and 3 were implanted with CII CI system (Advanced Bionics Corporation, Valencia, CA). Eight subjects received a 24RE(CA) CI and the remaining 2 received a CI512 (Cochlear Ltd., Lane Cove, Australia). Impedance measures were obtained for all subjects before data collection to identify potential electrode integrity issues. All subjects were native Dutch speakers and had at least 3 months of experience with their CI. Details on participant characteristics are listed in **Table 6.1**. The study was approved by the Institutional Review Board of the Ghent University Hospital and was conducted in accordance with the ethical standard stipulated in the Helsinki Declaration (2008) for research involving human subjects.

Table 6.1
Details on participants characteristics.

subject	gender	internal device and electrode array	external device	ear	duration of deafness (yr)	age at CI (yr)	etiology	exp1	exp2	exp3
AB1	F	HiRes90K HiFocus Helix	Harmony	R	1.0	76	un, progr			x
AB2	F	HiRes90K HiFocus Helix	Harmony	L	5.0	77	un, progr			x
AB3	F	CII	Harmony	L	2.0	58	un, progr	x	x	x
AB4	F	CII	Harmony	L	0.8	38	meningitis	x	x	x
AB5	M	HiRes90K HiFocus Helix	Auria	R	2.0	62	un, progr	x	x	x
AB6	F	HiRes90K HiFocus Helix	Harmony	R	3.0	69	Menière, progr	x	x	x
AB7	M	HiRes90K HiFocus Helix	Harmony	R	1.0	60	un, sudden	x	x	x
AB8	F	HiRes90K HiFocus Helix	Harmony	L	1.0	61	un, progr			x
AB9	F	HiRes90K HiFocus Helix	Harmony	R	2.0	34	Usher, progr	x		x
AB10	M	HiRes90K HiFocus Helix	Auria	L	3.0	55	un, progr	x	x	x
AB11	F	HiRes90K HiFocus Helix	Auria	R	10.0	52	un, progr	x	x	
AB12	M	CII	Auria	L	10.0	72	Menière, progr	x		
AB13	M	HiRes90K HiFocus Helix	Harmony	R	10.0	37	un, progr	x		
AB14	F	HiRes90K HiFocus Helix	Auria	R	5.0	41	un, progr	x		
CO1	F	Nucleus CI24RE(CA)	FDM	R	3.0	70	un, otosclerosis		x	x
CO2	F	Nucleus CI24RE(CA)	FDM	R	5.0	69	un, progr		x	x
CO3	M	Nucleus CI24RE(CA)	FDM	L	3.0	72	un, sudden		x	x
CO4	F	Nucleus CI24RE(CA)	FDM	R	3.0	69	un, progr		x	x
CO5	M	Nucleus CI24RE(CA)	FDM	R	3.0	64	un, progr		x	x
CO6	F	Nucleus CI24RE(CA)	FDM	R	1.0	58	un, progr		x	x
CO7	F	Nucleus CI512	CP810	R	12.0	65	un, progr			x
CO8	M	Nucleus CI24RE(CA)	FDM	R	0.4	44	un, sudden			x
CO9	F	Nucleus CI512	CP810	R	10.0	70	un, progr			x

AB= Advanced Bioncics, CO=Cochlear, M=male, F=female, FDM=Freedom, L=left, R=right, yr=years, CI=Cochlear Implant, exp=experiment, progr=progressive, un=unknown

6.2.2 Equipment setup

For subjects with the Advanced Bionics devices, ECAPs were obtained using the RSPOM (Research Studies Platform – Objective Measures module) v.1.3.0. software (Advanced Bionics, Niel, Belgium). For subjects with the Nucleus Freedom device, ECAPs were obtained using the Nucleus Custom Sound EP software v. 3.1 (Cochlear Ltd, Lane Cove, Australia). All subjects were tested with a laboratory speech processor. For Advanced Bionics subjects, a Platinum Speech Processor was used, for Nucleus subjects a Freedom speech processor was used.

6.2.3 Stimulation and recording parameters

In this paper two methods to retrieve ECAP profiles will be used. The first method (SOE-VR) makes use of stimulation on a fixed electrode contact and measures the evoked response on all other contacts along the array (Frijns et al 2002, Cohen et al 2004). To minimize the influence of the stimulus artifact on the measured ECAP waveform, alternating polarity is used as artifact reduction technique.

In the second method (SOE-VM) probe and recording electrodes are fixed and the masker electrode is variable. The resulting ECAP amplitude pattern indicates the amount of spatial overlap between neural populations stimulated by masker and probe electrodes. To minimize the influence of the stimulus artifact on the measured ECAP waveform, the masker-probe or forward-masking method is applied within both Advanced Bionics and Cochlear recipients. Default recording electrode setting is two electrode positions apical to the stimulating electrode (Abbas et al, 1999).

Both SOE-VR and SOE-VM measurements were performed at three probe electrode (el) stimulation locations: one apical, one mid and one basal. For Advanced Bionic electrodes, stimulation electrodes 3 (apical), 7 (mid) and 11 (basal) were used. For Cochlear electrodes, stimulation electrodes 17 (apical), 11 (mid) and 5 (basal) were used. Each measurement was performed at patient’s most comfortable level (MCL). The MCL was determined using a 10-point loudness scale by increasing the ECAP stimulus in a stimulation-only condition, until it was deemed loud, but just below the onset of discomfort. For each subject and at each of the three probe electrodes, MCL was determined. For the SOE-VM patterns, the same current level was used on the probe and masker electrodes.

6.2.4 Data analysis on spatial profiles

A typical ECAP consists of a biphasic waveform with a leading negative trough (N1) at about 0.2-0.4 ms, followed by a much smaller positive peak (P2) or plateau, occurring at about 0.6-0.8 ms (Abbas et al 1999). The amplitude of the ECAP is defined as the absolute difference (in µV) between N1 and P2. For both Cochlear and Advanced Bionics, the default software peak detector (resp. Custom Sound EP v. 3.1 and RSPOM v. 1.3) was initially used for data collection, and the N1 and P1 locations were then verified by visual inspection by an experienced audiologist.

The excitation patterns were normalized by dividing the amplitude of the evoked potential measured at each electrode by the peak amplitude of each respective excitation pattern. For each measurement, spatial profiles were modeled using a linear regression fit and width values were calculated at 50% of the peak of each fitted curve.

6.2.5 Speech perception measurements

Speech perception tests were performed in a sound booth, with speech and noise materials presented at 0° azimuth at 1 meter distance from the CI recipients. Tests were performed with the recipients own speech processor at their daily program. Three speech perception tests were collected in the aided condition (CI alone), varying in their demand on central function ranging from lower to more higher-level auditory and linguistic mechanisms. The first speech perception task was a closed-set identification of consonants (C) in an intervocalic /a/ context (12 /aCa/ nonsense syllables) with C being /m, n, p, b, k, s, z, f, v, t, d and ɣ/. The stimuli were presented at 65 dB SPL. Each /aCa/ nonsense syllable was presented 6 times in random order. Participants responded to the stimuli by clicking one of the 12 /aCa/ nonsense syllables on a computer screen. Tests were self-paced, they did not proceed before an answer was given. No training or feedback was provided, except for familiarization with the different response alternatives before testing.

The second perception task was an open-set performance by using Dutch recorded CVC (Consonant-Vowel-Consonant) 3 phoneme monosyllabic words (NVA, Wouters et al, 1994).

Hence, the demand on central function was increased, compared to the closed-set identification of consonants. Former studies never included speech testing with measures that approximate everyday listening situations, since stimuli were presented at only one intensity level (mostly 70 dB SPL). Unfortunately, stimuli presented at 70 dB SPL are not representative of real-life listening situations (Firszt et al, 2004). Therefore in this study, speech stimuli will be presented at three intensity levels (70, 55 and 40 dB SPL). For each condition a list of 11 words was used and scoring was based on percent-correct performance at phoneme level.

Because good spectral resolution may lead to high results on a speech-in-noise perception task, this task was included within the third speech perception test. SRT was measured adaptively in noise (SRT_n) by means of the LIST sentences, representing speech in daily life (van Wieringen and Wouters, 2008). The speech level was varied and the speech-weighted noise available for the LIST was kept constant at 60 dB SPL. The SRT is the sound level of the speech signal at which 50% of the presented speech sentences is correctly identified. The SRT was determined using a simple up-down (1 up, 1 down) procedure with a 2-dB step size. The first sentence was repeated with increasing level until it was correctly identified by the subject. The level of subsequent sentences was presented once at a 2 dB lower or higher level than the previous sentence, depending on whether the latter was identified correctly or not. A response was judged correct if and only if all key words of the sentence were repeated correctly. The adaptive procedure continued until 10 reversals occurred, and then the speech levels of sentences 6 to 10 and the imaginary 11th sentence were averaged to obtain the SRT value.

6.2.6 Feature analyses for the consonants

The relative information transmitted (ITr) was calculated for five features for each of the subjects (Miller and Nicely, 1955). ITr is expressed as a value between 0 and 1 and is the ratio of the transmitted information calculated from the confusion matrix to the maximal possible information transferred by the stimuli and categories under test. The consonants were classified according to the features ‘voicing, place of articulation, affrication, manner of articulation and nasality’. The classification of the consonants is shown in **Table 6.2**.

Table 6.2
Classification of the consonant features.

	p	t	k	b	d	m	n	v	z	s	f	ɣ
voicing	0	0	0	1	1	1	1	1	1	0	0	1
place	0	1	2	0	1	0	1	3	1	1	3	2
affrication	0	0	0	0	0	0	0	1	1	1	1	0
manner	0	0	0	0	0	1	1	2	2	2	2	0
nasality	0	0	0	0	0	1	1	0	0	0	0	0

6.3 Results

6.3.1 Experiment 1: SOE-VR versus SOE-VM patters

The goal of experiment 1 was to determine whether ECAP SOE-VR profiles were significantly different from SOE-VM profiles. Eight HiRes90K and three CII recipients were included (**Table 6.1**). Comparisons were made for a basal (el11), mid (el7) and apical (el3) electrode. **Figure 6.3** shows average normalized SOE-VM and SOE-VR functions for the three probe electrode locations. First, note the considerable standard deviations in both SOE-VR and SOE-VM profiles, at all electrodes, implying between-subject differences. Second, SOE-VR profiles were more bell-shaped, compared to SOE-VM profiles. These latter profiles were more exponential-like, especially at the apical and mid probe electrode. Furthermore, large within-subject variability was seen. **Figure 6.4** shows SOE-VR and SOE-VM profiles (basal, mid and apical) for recipient AB7. The SOE-VR profile at the apical and mid probe electrodes is asymmetrical and relatively broad, while the SOE-VR profile at the basal electrode is less asymmetric and narrower.

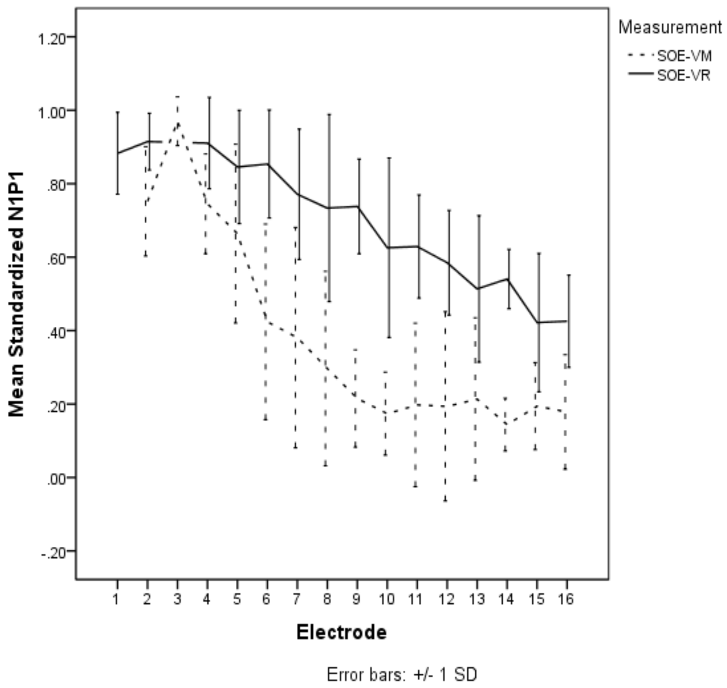

Figure 6.3a Mean (+- 1SD) normalized ECAP N1P1 amplitudes across Advanced Bionics recipients for SOE-VR (full line) and SOE-VM (dotted line) functions for *apical* (el3) probe locations.
ECAP=Electrically Evoked Compound Action Potential, SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

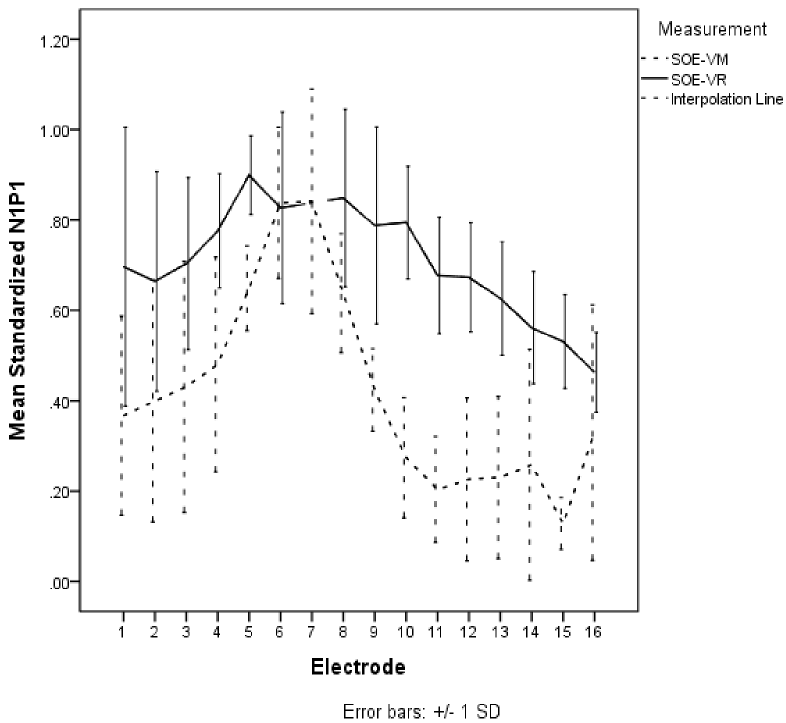

Figure 6.3b Mean (+- 1SD) normalized ECAP N1P1 amplitudes across Advanced Bionics recipients for SOE-VR (full line) and SOE-VM (dotted line) functions for *middle* (el7) probe locations.
ECAP=Electrically Evoked Compound Action Potential, SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

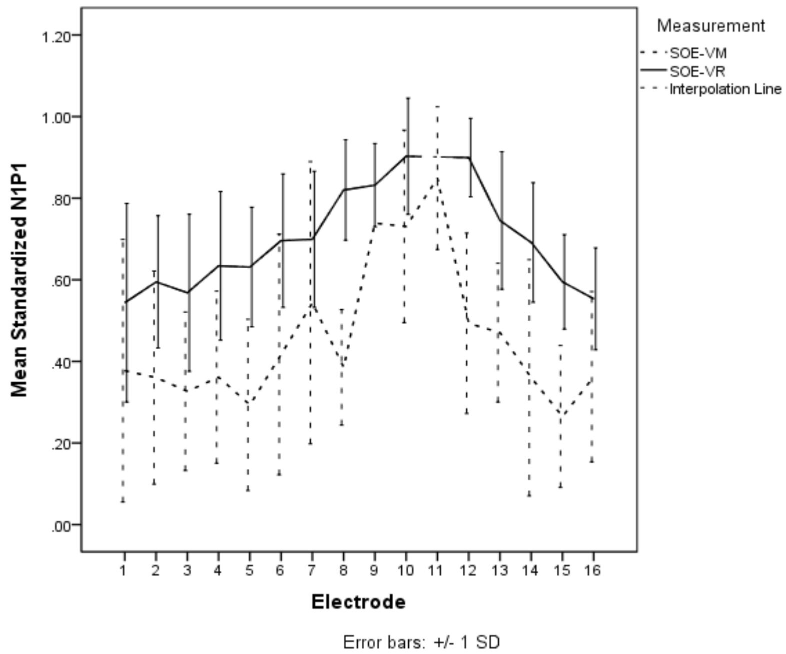

Figure 6.3c Mean (+- 1SD) normalized ECAP N1P1 amplitudes across Advanced Bionics recipients for SOE-VR (full line) and SOE-VM (dotted line) functions for *basal* (el11) probe locations.
ECAP=Electrically Evoked Compound Action Potential, SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

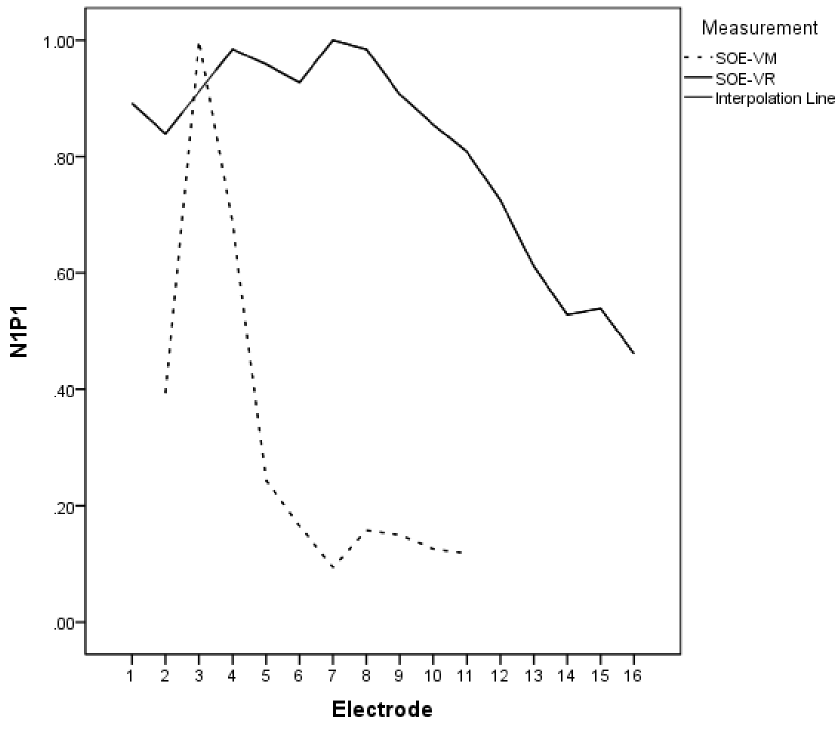

Figure 6.4a Individual example of normalized ECAP N1P1 amplitudes for recipient AB7 for SOE-VR (full line) and SOE-VM (dotted line) at *apical* (el3) probe electrode location.
SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

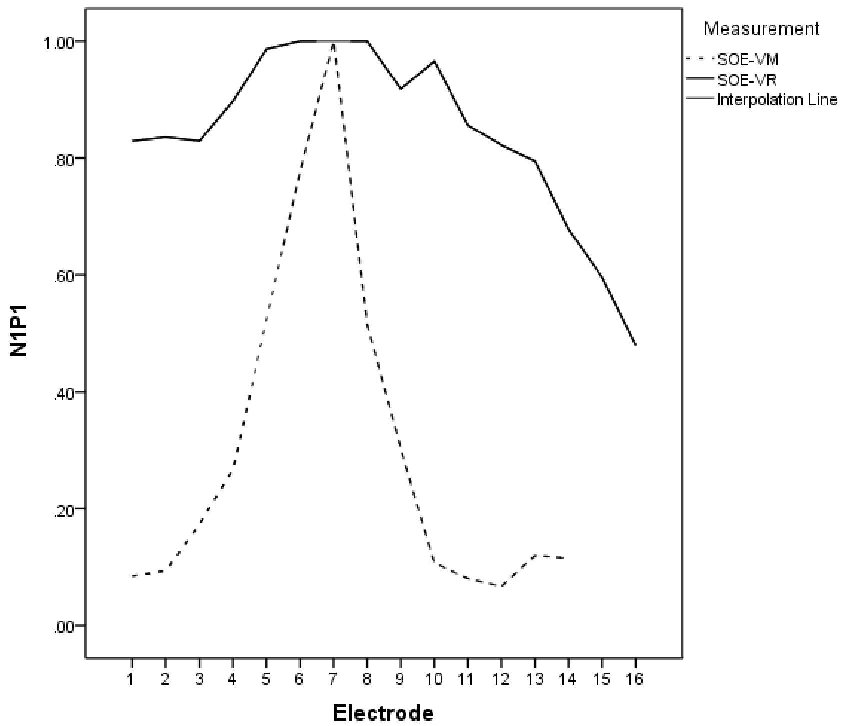

Figure 6.4b Individual example of normalized ECAP N1P1 amplitudes for recipient AB7 for SOE-VR (full line) and SOE-VM (dotted line) at the *middle* (el7) probe electrode locations.
SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

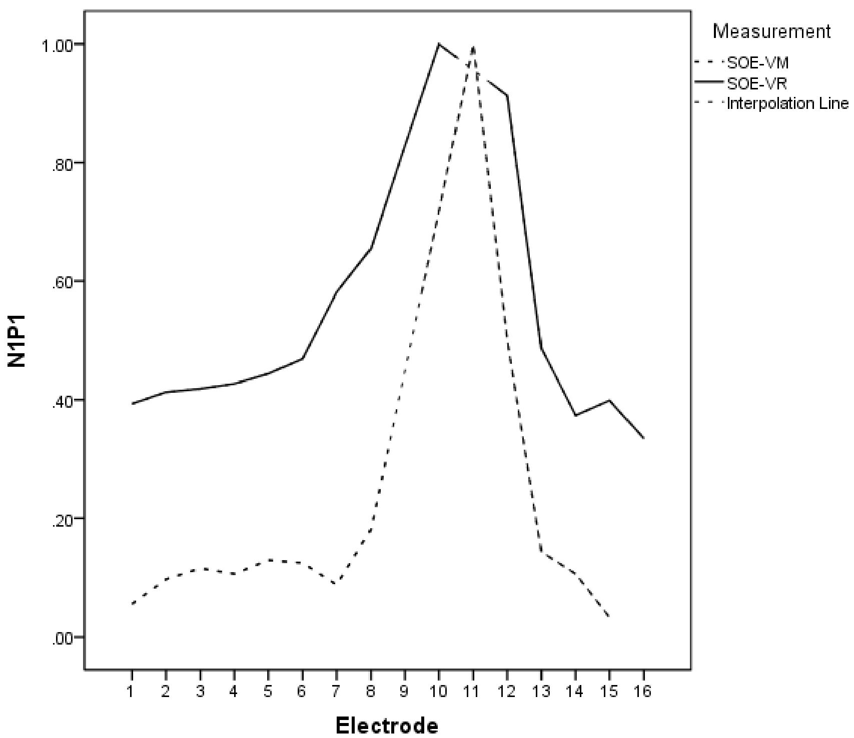

Figure 6.4c Individual example of normalized ECAP N1P1 amplitudes for recipient AB7 for SOE-VR (full line) and SOE-VM (dotted line) at the *basal* (el11) probe electrode location.
SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, el=electrode

A repeated–measures analysis of variance (ANOVA) was performed to examine the effects of probe electrode (el11, el7, el3) and measurement technique (SOE-VR, SOE-VM) on the mean normalized ECAP amplitude across each function. Analysis revealed a statistically significant result for measurement technique ($F(1,7) = 42.11$, $p < .001$), but no statistically significant result for probe electrode ($F(2,14) = 2.93$, $p > .05$). It was found that SOE-VR measurements were broader compared to SOE-VM measurements.

6.3.2 Experiment 2: Long-term stability of SOE-VM patterns over repeated trials

Studies comparing ECAP measures and psychophysical measures mostly report one trial for ECAP measures, whereas several trials for psychophysical measures are averaged. Hence assuming little variability in ECAP measures across repeated trials, however, this has not been systematically evaluated within different electrodes. This experiment examined whether ECAP SOE-VM patterns for three different probe electrodes differed significantly across repeated trials within recipients.

Thirteen recipients were included (**Table 6.1**) and for each recipient SOE-VM patterns for three probe electrodes were obtained at two test moments, with the second test moment planned approximately one year after the first one. For each measurement, probe level was defined as earlier described.

To obtain a more general comparison with respect to SOE-VM patterns, ECAP amplitudes were normalized within each subject and then averaged across subjects. **Figure 6.5** shows average normalized SOE-VM functions for both devices for three electrode locations at two test moments. The top and bottom row show data for Advanced Bionics and Cochlear subjects, respectively. For each device, two one-way ANOVA's were performed (test moment and electrode) on the mean normalized ECAP amplitude across functions. For both devices, no significant difference was found between both test moments (resp. $F(1,5) = 3.322$, $p > .05$ and $F(1,5) = .631$, $p > .05$) nor electrodes (resp. $F(2,5) = .798$, $p > .05$ and $F(2,5) = 2.526$, $p > .05$).

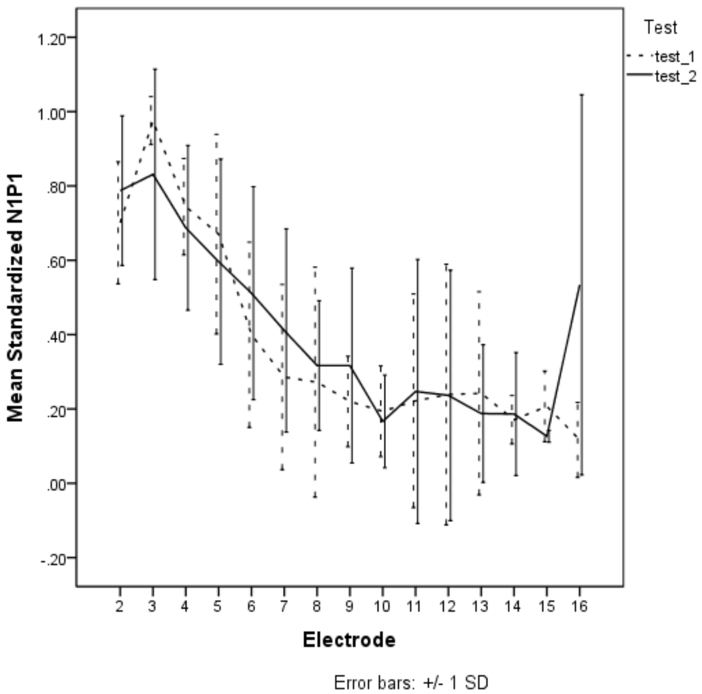

Figure 6.5a Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes across *Advanced Bionics* recipients *apical* (el3) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

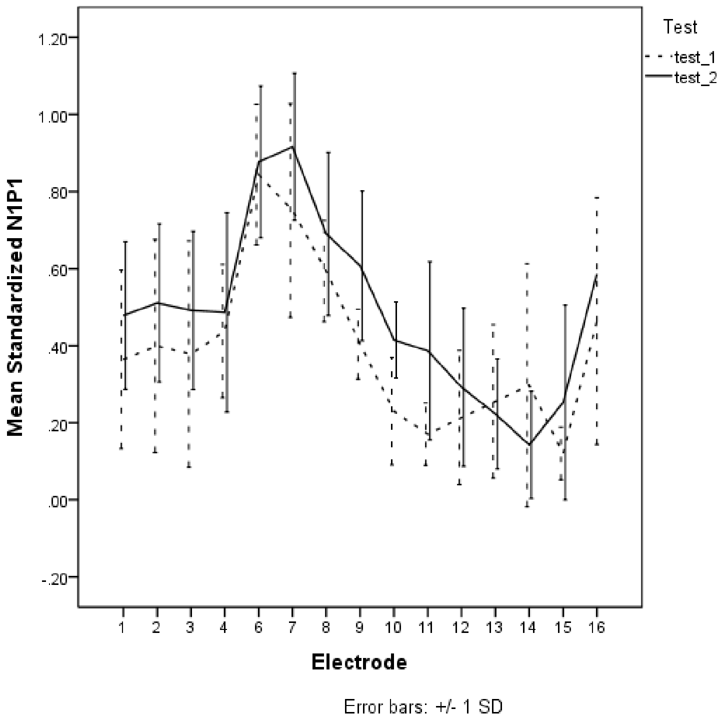

Figure 6.5b Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes across *Advanced Bionics* recipients *middle* (el7) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

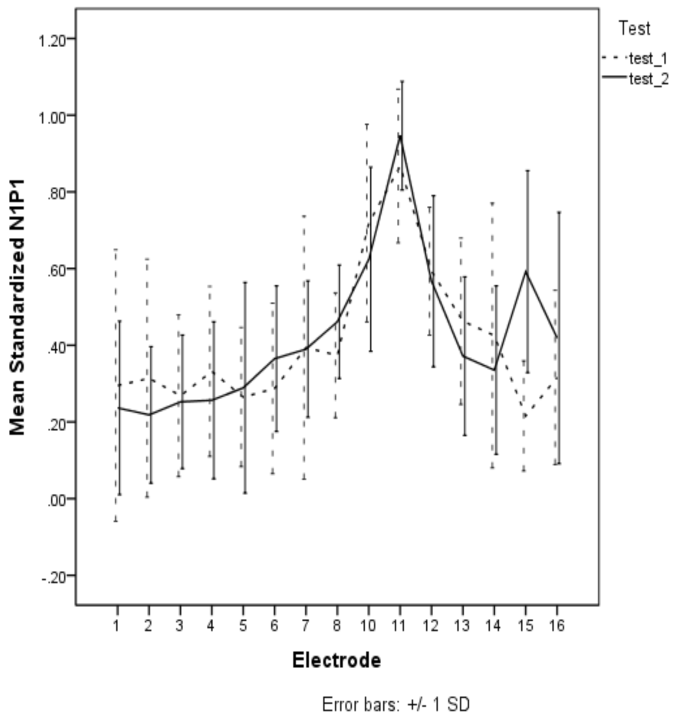

Figure 6.5c Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes across *Advanced Bionics* recipients *basal* (el11) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

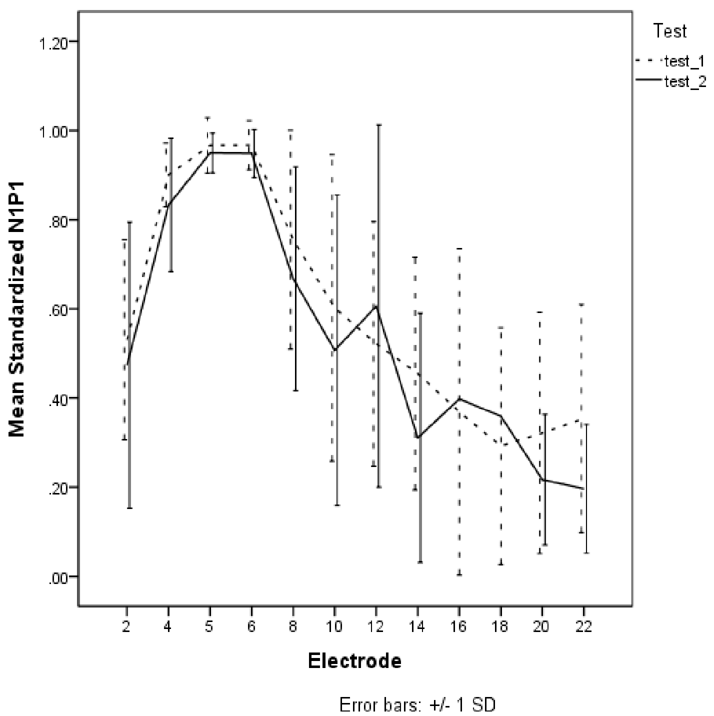

Figure 6.5d Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes across *Cochlear* recipients *apical* (el5) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

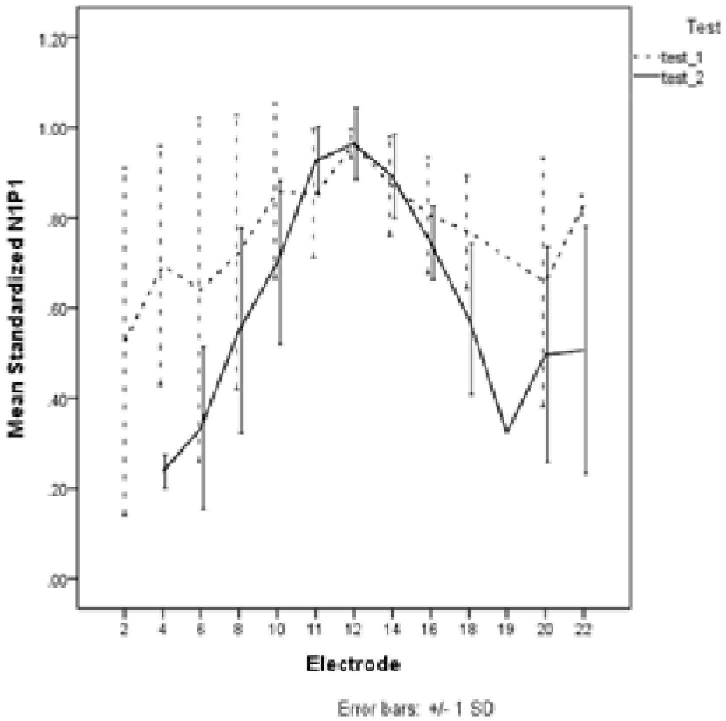

Figure 6.5e Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes for *Cochlear* recipients for *middle* (el 11) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

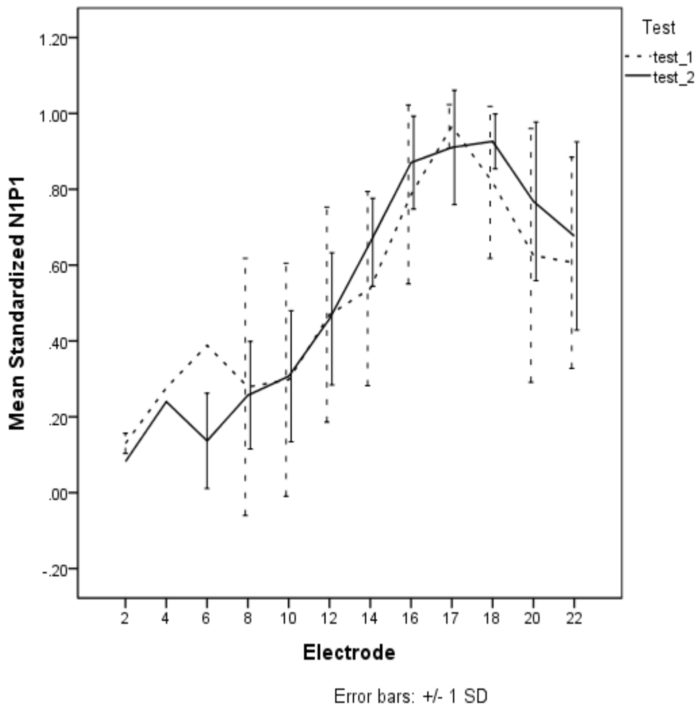

Figure 6.5f Mean (+-1SD) normalized repeated trails SOE-VM ECAP amplitudes for *Cochlear* recipients for basal (el 5) probe electrodes
ECAP=Electrically Evoked Compound Action Potential, el=electrode

Finally, to compare ECAP amplitudes across both devices, the width values were transposed from width in number of electrodes to width in millimeters (mm) by using an electrode spacing of 1.1 mm for the Advanced Bionics and 0.7 mm for the Cochlear internal devices.

A three-way ANOVA with device , test moment and electrode as independent variables and width (in mm) as dependent variable revealed no interaction effects, nor main effects for device ($F(1,60) = 3.587$, $p > .05$), test moment ($F(1,60) = .023$, $p > .05$) and electrode ($F(2,60) = .276$, $p > .05$).

6.3.3 Experiment 3: Correlation between speech perception and spatial profiles

The third goal of this study was to investigate the possible relationship between speech perception and ECAP spatial profiles. It was hypothesized that subjects with smaller widths would have better speech perception performance. Twenty adult CI recipients were included (**Table 6.1**). Results of the speech perception measurements in quiet (NVA40, NVA55, NVA70 and /aCa/) and in noise (SRT_n) are shown in **Table 6.3**. NVA scores and /aCa/ scores are shown in percentages correct, SRT_n results show the dB SNR level. Some subjects performed well on these tasks, (e.g. subject AB10) while others experienced great difficulties even in quiet (e.g. subject AB8). No significant differences were found between the devices with respect to speech perception data.

Table 6.3
Individual results on speech perception tests.

subject	NVA40 (%)	NVA55 (%)	NVA70 (%)	SRT_n (dB SNR)	aCa (%)
AB1	0	18	73	26	51
AB2	0	57	42	8	51
AB3	6	58	48	19	42
AB4	0	52	73	12	50
AB5	0	36	42	20	33
AB6	3	73	88	26	44
AB7	21	76	85	7	68
AB8	0	18	30	30	14
AB9	30	70	79	10	92
AB10	55	76	94	4	56
CO1	22	85	88	6	36
CO2	33	45	42	26	33
CO3	15	82	64	-3	58
CO4	0	10	24	24	11
CO5	0	73	76	8	29
CO6	24	67	73	12	43
CO7	18	52	76	8	69
CO8	52	69	91	3	61
CO9	0	39	55	15	50
CO10	55	82	88	6	60

AB= Advanced Bionics, CO=Cochlear, SRT_n= Speech Reception Threshold in noise, dB= decibel

Also relative information transmission scores for the features of voicing, place of articulation, affrication, manner of articulation and nasality are shown, both individual and averaged across all recipients (**Table 6.4**). Results revealed that overall, place cues were the least transmitted. Moreover, a large variability was seen across subjects: some recipients had difficulties across all five features (e.g. subject number CO14), while others obtained 100% scores (e.g. subject CO8).

Table 6.4
Individual scores on relative information transmission for five consonant features.

subject	ITr voicing	ITr place	ITr affrication	ITr manner	ITr nasality
AB1	0.57	0.37	0.9	0.84	0.63
AB2	0.31	0.4	0.85	0.9	1
AB3	0.54	0.11	0.61	0.76	0.82
AB4	0.8	0.39	1	0.82	0.57
AB5	0.3	0.26	0.29	0.32	0.22
AB6	0.74	0.25	0.54	0.66	0.62
AB7	0.55	0.6	1	1	1
AB8	0.03	0.07	0.02	0.03	0.02
AB9	0.82	0.81	1	1	1
AB10	0.28	0.43	0.89	0.94	1
CO1	0.1	0.3	0.61	0.58	0.25
CO2	0.19	0.22	0.61	0.7	0.85
CO3	0.84	0.4	0.33	0.61	1
CO4	0.01	0.06	0.14	0.16	0.17
CO5	0.49	0.14	0.41	0.52	0.63
CO6	0.22	0.4	0.54	0.67	0.72
CO7	0.84	0.48	0.82	0.84	0.89
CO8	0.74	0.49	1	1	1
CO9	0.84	0.14	0.61	0.7	0.72
CO10	0.55	0.42	0.79	0.87	1
mean	0.49	0.34	0.65	0.7	0.71
SD	0.29	0.19	0.3	0.27	0.32

AB=Advanced Bionics, CO=Cochlear, ITr=relative information transmitted, SD=standard deviation

Five multiple regression analyses were conducted to examine the effect of SOE-VM widths measured on 3 electrode locations (apical, middle and basal) for each of the five speech perception scores (NVA40, NVA55, NVA70, SRT_n, /aCa/). None of these regression analyses revealed a statistical significant result.

Furthermore, five multiple regression analyses were performed to gain insight in the possible relationship between ECAP widths on 3 electrode locations (apical, middle and basal) and each of the consonants features (voicing, place of articulation, affrication, manner of articulation and nasality). Again, no statistical significant results were obtained.

During the analysis of the raw data, it was found that for a number of SOE-VM profiles, maximum amount of masking did not occur with masker and probe on the same electrode. Overall, 47% of the included ECAP spatial profiles had a displaced peak of which 11% were double peaks. Twenty-five percent of ECAP profiles were skewed in an apical direction, while 29% toward the base. The majority of these displacements (in both directions) had a maximum amount of masking at an adjacent electrode of the probe electrode. For example, the maximum amount of masking for subject AB4 at the middle electrode occurred at electrode 6 instead of electrode 7. For only two SOE-VM profiles, the maximum amount of masking occurred at a non-adjacent electrode. For example, the maximum amount of masking for subject CO4 at the apical electrode occurred at electrode 20 instead of electrode 17. No differences were seen with respect to location of electrode on amount of displaced peaks. For the middle and basal electrode, 50% of ECAP profiles and for the apical electrode 44% was displaced.

To investigate the relation between the SOE-VM profiles and the speech perception tasks, five regression analyses were carried out with each speech perception measurement as dependent variable and presence of displaced peaks within each subject as independent variable, but no statistical significant results were found. Also the relation between the presence of displacements and ITr scores was examined by means of five regression analyses. Again, results revealed no statistical significant results. Furthermore, although not significantly different, highest ITr scores were obtained when no displacements were found. Recipients without displacements at the mid and/or apical electrode, achieved higher ITr scores for affrication, manner and nasality.

6.4 Discussion

6.4.1 Experiment 1: SOE-VR versus SOE-VM patters

The primary objective of this study was to compare two spatial measurement techniques: SOE-VR and SOE-VM. By means of both measurement techniques, spatial selectivity can be successfully measured along the electrode array. This study shows that spatial profiles across all subjects and for both measurement techniques have the same characteristics: a peak in the region of the masker electrode and a variable shape profile from exponential-like to more bell-shaped. Furthermore, not all profiles are symmetrically shaped. This is in accordance with Chatterjee and Shannon (1998). To gain more insight in the shape of the profiles, Electric Field Imaging measurements (EFI) of 5 recipients (AB3, AB5, AB6, AB9 and AB10) were collected (EFI, Advanced Bionics Corporation, Valencia, CA). These spatial profiles were obtained by applying an electric pulse at one electrode (el 11, 7 and 3) while recording the voltage potential at the remaining electrodes along the array. **Figure 6.6** shows the number of symmetrical and asymmetrical shaped profiles for SOE-VR, SOE-VM and EFI measurements for these 5 recipients. The majority of SOE-VM measurements was symmetrically shaped (11/15), while the majority of SOE-VR (8/15) and EFI (7/15) measurements was asymmetrically shaped. This asymmetry in EFI data was noted in former studies (Vanpoucke et al 2004a, 2004b). Probably, this asymmetry is due to the low-resistance path of the current flow from the apex towards the base, which is also seen within the SOE-VR measurements.

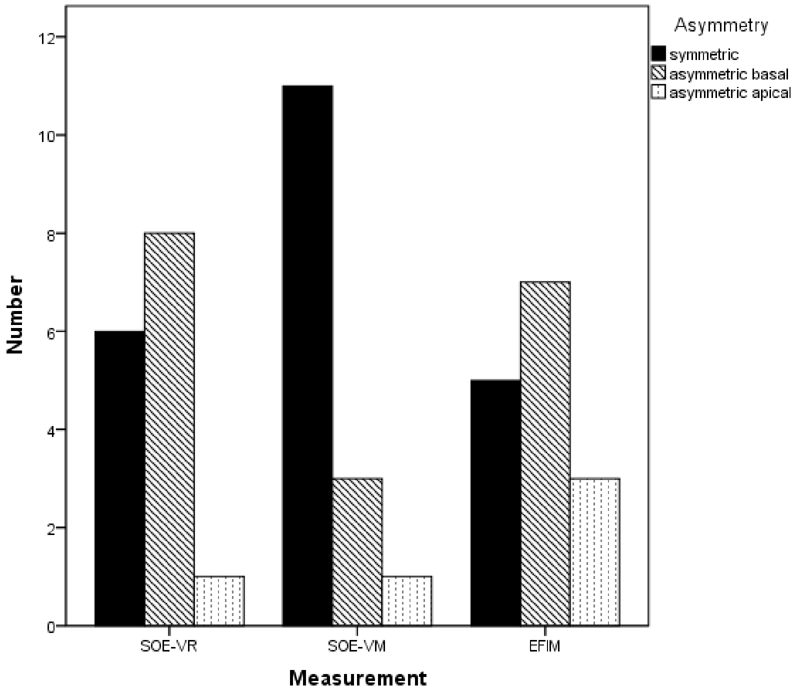

Figure 6.6 Bar charts for three spatial measurements (SOE-VR, SOE-VM and EFIM) showing the direction of displaced peaks (symmetric, asymmetric basal and asymmetric apical).
SOE-VR=Spread Of Excitation, Variable Recording, SOE-VM=Spread Of Excitation, Variable Masker, EFIM=Electrical Field Imaging Measurements

We also observed that SOE-VR patterns were significantly broader than SOE-VM patterns. This result is in accordance with Abbas et al (2004), Cohen et al (2004), Hughes and Stille (2010) and van der Beek et al (2012). As already explained by Hughes and Stille (2010) and van der Beek et al (2012) SOE-VR patterns might be broader because the current from both the stimulating contact and the nerve fibers spread through the surrounding fluid along the entire length of the cochlea since recordings are made at several contact points along the electrode array. SOE-VM patterns, on the other hand, are narrower because a fixed recording contact is used and consequently, the excitation of the stimulating pulse is shown.

Furthermore, no variability was found across different probe electrodes. This finding is in accordance with psychophysical studies of Hughes and Stille (2008) and Bierer and Faulkner (2010), who did not find tuning properties to be affected by channel location. Hughes and Abbas (2006a) performed ECAP measurements as a measure of channel interaction in individuals with a straight electrode array (Nucleus 24M) versus a perimodiolar array (Nucleus 24R(CS) Contour). Their results also showed no significant effect of electrode place on the width of the ECAP functions.

6.4.2 Experiment 2: Long-term stability of SOE-VM patterns over repeated trials

This experiment assessed the long-term stability of ECAP SOE-VM functions for three probe electrodes within adult CI recipients by examining possible width variations over repeated trials. Results showed that no significant differences could be found with respect to width measurements within our recipients, implying a good long-term stability. This result is in accordance with Hughes and Stille (2010). This study assessed the stability of ECAP functions by examining amplitude variations over repeated trials within 6 Cochlear and 6 Advanced Bionics adult CI recipients. They found that, in general, ECAP amplitudes were highly repeatable across measurements. However, Hughes and Stille (2010) repeated ECAP functions three or four times within one single data collection and did not repeat across longer time intervals. Moreover, for each subject, ECAP patterns were repeated for only one randomly chosen probe electrode. The current study, however, assessed possible variances in ECAP patterns across trials spaced across a one-yr interval for three probe electrodes within each subject.

With respect to probe electrode location, no significant statistical differences were found. Several authors have compared spatial measurements along the electrode array. Eisen and Franck (2005) used a forward masking paradigm to explore ECAP electrode interaction. They included 27 pediatric subjects (N = 16 AB HiFocus, N = 11 Cochlear) and performed ECAP functions at three probe electrode locations (AB: el5, el9, el13: Cochlear: el17, el11, el5). They found more electrode interaction at apical probe locations than at basal probe locations. Cohen et al (2004) performed spatial measurements on seven Nucleus 24 patients by means of the subtraction paradigm. They found no effect of probe electrode location when including a basal, a mid and an apical probe electrode. Busby et al (2008) measured spread of excitations on a dual electrode and two adjacent single electrodes at three positions on the array (el17+18, el11+12, el5+6) in nine adult Cochlear Freedom subjects by means of the ECAP forward masking method and found no significant difference between the 75% widths on linear best-fits at electrodes at the apical, mid and basal positions.

6.4.3 Experiment 3: Correlation between speech perception and spatial profiles

Theoretically, subjects with smaller widths at ECAP profiles should have better speech perception scores as narrow spatial profiles would allow for more independent information channels. Former studies could not confirm a relation between ECAP and speech perception scores (Hughes and Abbas, 2006b; Hughes and Stille, 2008; Tang et al 2011; van der Beek et al 2012).

Several reasons have been proposed by other researchers for the lack of correlation between ECAP and speech perception scores. Van der Beek et al (2012) argued that comparing their intra-operatively derived ECAP spatial profiles (and thus high level current) to speech tests obtained two years later, could account for the lack of correlation. Tang et al (2011) explained their results by the discrepancy in stimulation rate between the present measures (<= 100Hz) and the current speech processors (> 500Hz), besides a small sample size (N=5). Hughes and Stille (2008) revealed that subjects whose ECAP FM patterns correlated well with PFM patterns generally had the poorest speech perception and subjects with the poorest correlations had the best perception on monosyllabic word en phoneme scores (CNC monosyllabic word test) and sentences in noise (BKB-SIN). They hypothesized that better-performing subjects using higher-level auditory mechanisms within psychophysical tasks can better compensate for a shifted peak measured within the peripheral ECAP function. However, these studies only included speech perception percentage correct scores. Because ECAP spatial profiles are measured at the electrode-neural interface, the current study also included ITr scores of consonants by means of five features (voicing, place of articulation, affrication, manner of articulation and nasality). Furthermore, this test material is known to be less redundant compared to for example sentences. Nevertheless, results revealed no statistical significant results which could correlate ECAP derived spatial profiles with percentage correct scores nor ITr. To our knowledge, no other studies have explored the possible relationship between ITr and ECAP derived spatial profiles. Henry and co-workers (2005) used a psychophysical spectral ripple test to examine spectral peak resolution and vowel and consonant percentages correct scores in quiet. Regression analyses showed a significant moderate linear correlation between spectral peak resolution thresholds and both vowel and consonant recognition, suggesting that the ability to resolve spectral peaks in a complex acoustic spectrum is associated with speech recognition. In addition, Nelson and colleagues (2011) found a weak relation between psychophysically obtained forward-masking spatial tuning curves at a mid electrode location and transmitted information for vowel second formant frequency.

However, using spectral ripple or any other psychophysical test routinely in a clinical setting is impossible due to the large amount of time needed to perform these measurements.

Within this study, 47% of ECAP derived spatial profiles had a displaced peak. Hughes and Stille (2008) listed the displaced peaks both in PFM and ECAP FM measurements. Displaced peaks in ECAP FM occurred in twenty-one percent. Within their study, a difference between subjects with a poor correlation between ECAP FM and PFM on the one hand and subjects with a strong correlation between both measurements on the other hand, was found. It was shown that 39% of all ECAP masking functions in the poor correlation group had displaced peaks as opposed to the strong correlation group in which only 11% of displaced peaks was found. A t-test was performed to examine the possible relationship between displaced peaks and speech perception performance and no statistically significant difference was found. Also within the current study, no significant correlation was obtained between displaced peaks and speech perception measurements even when the consonant features were taken into account. Throckmorton and Collins (1999) did find a significant correlation between averaged PFM and speech perception tests (Iowa Medial Consonants, NU-6 phonemes and CID Everyday Sentences). However, the sample size was relatively small (N=7).

To explore the possible relationship between the presence of displaced peaks and speech perception, data of 9 recipients were further analyzed in the present study. Data of recipients without displacements (AB1, AB6, AB7 and AB8) were compared to data of recipients whose SOE-VM profiles at all 3 probe electrodes were displaced (AB4, CO2, CO3, CO4 and CO9). It was hypothesized that recipients without displacements would achieve better scores on speech perception tests. Independent t-tests were performed and no statistical significant differences between both groups were found. These results tentatively suggest that the presence of displaced peaks do not equate to lower speech perception scores. Hughes and Stille (2008) hypothesized that the presence of displaced peaks could be compensated by the use of higher-level auditory mechanisms. However, in this study, the closed-set identification test of consonants which makes use of lower-order auditory and linguistic mechanisms, did not show a significant difference between recipients with or without displaced peaks. When accounting for duration of deafness prior to CI, it was found that recipients whose SOE-VM profiles were all displaced, had a longer duration of deafness prior to CI compared to recipients without displacements (except for AB4: meningitis). Spiral ganglion loss might be accounted for the presence of displaced peaks since a longer duration of deafness is sometimes characterized by a lower degree of spiral ganglion survival. Another possible explanation for the absence of differences in speech perception between recipients with and without peak displacements, might be found within temporal and/or intensity cues. Recipients having lower spectral integration might compensate for this by having higher temporal and/or intensity integration cues. Gaining insight in the temporal and intensity cues was not the scope of the current study. In future, it would be interesting to assess spectral, temporal and intensity integration cues in a large group of CI recipients to explore possible compensation mechanisms within these integration cues, leading to better speech perception scores.

6.5 Conclusions

Research has shown that as a consequence of broader excitation patterns, channel interaction might increase (Boëx et al 2003; Cohen et al 2004; Abbas et al 2004), which may result in poor spectral resolution and consequently poorer speech recognition (Throckmorton and Collins 1999; Henry et al 2005; Litvak et al 2007; Won et al 2007). The primary purpose of this study was to gain insight in SOE-VR and SOE-VM patterns for three probe electrode locations. It was found that normalized SOE-VR functions were broader than normalized SOE-VM functions. As stated by Hughes and Stille (2010) this can be explained by the evidence that SOE-VR measures reflect volume conduction of the ECAP response along the length of the cochlea whereas SOE-VM functions reflect the relative overlap of neural populations recruited by the masker versus probe. Furthermore, the long-term stability of SOE-VM patterns for three probe electrode locations over repeated trials was investigated. Normalized data showed that SOE-VM ECAP measures were highly stable across repeated trials over a one-yr time period. This suggests that physiological spatial forward masking measurements, obtained at least three months after the first fitting, can be re-used when conducting studies that investigate possible relationships between these types of ECAP measures and psychophysical measurements. In clinical settings a large variation is seen on speech perception measurements within CI recipients. This study showed no significant correlation between ECAP derived spatial profiles measured at three electrode locations and speech perception measurements nor relative information transmitted for five consonant features. Moreover, the presence of peak displacements could not account for poorer speech perception results or lower relative information transmitted. These results suggest that ECAP derived spatial profiles with respect to width measurements and peak displacements should not be used to predict speech perception measurements within clinical settings.

Chapter 7

Assessing and predicting speech perception in early-deafened, late-implanted adults

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Submitted to Audiology and Neuro-otology

Abstract

Objectives

First, speech perception outcomes of early-deafened, late-implanted CI users are evaluated by using a thorough test battery including speech perception tests in quiet and in noise varying in their demand on central function ranging from lower to more higher-level auditory and linguistic mechanisms. Second, errors in speech production are used as a predictive factor for speech perception outcome postimplantation.

Methods

Data were collected from 19 early-deafened, late-implanted adults (N = 12 Nucleus, N = 5 Advanced Bionics, N = 2 Neurelec).

Results

After implantation, averaged speech perception scores improved, with a mean phoneme score of 53.03% at 70 dB SPL. Multiple regression analyses showed that errors in speech production could account for 78% of the variance for speech perception at 70 dB SPL, indicating that fewer errors in speech production are a reliable predictor of good speech perception outcome postimplantation.

Conclusions

Early-deafened, late-implanted adults obtain postimplant speech perception results beyond their preimplant state results for measurements that do not require higher-level auditory and linguistic mechanisms such as pure tone audiometry and consonant identification. Secondly, errors in speech production can be used as a predictor of speech perception outcome postimplantation in quiet and in noise for early-deafened, late-implanted adults. Using errors in speech production as a predictor for speech perception postimplantation will facilitate candidate counseling and selection in the group of early-deafened adult CI candidates.

7.1 Introduction

Over the last twenty years, cochlear implantation has widely been accepted as an effective treatment for patients with bilateral profound SNHL. It is well known that speech perception scores improve considerably in congenitally deafened early implanted children and in postlingually deafened adults. One category of patients that remains to be explored is the group of early-deafened, late-implanted adults.

Since 2002, less than fifteen studies have been published that gained insight in the speech perception outcome of this particular group (Kaplan et al, 2003, Schramm et al, 2002, Waltzman et al, 2002, Teoh et al, 2004a, 2004b, Moody-Antonio et al, 2005, Klop et al, 2007, Santarelli et al, 2008, Yoshida et al, 2008, Kos et al, 2009, Shpak et al, 2009, van Dijkhuizen et al, 2011, Yang et al, 2011, Caposecco et al, 2012). Overall, these studies reported statistically significant, but modest improvement in speech perception postimplantation compared to the preimplant levels. However, the averaged achieved performance of early-deafened, late-implanted adults never reached the same results obtained within postlingually deafened adults.

There are a number of shortcomings in the available literature. In the majority of studies investigating CI performance in early-deafened, late-implanted adults, speech perception was represented by a single measurement. Since stimuli were presented at only one intensity level (mostly 70 dB SPL), speech testing was never assessed with measures that approximate everyday listening situations (Firszt et al, 2004). Furthermore, few studies included a test battery that makes use of several speech tests that vary on their demand on central function ranging from lower to more higher-level auditory and linguistic mechanisms. Mostly, word scores on Consonant-Vowel-Consonant (CVC) tests and/or sentence recognition scores in quiet were used (Klop et al, 2007, Schramm et al, 2002, van Dijkhuizen et al, 2011). Only Santarelli and colleagues (2008) implemented levels with different degrees of difficulty and evaluated speech perception performance in quiet by means of word discrimination length, word and sentence identification, phoneme identification and word and sentence recognition. Scores on all tests significantly improved after CI, although substantial inter-subject variability was found. However, speech perception measurements in noise were not included. To mimic complex daily life situations, speech perception in noise must be included in the postimplantation test battery (Teoh et al, 2004a, 2004b, Waltzman et al 2002, Sphak et al, 2009).

Because of the large inter-subject variability within early-deafened, late-implanted CI users, it would be of interest to gain more insight in factors affecting speech perception postimplantation. Recently, two studies have investigated the relative contribution of several factors to speech perception outcome in early-deafened, late-implanted adults. Van Dijkhuizen and colleagues (2011) implemented speech intelligibility as a predicting variable for speech perception in early-deafened, late-implanted adults. They presented speech samples from 25 CI candidates to two normal-hearing listeners. From this group of 25 CI candidates, 9 participants with above averaged intelligibility were selected for CI. Speech perception data from these 9 CI users, obtained 12 months postoperatively (CVC) at 65 dB SPL, were used as the dependent variable within a stepwise multiple regression analysis. This analysis showed that speech intelligibility measured before CI was the only significant predictor, accounting for 62% of the variance in speech perception postimplantation. However, in the study by van Dijkhuizen and colleagues (2011) only the subjects obtaining above-average speech intelligibility results were selected for CI. This restriction possibly had a moderating effect on the correlation between intelligibility and postimplant speech perception. Furthermore, speech intelligibility was expressed as the percentage of correctly identified vowels/diphthongs, consonants, words or sentences. No in-depth analysis of errors in speech production was made. Caposecco and colleagues (2012) also identified prognostic factors associated with positive speech perception outcomes. Higher scores postimplantation were predicted by an oral mode of communication in childhood, a progressive HL and a HA worn on the implant ear up to the time of implantation. Sixty-three percent of the variance in speech perception scores was explained by these three variables.

Overall, there are two major shortcomings in literature to date. Resulting from these shortcomings in literature, the aims of the current study are two-folded. First, speech perception outcomes of early-deafened, late-implanted CI users were assessed using a thorough test battery that permits evaluating a hierarchy of auditory skills ranging from sound detection through comprehension of sentences in noise. Therefore, pure-tone audiometry (PTA), closed-set consonant identification (/aCa/), speech perception (CVC) at three intensity levels (40, 55 and 70 dB SPL), speech perception (sentences) in quiet and noise will be included in this study. Second, the relationship between speech production and speech perception in this population was assessed including data with a large range in speech production performance. Errors in speech production (omissions, substitutions and distortions) will be used as a predictive factor for speech perception outcome postimplantation.

7.2 Materials

7.2.1 Participants

Participants were recruited using the following criteria: (1) early-onset bilateral sensorineural HL, (2) unilaterally implanted at 13 yr or older, and (3) minimum one year of CI experience. Nineteen adult patients met the inclusion criteria. **Table 7.1** gives an overview of relevant patient characteristics. All participants had a history of HA use. Average age at fitting first HA was 4.58 yr (SD 3.25, range 1 to 14 yr). PTA (measured at 0.5, 1 and 2 kHz) unaided for the implanted ear prior to implantation was 106 dB HL (SD = 12.98, range 75-123 dB). Average age at implantation was 32 yr (SD = 8.87; range 13 to 50 yr). None of the participants primarily used sign language.

Table 7.1
Details on patient characteristics.

patientnumber	gender	age (yr)	etiology	communication mode	internal part	external part	participation in speech perception	participation in speech production
1	M	38	mat. rubella	oral	CI24R	Freedom	x	x
2	F	52	unknown	oral+manual	CI24RECA	Freedom	x	
3	F	34	Usher type 2	oral+manual	HiRes90K	Harmony	x	x
4	M	43	measles	oral+manual	CI24RECA	Freedom	x	x
5	M	42	mumps	oral	DX10	Digisonic	x	x
6	M	45	unknown	oral	CI24RCA	Freedom	x	x
7	F	38	Noonan	oral	DX10	Digisonic	x	
8	F	46	rubella	oral+manual	CI24RECA	Freedom	x	x
9	F	24	unknown	oral	CI512	CP810	x	
10	M	40	unknown	oral	HiRes90K	Harmony	x	
11	M	25	meningitis	oral+manual	CI24M	Freedom	x	
12	F	41	unknown	oral	CI24RECA	Freedom	x	
13	F	32	Cx26	oral	CI512	CP810	x	x
14	F	40	unknown	oral+manual	CI512	CP810	x	x
15	F	39	rubella	oral	CI24RECA	Freedom	x	
16	M	41	unknown	oral	HiRes90K	Harmony	x	x
17	F	32	unknown	oral	HiRes90K	Harmony	x	x
18	F	48	unknown	oral	HiRes90K	Harmony	x	x
19	F	24	cCMV	oral+manual	CI24RECA	Freedom	x	

mat. rubella= maternal rubella, M=male, F=female, yr=year, Cx26=Connexine 26, cCMV=congenital cytomegalovirus

7.2.2 Outcome measures

The test battery included PTA, speech perception tests and speech production tests.

7.2.2.1 Pure-tone audiometry and speech perception

PTA and speech perception tests were performed 12 months postoperatively in a sound booth, with warble tones, speech and noise materials presented at 0° azimuth at 1 meter distance from the CI recipients. Tests were performed with the recipients own HA (preimplantation) or speech processor (postimplantation) at their daily program. Three speech perception tests were collected in the aided condition (CI alone). The first speech perception task was an identification of consonants (C) in an intervocalic /a/ context (12 /aCa/ nonsense syllables) with C being /m, n, p, b, k, s, z, f, v, t, d and ɣ/. The stimuli were presented at 65 dB SPL. Each /aCa/ nonsense syllable was presented 6 times in random order. Participants responded to the stimuli by clicking one of the 12 /aCa/ nonsense syllables on a computer screen. Tests were self-paced, they did not proceed before an answer was given. No training or feedback was provided, except for familiarization with the different response alternatives before testing. Relative information transmitted (ITr) was calculated for five features for each of the subjects (Miller and Nicely, 1955). ITr is expressed as a value between 0 and 1 and is the ratio of the transmitted information calculated from the confusion matrix to the maximal possible information transferred by the stimuli and categories under test. The consonants were classified according to the features 'voicing, place of articulation, affrication, manner of articulation and nasality'. The classification of the consonants is shown in **Table 7.2**. Second, percent-correct performance at phoneme level was measured using Dutch recorded CVC 3 phoneme monosyllabic words (NVA, Wouters et al, 1994). Speech perception was measured in quiet at 70, 55 and 40 dB SPL. For each condition a list of 11 words was used and scoring was based on percent-correct performance at phoneme level. Within the third speech perception test, Speech Reception Threshold (SRT) was measured adaptively in quiet (SRT_q) and noise (SRT_n) by means of the LIST sentences, representing speech in daily life (van Wieringen and Wouters, 2008). The speech level was varied and the speech-weighted noise available for the LIST was kept constant at 60 dB SPL. The SRT is the sound level of the speech signal at which 50% of the presented speech sentences is correctly identified. The SRT was determined using a simple up-down (1 up, 1 down) procedure with a 2-dB step size. The first sentence was repeated with increasing level until it was correctly identified by the subject. The level of subsequent sentences was presented once at a 2 dB lower or higher level than the previous sentence, depending on whether the latter was identified correctly or not. A response was judged correct if and only if all key words of the sentence were repeated correctly. The adaptive procedure continued until 10 reversals occurred, and then the speech levels of sentences 6 to 10 and the imaginary 11th sentence were averaged to obtain the SRT value.

Table 7.2
Classification of the consonant features.

	p	t	k	b	d	m	n	v	z	s	f	ɣ
voicing	0	0	0	1	1	1	1	1	1	0	0	1
place	0	1	2	0	1	0	1	3	1	1	3	2
affrication	0	0	0	0	0	0	0	1	1	1	1	0
manner	0	0	0	0	0	1	1	2	2	2	2	0
nasality	0	0	0	0	0	1	1	0	0	0	0	0

7.2.2.2 Speech production

To assess the participants' speech production, speech samples were elicited by means of a picture naming test 12 months postoperatively (Van Borsel, 1996). These speech samples were elicited in a sound-proof booth. Participants were asked to name 135 drawings of common objects and actions. Hence, a speech sample was elicited containing instances of all Dutch single speech sounds and most clusters in all permissible syllable positions. A directional microphone was placed at about 30 cm from the patient's mouth. Furthermore, those speech samples were video-recorded (Sony HDR-SR1) for further phonetic analysis. The consonant productions were compared with target productions and analyzed for error types at the segmental level making a distinction between distortions (i.e. the deviant production of a target sound not crossing phoneme boundaries), substitutions (i.e. the deviant production of a target sound crossing phoneme boundaries) and omissions (i.e. non-productions of a target sound) of consonants (Van Borsel, 1996). The analyses were based on a narrow phonetic transcription made by an experienced professional in speech and language pathology using the symbols and diacritics of the International Phonetic Alphabet

(IPA, 1949). Intra en inter judge sound-for-sound agreements were calculated based on a second transcription on 10% of all speech samples. For the intra judge agreement, the same professional transcribed the selected speech samples within a six-week time interval, without referring to the first transcription. For the inter judge agreement, another experienced professional in speech and language pathology transcribed 10% of all speech samples. The concordance values of these intra en inter judges agreements were 92 and 87% respectively. Because speech production testing is not routinely performed during yearly follow-up measurements, only 11 subjects were willing to come over to the clinic to participate.

All statistical analyses were conducted using SPSS version 19.0. The study was approved by the Institutional Review Board of the Ghent University Hospital and was conducted in accordance with the ethical standard stipulated in the Helsinki Declaration (2008) for research involving human subjects.

7.3 Results

7.3.1 Pure-tone audiometry and speech perception outcomes

Preimplant en postimplant performance for aided PTA thresholds and aided speech perception scores (NVA) at 70 dB SPL were compared. Aided measurements before implantation were obtained while wearing the HA at the side of implantation. Before implantation mean aided PTA thresholds were 66.0 dB HL (SD = 13.92, range 40-93) and aided speech perception at 70 dB SPL was 10.72% (SD = 16.36, range 0-60). After implantation, PTA results revealed an average aided threshold of 28.95 dB HL (SD = 9.05, range 18-48). Also averaged speech perception scores improved (53.05%, SD = 33.82, range 0-100). Paired samples t-tests revealed that on average both differences between preimplant and postimplant scores were significant, implying better scores postimplantation (PTA $t(16) = 11.88$, $p < .001$; NVA70 $t(17) = -5.76$, $p < .001$).

Table 7.3 displays the results obtained for the speech perception tests postimplantation. Average consonant identification scores (/aCa/) were 40.25% (SD = 27.82, range 11-92). Average speech perception scores at 40, 55 and 70 dB SPL were 9.88% (SD=17.05, range 0-48), 43.82% (SD = 30.89, range 0-82) and 53.05% (SD = 33.82, range 0-100), respectively.

Also, SRT's in quiet and noise were gained (**Figure 7.1**). Six (out of 14, patientnumber 1, 2, 7, 8, 11 and 19) subjects were not able to perform this test in quiet, even when the intensity level of the speech material was set to the maximal output level. The average SRT_q for the remaining 8 participating subjects was 60.0 dB (SD = 9.63, range 47-77). Similarly, 8 (out of 14, patientnumber 1, 2, 4, 6, 7, 8, 11 and 19) subjects were not able to perform this test in noise. The average SRT_n (with a constant speech-weighted noise level of 60 dBSPL) for the remaining 6 subjects was 69.17 dB (SD = 2.99, range 66-73).

Table 7.3
Speech perception outcomes 12 months post-implantation

Patientnumber	NVA40 (%)	NVA55 (%)	NVA70 (%)	aCa (%)
1	0	0	3	11
2	0	0	4	18
3	30	70	79	92
4	0	55	48	24
5	6	73	91	/
6	0	58	48	22
7	0	52	52	/
8	0	0	9	18
9	48	76	73	51
10	36	67	84	/
11	0	10	15	21
12	/	/	100	/
13	0	34	28	/
14	0	31	51	22
15	/	/	83	/
16	6	70	79	69
17	0	67	76	79
18	42	82	85	56
19	0	0	0	/

aCa=consonant identification task, NVA40=speech perception (phonemescore) at 40 dB SPL, NVA55= speech perception (phonemescore) at 55 dB SPL, NVA70=speech perception (phonemescore) at 70 dB SPL, '/' implies that the test has not been performed.

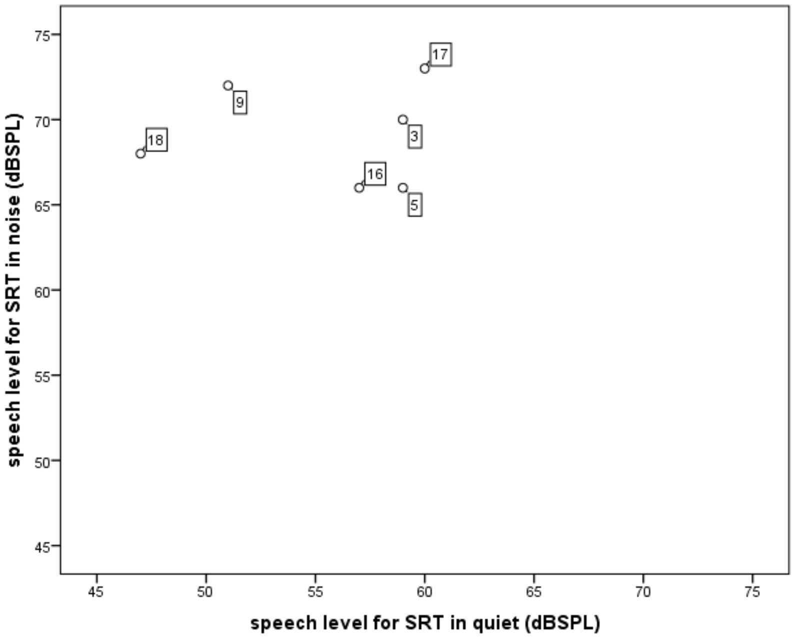

Figure 7.1 Scatter plot for speech level for SRT in quiet (dBSPL) at the X-axis and speech level for SRT in noise (dBSPL) at the Y-axis obtained 12 months postoperative. The noise level was kept constant during SRT in noise testing at 60 dBSPL. Each dot represents a single subject (patientnumber is indicated in the squares).
SRT=Speech Reception Threshold.

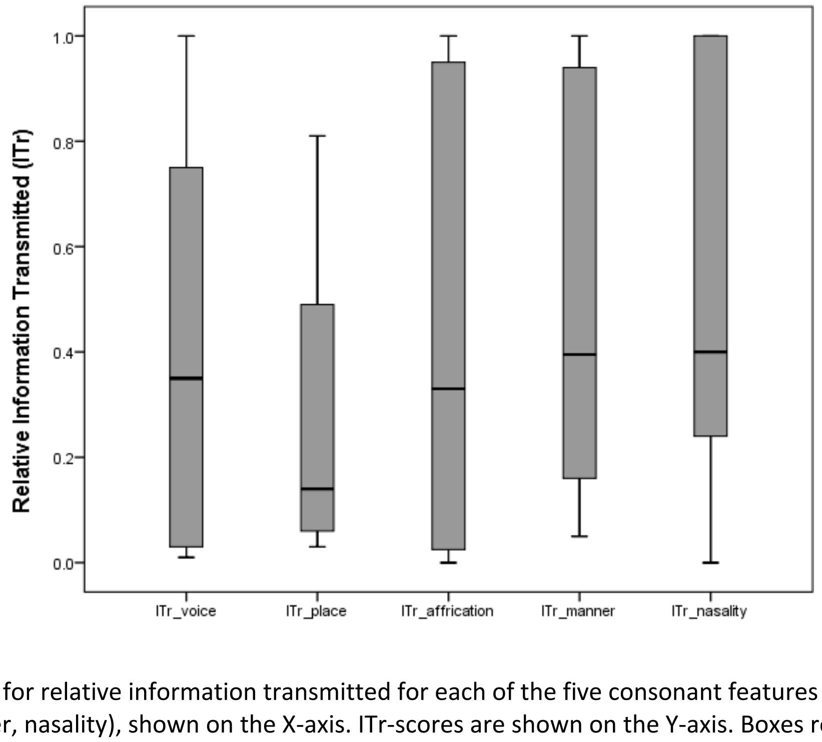

Figure 7.2 Box plots for relative information transmitted for each of the five consonant features (voicing, place, affrication manner, nasality), shown on the X-axis. ITr-scores are shown on the Y-axis. Boxes represent the median (thick horizontal line), lower and upper quartiles (ends of boxes), minimum and maximum values (ends of whiskers).
ITr=relative information transmitted

Relative information transmission scores for the consonants were obtained for the features of voicing, place of articulation, affrication, manner of articulation and nasality (**Figure 7.2**). A higher score indicates a better relative information transmission. Results revealed that overall, place cues were the least transmitted and a large inter-subject variability was found.

7.3.2 Speech production outcomes

Figure 7.3 demonstrates the results of speech production data. On average, the largest amount of errors was found within distortions (12.97%, SD = 9.63, range 0-24.71), followed by substitutions (6.59%, SD = 5.63, range 0-13.71) and omissions (3.90%, SD = 5.66, range 0-18.71). **Figure 7.4** shows the averaged relative distribution of errors (omissions, substitutions and distortions) for each consonant. First, note that a large difference in error rate is found among the consonants. With an error rate of 66.33%, most errors are made for /z/, while the least errors (2.57%) are found for /k/. Second, the type of errors made, also differ between the consonants. For the /S/, 42.74% (out of 56.02% of errors) was produced with a distortion, while for /b/ the only errors made were omissions (16.12%).

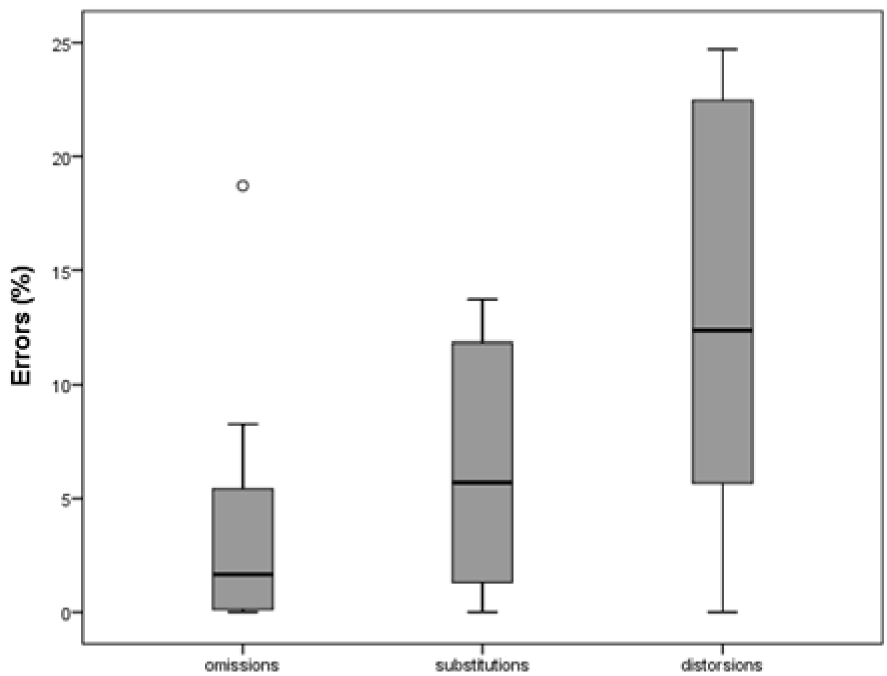

Figure 7.3 Box plots for errors in speech production (omissions, substitutions and distortions), shown on the X-axis. Percentage of errors is shown on the Y-axis. Boxes represent the median (thick horizontal line), lower and upper quartiles (ends of boxes), minimum and maximum values (ends of whiskers), outliers (values between 1,5 and 3 box lengths from either end of the box - circles).

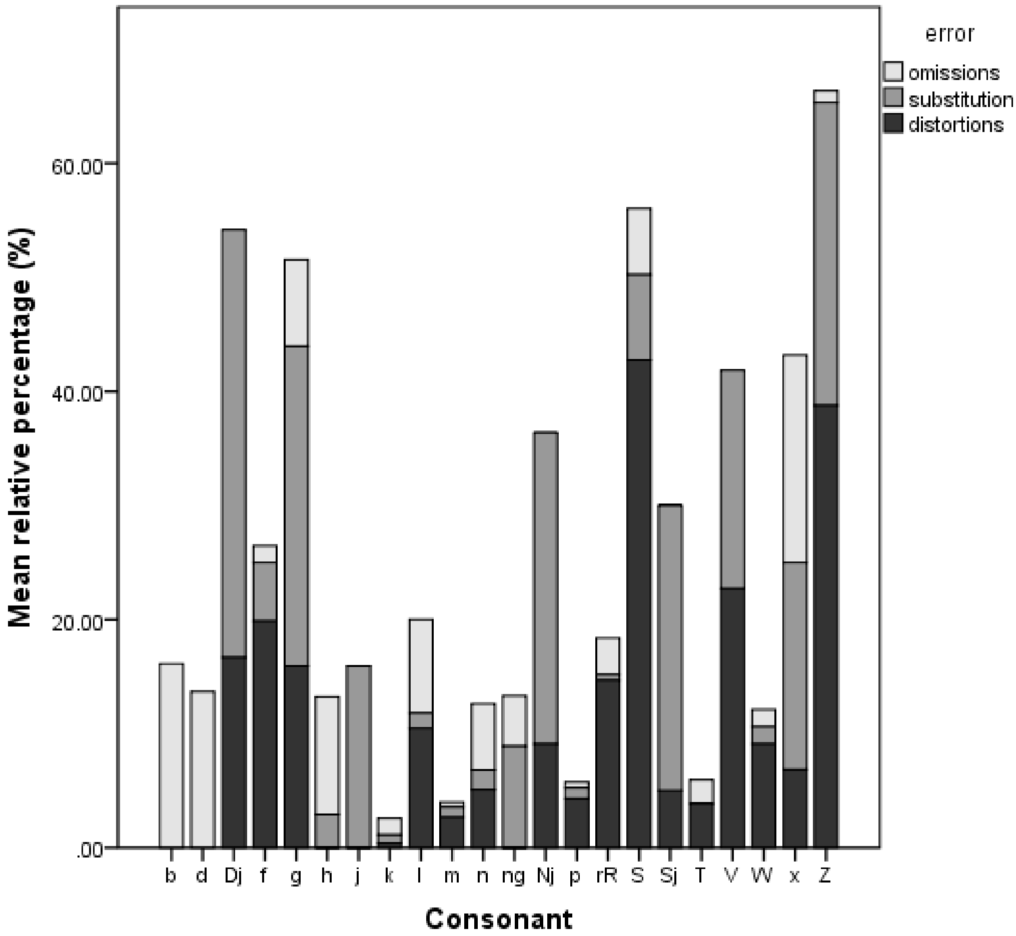

Figure 7.4 Bar chart of errors in speech production for each of the consonants. Consonants are shown on the X-axis. Mean relative percentage of errors in speech production (omissions, substitutions and distortions) is shown on the Y-axis.

7.3.3 Factors related to speech perception outcomes

The correlations between speech perception measurements (NVA40, NVA55, NVA70 and /aCa/) and several variables related to the patient's characteristics are shown in **Table 7.4** (Bonferroni correction applied). A significant negative correlation between omissions, substitutions and distortions was found for NVA55 and NVA70, implying higher speech perception scores when fewer errors in speech production were seen.

Table 7.4
Correlation between speech perception postimplantation and several independent variables.

Patient Characteristic	**Correlation Coefficient**				
	NVA40	NVA55	NVA70	aCa	PTA post
Omissions	-0.392	-0.89**	-0.844**	-0.643	0.003
Substitutions	-0.587	-0.85**	-0.917**	-0.813	-0.321
Distortions	-0.498	-0.644	-0.764*	-0.587	-0.356
PTA pre unaided CI side (dB)	0.071	0.211	0.138	-0.468	0.413
PTA pre aided CI side (dB)	-0.127	-0.171	-0.03	-0.154	0.307
NVA70 pre aided CI (%)	0.257	0.415	0.505	0.692	-0.243
communication mode	-0.371	-0.541	-0.517	-203	0
age at first HA (yr)	0.43	0.478	0.481	0.153	-0.437
congenital or non-congenital	0.507	0.473	0.453	0.435	-0.31

NVA40= speech perception (phonemescore) at 40 dB SPL, NVA55=speech perception (phonemescore) at 55 dB SPL, NVA70=speech perception (phonemescore) at 70 dB SPL, aCa=consonant identification perception test, PTA = Pure Tone Audiometry (0.5, 1 and 2 kHz), HA=hearing aid, yr=year.

Correlations are expressed as Pearson's r correlation coefficients
except for 'communication mode' and 'congenital or non-congenital'
(Spearman's rho coefficients (italicized))
*Significant at p <.05
**Significant at p <. 01

7.3.3.1 Errors in Speech Production

Spearman correlation's between omissions, substitutions and distortions showed a significant linear relationship. Furthermore, Cronbach's alpha between these measurements of speech errors, was .84, suggesting that the patient information captured by these three measurements was redundant. Therefore, a Principal Component Analysis (PCA) was conducted. PCA was performed on the scores of omissions, substitutions and distortions. One factor had an eigenvalue over Kaiser's criterion of 1 and explained 79.8% of the variance. This extracted factor stands for the speech errors made by the participants and is therefore referred to as 'Errors in Speech Production' (ErrSP). **Table 7.5** shows the ErrSP scores for each of the eleven subjects who participated in the speech production measurements.

Table 7.5
ErrSP scores for 11 early-deafened, late-implanted adults.

Case Number	ErrSP
1	1.52246
3	-.59382
4	.61539
5	-1.15594
6	-.27587
8	.79770
13	1.04577
14	.85409
16	-1.21447
17	-.39047
18	-1.20484

ErrSP= Errors in Speech Production.
ErrSP is derived from a one-factor Principal Component Analysis.

7.3.3.2 Multiple Regression Analyses

Assumptions for stepwise multiple regression analyses were met for NVA 55, NVA 70 and /aCa/. Hence, three stepwise multiple regression analyses were performed, with seven independent variables (**Table 7.6**) to gain insight in the contribution of each of those seven variables to the variance in four outcome measurements ((1) percentage correct at /aCa/ (ACA), (2) speech perception at 70 dB SPL (NVA70), and (3) speech perception at 55 dB SPL (NVA55). For the first analysis with /aCa/ as dependent variable, two significant predictors were found (ErrSP and age at first HA) and together accounted for 54% of the variance (adjusted R^2). For the remaining two analyses, ErrSP was the only significant predictor, accounting for 78% and 75% of the variance (adjusted R^2) for speech perception on NVA70 and NVA55, respectively. By adding any of the remaining 6 independent variables, no improvement of the explained variance was found. **Figure 7.6** shows scatter plots with the ErrSP on the X-axis and speech perception measurements NVA70 and NVA50 post-CI on the Y-axis, respectively. The solid line represents the linear regression line and the dashed lines represent the upper and lower limits of the 95% confidence interval.

Table 7.6
Predictors included in each of the multiple regression analyses.

1	ErrSP
2	PTA pre unaided CI side
3	PTA pre aided CI side
4	NVA70 pre aided CI
5	communication mode
6	age at first HA
7	congenital or non-congenital

ErrSP=Errors in Speech Production, PTA=Pure Tone Audiometry (0.5,1 and 2 kHz), HA=hearing aid, NVA70=speech perception (phonemescore) at 70 dB SPL.

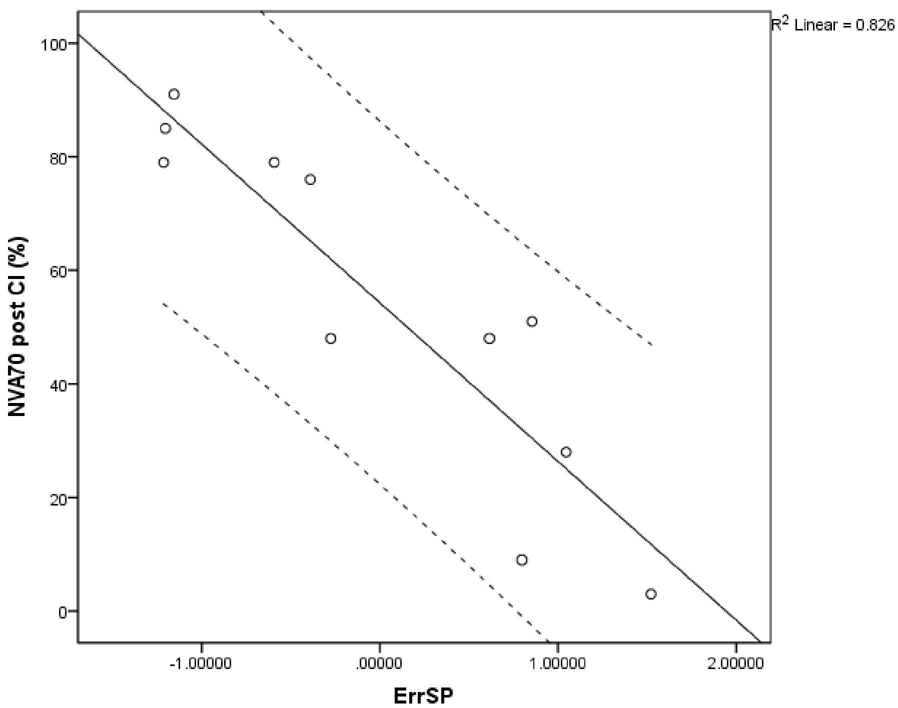

Figure 7.6a Scatter plot with the ErrSP on the X-axis and speech perception NVA70 (phonemescore at 70 dB SPL) at 12 months post-CI on the Y-axis (N=11). The solid line represents the linear regression line and the dashed lines represent the upper and lower limits of the 95% confidence interval.
ErrSP=Errors in Speech Production, CI=Cochlear Implant

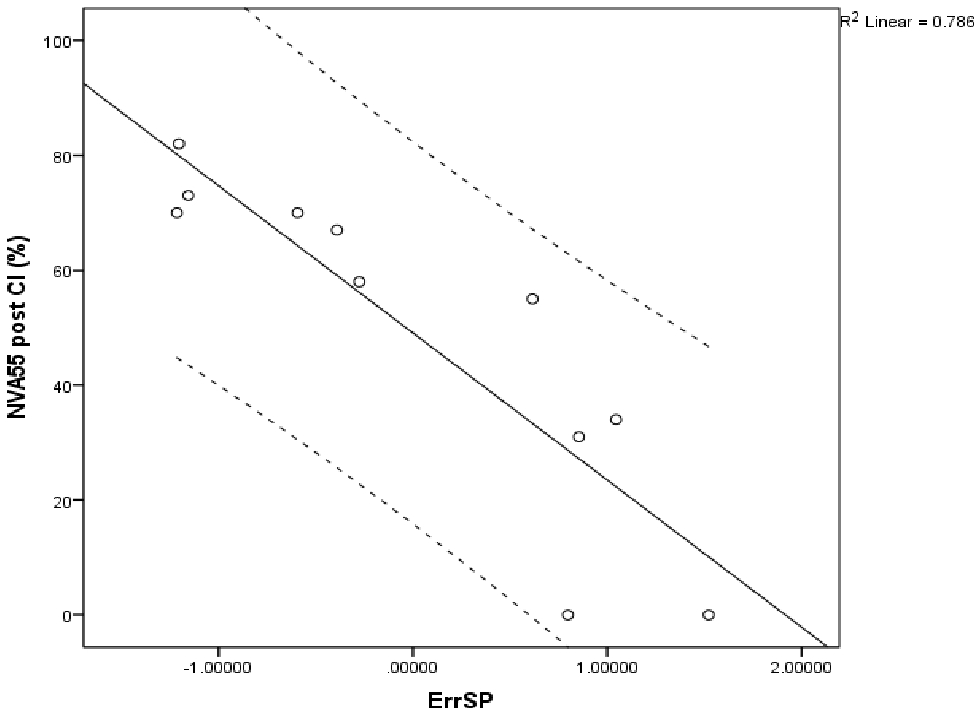

Figure 7.6b Scatter plot with the ErrSP on the X-axis and speech perception NVA55 (phonemescore at 55 dB SPL) at 12 months post-CI on the Y-axis (N=11). The solid line represents the linear regression line and the dashed lines represent the upper and lower limits of the 95% confidence interval.
ErrSP=Errors in Speech Production, CI=Cochlear Implant

7.3.3.3 Phoneme analysis perception-production

In order to make a more in-depth comparison between speech perception and speech production of consonants, relative averaged errors at phoneme level were gained (**Figure 7.7**). For the consonant perception, the /aCa/ test was analyzed and for consonant production relative averaged omissions, substitutions and distortions were taken into account. First, note that, except for /s/ and /z/, perception errors were higher than production errors. Second, the highest number of perception errors was made for the consonant /p/, while for production most errors were found for /z/. Third, for some perception-production pairs, a large discrepancy was found. For example, 65% of perceived /t/'s was wrongly identified, opposed to a 5% production error score.

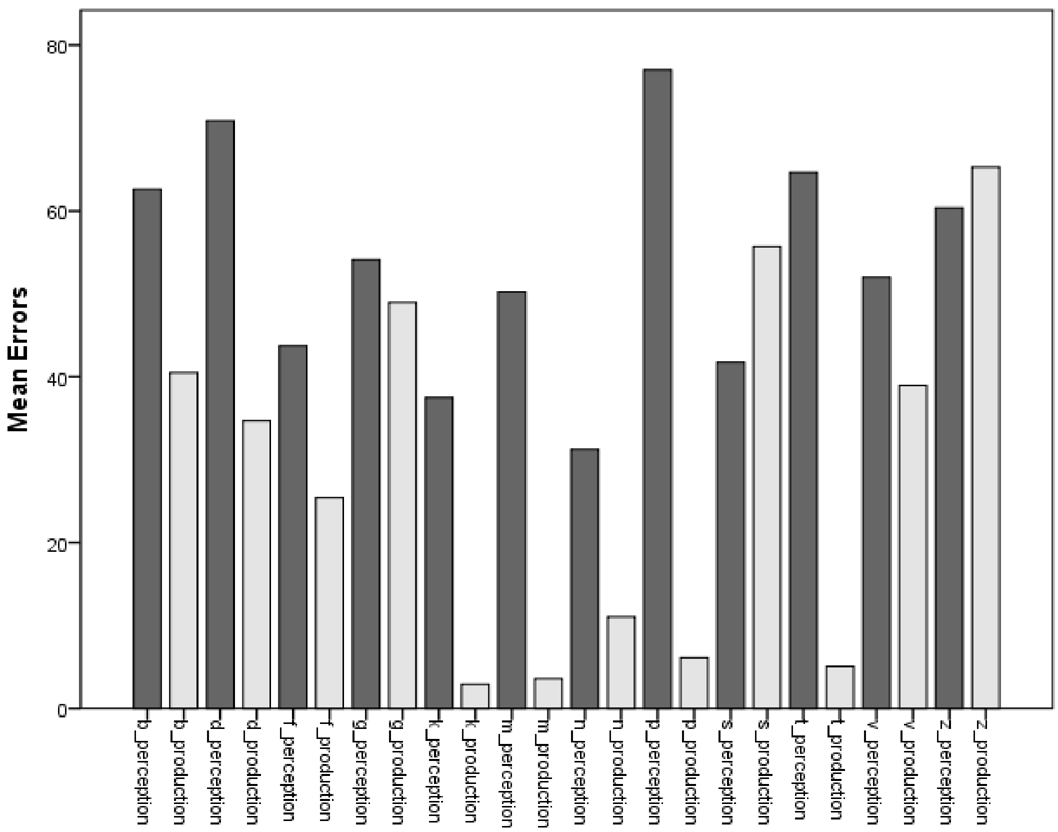

Figure 7.7 Bar charts of mean errors in speech perception (dark grey) and speech production (light grey) for each of the consonants.

7.4 Discussion

The first aim of this study was to investigate speech perception in early-deafened, late-implanted adults by means of a variety of speech perception material in quiet and in noise. Results revealed a statistically significant difference between pre- and postimplant scores on NVA70 and PTA. However, a large inter-subject variability was found, ranging from no perception up to excellent perception. This is in accordance with former studies reporting a wide range of performance (Schramm et al, 2002, Teoh et al, 2004a, 2004b, van Dijkhuizen et al, Caposecco et al, 2012). Nevertheless, on average, these postimplant performances are poorer compared to results of postlingually deafened adults (Schramm et al, 2002, Teoh et al, 2004a, 2004b, Santarelli et al, 2008). When the intensity of the speech perception test was decreased to 55 or 40 dB SPL, percentage correct scores were also reduced. Firszt et al (2004) performed a large-scale study to gain insight in speech recognition at soft to loud intensity levels. Results of these 78 subjects, of whom 10 were pre-linguistically deafened, on CVC word scores at 70, 60 and 50 dB SPL were on average 42%, 39% and 24% respectively (SD 23, 21 and 18 respectively). The mean score at 60 dB SPL of their pre-linguistic group was 46.07% (SD 26.94). This result lies between the scores obtained by our group at 70 dB SPL (53.05%, SD 33.82) and 55 dB SPL (43.82, SD 30.89).

Besides traditional PTA and speech audiometry, consonant identification was administered in a closed-set test (/aCa/) and on average 40.25% of the consonants were correctly identified. Santarelli and colleagues (2008) obtained 40.7% of correct responses after three years of implantation. Furthermore, relative information transmission scores for the consonants were obtained for the features of voicing, place of articulation, affrication, manner of articulation and nasality. Overall, results revealed that place cues were the least transmitted. Since relative information transmission scores had never been studied before in a group early-deafened, late-implanted adults, results were compared with studies including predominantly postlingually deafened adults. Van Wieringen and Wouters (1999) obtained ITr scores in 24 Laura implantees, of which 3 were prelingually deafened, with an average result of 33% correct (SD 13%). Philips et al (2013, unpublished data) used the /aCa/-test within a group of 20 postlingually deafened CI subjects and on average, 47.6% was correctly identified. Furthermore, in both studies within postlingually deafened subjects, place cues were least transmitted, which is in accordance with the results obtained in the present study.

The results of the speech perception in noise test (SRT_n) showed that on average, SRT was 69.17 dB (SD 2.99) implying that speech stimuli had to be 9.17 dB louder than the speech-weighted noise (presented at 60 dB SPL) to obtain 50% correct, whereas in quiet, SRT was 60.0 dB (SD 9.63). As for the /aCa/-test, SRT’s in early-deafened, late-implanted adults had never been reported before. Shpak and colleagues (2009) investigated sentence recognition in noise (SNR +10 dB) with the noise from the speech audiometer used within 20 prelingually deafened adolescents and results revealed 34% correct. Waltzman and colleagues (2002) obtained postimplantation 19.9% correct for CUNY sentences in noise scores for 14 congenitally deaf adults. However, it should be noted that some patients were unable perform this test (6/14 for SRT_q and 8/14 for SRT_n). The patients that were not able to perform this sentencetest (LIST) in quiet, also obtained very low scores on the CVC test (< 20% correct at 70 dB SPL (phonemescore), except for patient number 7 (52% correct at 70 dBSPL (phonemescore). In this particular patient, Noonan syndrome has been diagnosed, and she suffers from a mild mental retardation. Possibly, this sentence task might have been to complicated for her. Two patients, 4 and 6, were able to perform the test in quiet, but failed when background noise was presented. Possibly, lowering the level of the background noise, e.g. to 50 dB SPL, might enhance this test for them.

Overall, early-deafened, late-implanted adults progress beyond their preimplant state on sound detection and consonant identification, whereas results on more demanding measurements (words and sentences in noise) are limited. It has been shown by means of positron emission tomography (PET) that the primary auditory cortex of prelingually deafened adults remains responsive to intracochlear stimulation, while speech activation of the secondary auditory cortex is poorer compared to normal-hearing or postlingually deaf subjects (Truy et al, 1995, Naito et al, 1997). Hence, Teoh and colleagues (2004b) suggest that although the auditory pathways to the primary auditory cortex remain functional for a long period after auditory deprivation, the auditory input may not be interpreted as meaningful sound by some CI subjects, possibly because of their inability to process the incoming signals in the appropriate higher-order speech and language centers.

The second aim of the study was to gain insight in the relationship between errors made in speech production and speech perception. By means of a single factor, ErrSP (Errors in Speech Production), 78% of the variance of postimplantation speech perception at NVA70 can be explained. Hence, participants with fewer errors in speech production achieve better scores on speech perception. Multiple regression analyses performed in former studies could never explain such a large portion of the variance. Caposecco and colleagues (2012) included three factors (oral communication in childhood, progressive HL and no time without a HA on the implant ear) that - together - accounted for 63% of the variance on open-set sentence scores, whereas van Dijkhuizen and colleagues (2011) by implementing speech intelligibility as a predictive factor could explain 62% of the variance on open-set CVC words.

Using ErrSP as a predictor will facilitate counseling and candidate selection in this particular patient population since this factor does not depend on patients self-reported factors such as age at onset of deafness or age at first HA. These factors are therefore not very effective as a selection criteria for cochlear implantation. In accordance with van Dijkhuizen et al (2011) we discard early age at onset of deafness as an exclusion criteria for cochlear implantation. With respect to communication mode, former studies have shown that subjects using oral communication in childhood were more likely to obtain better speech perception results compared to those using total communication or sign language (Kaplan et al, 2003, Kos et al, 2009, Caposecco et al, 2012). None of the subjects in the present study exclusively communicated through sign language and indeed, a significant correlation between communication mode and speech perception revealed better scores for subjects using oral communication. Moreover, subjects using oral communication achieved lower ErrSP scores implying less errors in speech production. Hence, one might suggest that the development of speech containing few errors is dependent on the usage of oral communication. However, offering oral communication is not the only factor contributing to good speech production. The success also depends on the availability of sufficient auditory input (van Dijkhuizen et al 2011) and the absence of colonization of the auditory cortex by other sensory modalities (Teoh et al, 2004b).

The comparison of speech perception and speech production of consonants revealed that overall, perception errors were more common than production errors, except for /s/ and /z/. It should be noted that the speech perception task (closed-set /aCa/) differed from the speech production task (open-set picture-naming task). In future, it might be interesting to repeat the /aCa/ task for both speech perception and speech production.

A limitation of the present study was the usage of speech production measurements 12 months postimplantation. Including the preimplantation speech production data might have been more precise. Since improving speech perception is the primary focus within speech therapy of early-deafened, late-implanted adults postimplantation, speech production capabilities postimplantation are equivalent to their preimplantation levels (Wong, 2007).

7.5 Conclusions

Overall, the findings from this study indicate that, on average, early-deafened, late-implanted adults progress beyond their preimplant state for speech perception measurements that do not require high-level auditory mechanisms such as PTA and consonant identification whereas for more demanding measures such as speech perception in noise, results are poorer. Secondly, errors in speech production can be used as a predictor of speech perception outcome postimplantation in quiet and in noise for early-deafened, late-implanted adults. Using errors in speech production as a predictor for speech perception postimplantation will facilitate candidate counseling and selection in the group of early-deafened adult CI candidates.

Chapter 8

Characteristics and determinants of music appreciation in adult CI users

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Abstract

Objectives

The main objective of this study was to assess the associations between self-reported listening habits and perception of music and speech perception outcomes in quiet and noise for both unilateral CI users and bimodal (CI in one ear, HA in contralateral ear) users.

Methods

Forty postlingually deaf CI recipients filled in the questionnaire. Furthermore, audiological data of 15 CI recipients were gathered and compared with music appraisal data.

Results

Information concerning music appreciation was gathered by means of a newly developed questionnaire. Moreover, audiological data (pure-tone audiometry, speech tests in noise and quiet) were gathered and the relationship between speech perception and music appreciation is studied. Bimodal users enjoy listening to music more in comparison with unilateral CI users. Also, music training within rehabilitation is still uncommon, while CI recipients believe that music training might be helpful to maximize their potential with current CI technology.

Conclusions

Music training should not be exclusively reserved for the good speech performers. Therefore, a music training program (MTP) that consists of different difficulty levels should be developed. Hopefully, early implementation of MTP in rehabilitation programs can enable adult CI users to enjoy and appreciate music and to maximize their potential with commercially available technology. Furthermore, because bimodal users consider the bimodal stimulation to be the most enjoyable way to listen to music, CI users with residual hearing in the contralateral ear should be encouraged to continue wearing their HA in that ear.

8.1 Introduction

The CI is an assistive hearing device designed for individuals with a severe to profound bilateral SNHL who receive little or no benefit from conventional HAs. Nowadays, modern CI systems are able to provide good speech understanding, at least in favorable listening conditions. In recent years, many CI recipients express hopes of enjoying music following cochlear implantation. However, while a CI is successful in delivering speech in quiet, its performance in delivering music is less than ideal. Because the perception of music requires a good perception of four basic perceptual attributes (pitch, rhythm, timbre and melody) most researchers have measured aspects of CI music perception using a variety of tasks that measures (a combination of) these four basic perceptual attributes (Drennan and Rubinstein, 2008).

The periodicity element of pitch is particularly necessary when listening to complex tones. The basis of good perception of complex-tone pitch lies in the repetition rate, which depends on fine-structural temporal encoding (Drennan and Rubinstein, 2008). CI recipients rely mostly on perceiving temporal envelopes at specific places and not on temporal fine-structure. By using pitch-based tests, such as pitch discrimination, pitch ranking and pitch perception, several researchers found significant lower results on pitch-based tests for CI users compared to normal hearing (NH) subjects (Fujita and Ito, 1999, Gfeller et al, 1997, Zeng, 2002, Galvin et al, 2007, Gfeller et al, 2007).

It is known that music perception in electric hearing is dominated by the rhythmic patterns rather than pitch information. In addition, some CI listeners can use low-frequency temporal information to extract pitch information. Zeng (2002) demonstrated perception of temporal pitch in CI users for stimulation rates up to 300 Hz.

Results of simple rhythm-based tests such as gap detection and amplitude modulations revealed fairly good rhythm perception in adult CI users (Collins et al, 1994, Gfeller et al, 1997, Gfeller et al, 2000, Zeng, 2002). However, the ability to process more complex rhythmic discrimination (Collins et al, 1994, Gfeller et al, 1997, Kong et al, 2004) is subject to large inter-subject differences (Leal et al, 2003, McDermott, 2004). Thus, rhythm discrimination is good but on average not as good as in NH listeners.

The third basic aspect, timbre, can be measured by timbre perception or musical instrument identification (Gfeller et al, 2002, McDermott, 2004). Timbre is encode by the temporal envelope and by the spectral shape of sound. While temporal envelopes are fairly well preserved in CI processing, the spectral information is reduced relative to NH listeners implying a better than chance but not nearly as good timbre recognition for CI users compared to NH users.

Melody perception is the fourth basic perceptual attribute. Several researchers have gained insight in melody perception by using familiar melody recognition/identification (Collins et al, 1994, Fujita and Ito, 1999, Gfeller et al, 2002, Zeng, 2002, Leal et al, 2003, Kong et al, 2004, 2005, Singh et al, 2009). Because using familiar melody recognition tasks in CI music perception research presents a few problems such as the reliance on a CI user's memory, Cooper et al (2008) used the Montreal Battery for Evaluation of Amusia (Peretz et al, 2003) to gain insight in musical abilities of CI users. Nevertheless, all the above mentioned studies revealed that melody perception in CI users is generally extremely poor.

Overall results suggest that CI users, as NH listeners, show a typical pattern of music perception in which scores on temporal aspects of music are higher than on spectral aspects of music.

The deconstruction of music into its four basic perceptual attributes is a useful approach to gain insight in music perception in CI users. It allows researchers to determine basic aspects of musical perception and to identify those areas in which CI users show great difficulty. However, this type of deconstruction is also artificial and far-removed from the challenges faced by CI recipients when listening to music. It is well known that music is among the most complex categories of acoustic sounds that are commonly heard in life, lacking the redundancy of speech, and the contextual aspects of conversation (Limb, 2006). Furthermore, subjective appreciation of music is not necessarily reflected by the perceptual accuracy exhibited in laboratory conditions, since it involves personal, situational, cultural and emotional variables. Moreover, recent research has shown that factors can contribute differentially to the variability among adult CI users concerning music perception and music appraisal (Gfeller et al, 2008). Hence, both the accuracy of music perception and the degree of music appraisal should be accounted for as distinguishable indices of CI outcome.

Therefore, different measurements should be used to gain insight in accuracy of music perception and the degree of music appreciation. The majority of previous researchers has tended to focus on music perception, rather than on music appreciation in adult CI users (Leal et al, 2003, Kong et al, 2004, Gfeller et al, 2005, 2007, Cooper et al, 2008, Looi et al, 2008, Nimmons et al, 2008, Spahr et al, 2008, Spitzer et al, 2008, Singh et al, 2009). A smaller number of studies focused on music appraisal or enjoyment by means of self-reported questionnaires or testing appraisal of musical excerpts (Gfeller et al, 2000, 2008, 2010, Mirza et al, 2003, Brockmeier et al, 2007, Lassaletta et al, 2007, Looi and She, 2010, El Fata et al, 2009). These studies have all shown a substantial inter-subject variability among CI recipients for music appraisal or enjoyment. Understanding the factors that contribute to the variability among CI recipients with respect to music appreciation could have important implications for implant design as well as advising CI users with respect to musical possibilities and benefits.

Therefore, the objective of this study was to assess the associations between self-reported listening habits and perception of music and speech perception outcomes in quiet and noise for both unilateral CI users and bimodal (CI+HA) users.

8.2. Materials and methods

Participants were recruited from the hospital database. All were current CI recipients with a postlingually acquired HL, had used their CI for longer than six months, were native Dutch speakers and had no major intellectual impairments. Questionnaires were posted out by the clinic to all of the patients who met the above-mentioned criteria. In total 57 questionnaires were posted. A coding system was developed to ensure the anonymity of the respondents from the researchers.

Patients were able to choose whether they wanted (1) to fill in the questionnaire, or (2) to fill in the questionnaire and come over to the clinic to participate in audiological tests. Of the 57 questionnaires posted, 40 replies (70%) were received. Furthermore, 15 out of these 40 patients were willing to participate in audiological tests.

8.2.1 Subjects

Data were collected from 40 postlingually deaf adults (23 female, 17 male, mean age 64 yrs SD 12.5 yrs, range 37-84 yrs) who were implanted at our university hospital. A total of 19 patients use a Cochlear device (18 Nucleus Freedom, 1 Esprit 3G), 16 patients use an Advanced Bionics device (9 Harmony, 7 Auria) and 5 patients use a Digisonic BTE device. Furthermore, 13 recipients wear a contralateral HA. This group is referred to as the bimodal group (**Table 8.1**).

Table 8.1
Demographic data of the CI recipients.

	CI-only group	Bimodal group
Number of participants	27	13
Gender	12F	11F
	15M	2M
Age at testing	62.03yr (SD: 12.37)	68.52yr (SD: 12.16)
Duration of deafness	4.46yr (SD: 2.40)	4.46yr (SD: 2.05)
Length of experience with CI	5.41yr (SD: 3.27)	3.51yr (SD: 2.17)
Type of CI	14 Cochlear	5 Cochlear
	8 Advanced Bionics	8 Advanced Bionics
	5 Digisonic	0 Digisonic
Speech coding strategy	1 SPEAK	5 ACE
	11 ACE	5 HiRes
	8 HiRes	3 HiRes120
	2 MP3000	
	5 MPIS	

CI=cochlear implant, F=female, M=male, SD=standard deviation, yr=years

The study was approved by the Ethical Committee of the Ghent University Hospital and was conducted in accordance with the ethical standard stipulated in the Helsinki declaration (2000) for research involving human subjects.

8.2.2 Protocol

8.2.2.1 Questionnaire

A Dutch questionnaire to gain insight in music appreciation was developed based on literature and clinical experience (Gfeller et al, 2000, 2008, Mirza et al, 2003, Looi et al, 2008, Lassaletta et al, 2008) (Appendix 1). Patients were asked a number of questions regarding their music listening habits before becoming deaf and after cochlear implantation. A draft of the expanded questionnaire was given to two audiologists who evaluated the document for clarity, appropriateness, and comprehensiveness of the items. A second draft was completed and reviewed again by two audiologists. Furthermore these questions were given to two CI recipients in structured interview. The data of these two recipients were not included in the final study. The final version consists of 60 multiple choice questions and Likert-type scales (1 point for strong disagreement and 5 points for strong agreement) about 4 items, namely (1) speech perception and listening habits (22 questions), (2) musical background (6 questions), (3) music appreciation (25 questions) and (4) contralateral HA and rehabilitation (7 questions).

8.2.2.2. Audiological tests

Audiological tests were performed in a sound booth, with speech and noise materials presented at 0° azimuth at 1 meter distance from the CI recipients. Tests were performed with the recipients own speech processor or HA at their daily program. Pure tone audiometry was performed both unaided (under headphones, using no hearing devices) and aided (free field, using their CI or HA separately) for the frequencies ranging from 0.125 up to 8 kHz. Speech perception data were collected in the aided condition (CI or CI and HA separately for bimodal users), based on percent-correct performance at phoneme level and on speech reception threshold (SRT) (Plomp and Mimpen, 1979). Percent-correct performance at phoneme level was measured using Dutch recorded CNC 3 phoneme monosyllabic words, spoken by a male speaker (NVA) (Wouters et al, 1994). Speech perception was measured in quiet at 70, 55 and 40 dB SPL. For each condition a list of 11 words was used, but scoring was based on percent-correct performance at phoneme level. This test was also conducted in noise. Therefore the steady NVA speech-weighted noise with the same long term-spectrum as the presented speech material was used. Two SNRs were tested, i.e. +5 dB and 0 dB at a noise level of 60 dB SPL (i.e. the average of 65 dB SPL and 60 dB SPL). Furthermore, SRTs were measured adaptively both in quiet and in noise by means of the LIST sentences, spoken by a female speaker (van Wieringen and Wouters, 2008). The SRT is the sound level of the speech signal at which 50% of the presented speech sentences is correctly identified. The SRT was determined using a simple up-down (1 up, 1 down) procedure with a 2-dB step size. The first sentence was repeated with increasing level until it was correctly identified by the subject. The level of subsequent sentences was presented once at a 2 dB lower or higher level than the previous sentence, depending on whether the latter was identified correctly or not. A response was judged correct if and only if all key words of the sentence were repeated correctly. The adaptive procedure continued until 10 reversals occurred, and then the speech levels of sentences 6 to 10 and the imaginary 11th sentence were averaged to obtain the SRT value. A lower SRT implies that the listener can understand speech in more adverse conditions. Two lists were used, one for obtaining an SRT score in quiet (SRT_q) and one for obtaining an SRT score in noise (SRT_n). In the latter procedure, the speech level was varied and the speech-weighted noise available for the LIST was kept constant at 60 dB SPL.

8.3 Results

Data were analyzed using SPSS version 15 (Chicago, IL). For all analyses the level of significance was set at 0.05.

8.3.1 Questionnaire

8.3.1.1 Section 1: speech perception and listening habits

Most CI users report a good speech understanding in quiet (93% can participate in a conversation with 2 or 3 people), but in more noisier conditions, speech perception drops (68% indicates having trouble to understand a salesman in a shop, 74% declares having trouble understanding a conversation at a reception). With respect to being able to differentiate incoming sound with the CI, 59% states not being able to differentiate a bus from a car and when it comes to voices, only 53% indicates being able to recognize a female voice from a male voice. Furthermore, in 59% of cases, family members are not recognized solely by their voice. With subtitles, 63% of implant users are able to understand the news on television, without subtitles this percentage drops to a mere 28%.

8.3.1.2 Section 2: musical background

Before implantation, 43% of the participants listened regularly to music. After implantation this number decreases considerably to only 13%. Although 50% of participants reports that they enjoyed listening to music prior to implantation, only 13% of the participants remains enthusiastic after implantation. Concerning particular styles of music, popular songs are preferred both prior and after implantation, no statistical difference was found ($L\chi^2$ (1) = 1.58; p = .209).

Most CI users have a limited musical background: only 20% has followed musical lessons, 13% has sung in a choir and 18% played a musical instrument prior to CI. After CI only 3% of the group continues playing a musical instrument.

8.3.1.3 Section 3: music appreciation

Only 28% of the CI recipients can appreciate music with the CI and 82% states that music does not sound naturally with the CI. Following the rhythm is easier (35%) than following the melody (15%) for our recipients. Those patients who do find that music sounds natural through their CI state that they can perceive melody ($L\chi^2$ (1) = 7.76; p = .005) and rhythm ($L\chi^2$ (1) = 8.73; p = .003) within music and vice versa. Three months after switch-on, 20% describes listening to music, but 42% states that lyrics are intelligible only after 12 months. No statistical significant difference was found between experienced and non-experienced music listeners with respect to likeliness of the sound of music through a CI ($L\chi^2$ (1) = .113; p = .736). Fifty-two percent indicates that being able to enjoy music is important and 35% answers that music is significant for their social life. If available, 68% of the respondents would opt for a CI that could transmit music perfectly.

8.3.1.4 Section 4: rehabilitation and contralateral hearing aid

Thirty-three percent reports having received a form of musical training during rehabilitation and 45% states practicing at home. Patients who train at home, do find it easier to follow the rhythm than patients who do not practice at home ($L\chi^2$ (1) = 8.00; p = .005). However, no significant difference was found for easily following melody between patients who do or do not train at home ($L\chi^2$ (1) = 1.07; p = .300). Moreover, 65% is convinced that learning to listen to music during rehabilitation is useful.

Half of the respondents favors listening to music in a quiet environment or even alone (68%). In answer to the question ‘What do you think about a specific music program on your speech processor’, answers are divers (**Figure 8.1**).

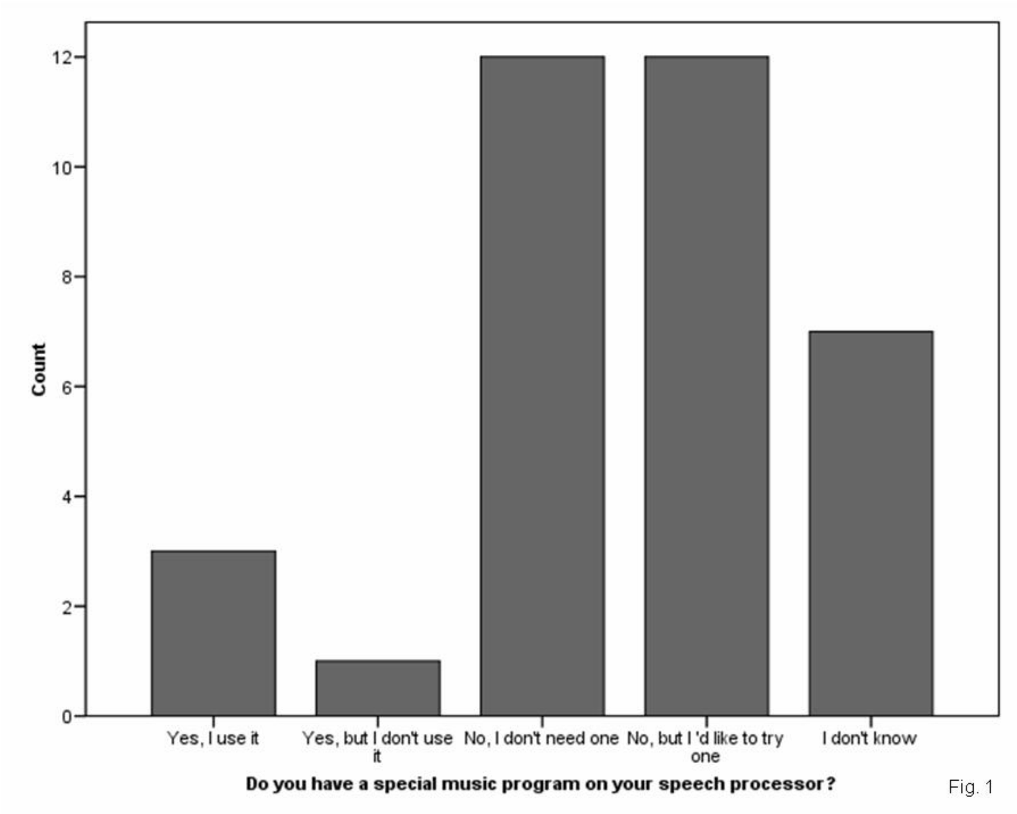

Figure 8.1 The x-axis describes the various answers concerning a special music program on CI recipients' speech processor; the y-axis is the number of 'yes' answers for the CI recipients.

Seventy percent of the bimodal listeners point out that their HA is helpful when listening to music. Furthermore, a statistical significant difference was seen between bimodal users and CI-only users for ease in following melody in favor of bimodal listeners ($L\chi^2$ (1) = 10.57; p = .001), but not for rhythm ($L\chi^2$ (1) = .06; p = .809) (**Figure 8.2**). Nevertheless, bimodal listeners in the CI-only condition, can less appreciate the sound of music through their CI compared to CI-only users ($L\chi^2$ (4) = 6.189 , p=.026). Also, more CI-only users (88%) point out the need for a speech processor able to better transmit music compared to bimodal users (54%) ($L\chi^2$ (1) = 4.39; p = .036). However, both groups believe that learning to listen to music in rehabilitation might be useful ($L\chi^2$ (1) = .68; p = .409).

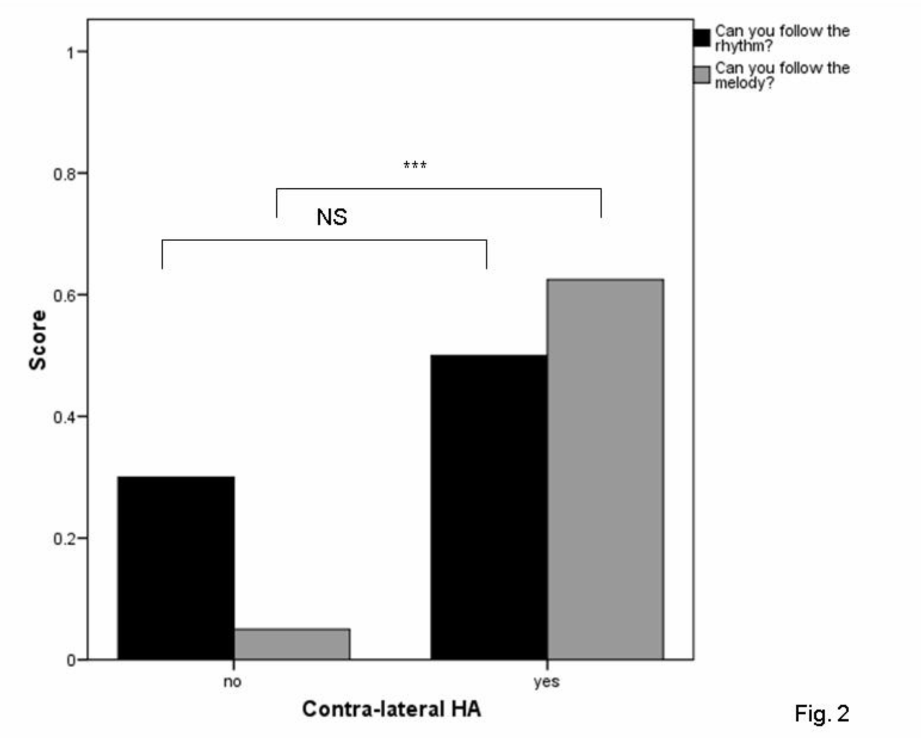

Figure 8.2 CI recipients wearing a contralateral HA (‘yes’) (bimodal group, N = 13) versus CI recipients who do not wear a contralateral HA (‘no’, N = 27) are depicted on the x-axis. The y-axis is the number of ‘yes’ answers for the CI recipients.
HA = hearing aid, NS = Not Significant, *** p< 0.001

8.3.2 Audiological tests

Fifteen patients (7 female, 8 male; mean age 63 yrs; SD 10.5 yrs; range 44-77 yrs) underwent audiological tests.

Pure-tone audiograms were performed both with and without prosthetic devices and good results (implying results for all frequencies < 30 dB HL) with their CI. Speech perception results showed a good speech perception in quiet, especially at higher intensity levels (**Figure 8.3**). As expected, speech perception scores decrease in less favorable conditions (**Figure 8.4 and 8.5**).

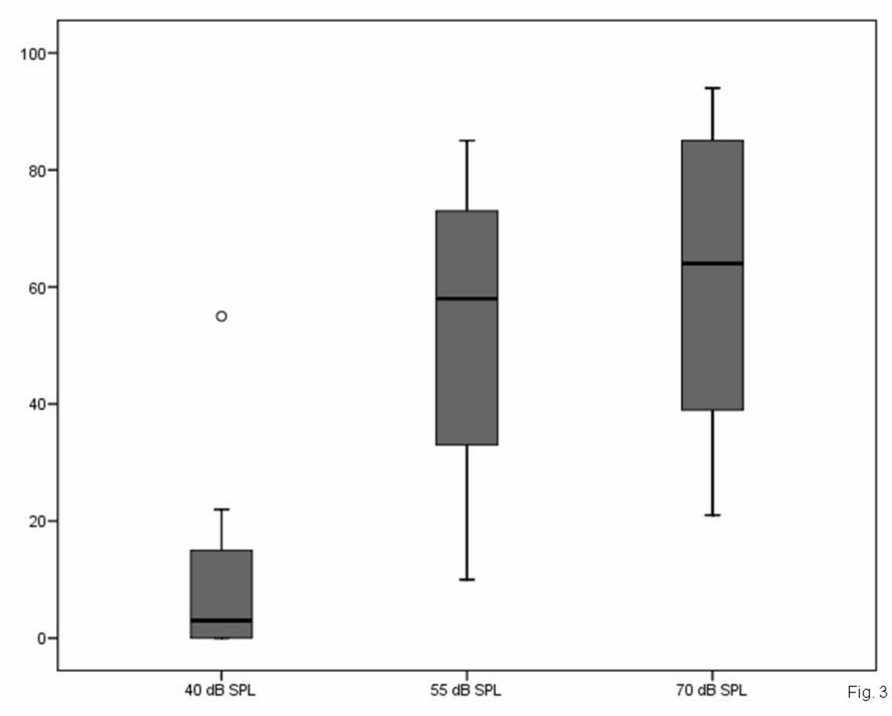

Figure 8.3 The x-axis describes the intensity levels at which speech testing is performed (NVA); the y-axis is the percentage correct obtained (phoneme score).

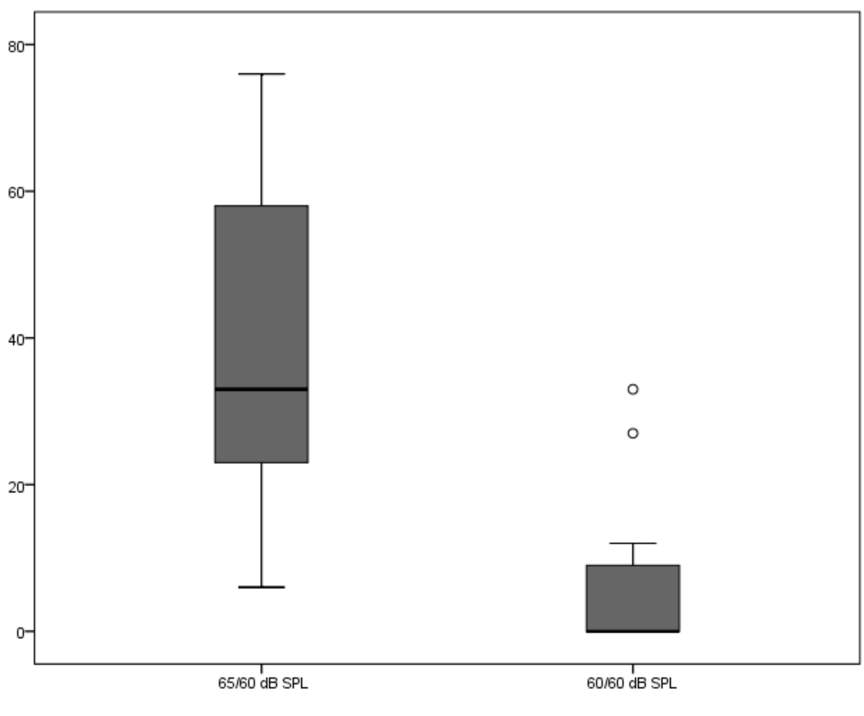

Figure 8.4 The x-axis describes the intensity levels at which speech testing is performed (NVA), speech level/noise level; the y-axis is the percentage correct obtained (phoneme score).

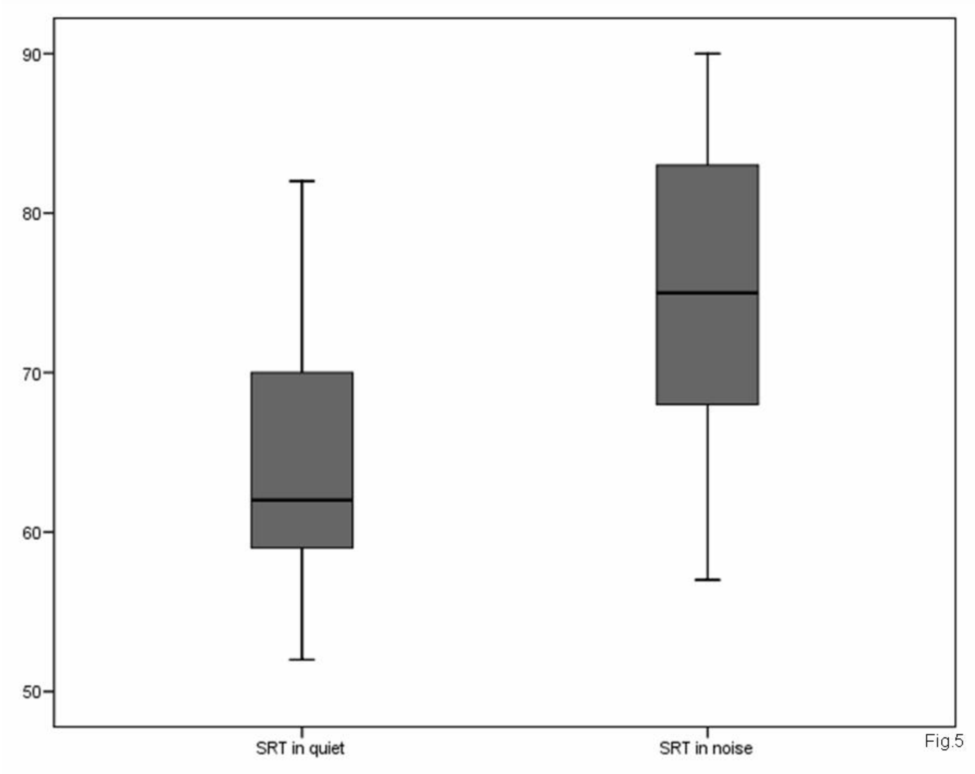

Figure 8.5. The x-axis describes the speech tests performed (LIST), both in quiet and in noise; the y-axis is the SRT-score (dB).
SRT= Speech Reception Threshold.

No differences were noticed with respect to speech perception scores, nor for SRT_q (F(1,14) = .023, p = .907), nor for SRT_n (F(1,14) = .106, p = 751) for the CI-only condition in both bimodal and CI-only groups. Patients with experience in listening to music prior to CI, do not have a better SRT_q (F(1,12) = .005, p= .953) nor SRT_n (F(1,12) = 1.236, p = .399).

A statistically significant correlation was found for both enjoyment of listening to music with CI and finding music with the CI sounding more natural and SRT_q (p = .001; p= .001) and for SRT_n (p = .035; p = .030), in a way that patients with good SRT_q and SRT_n enjoy listening to music through their CI more and find it sounding more natural in comparison with recipients having worse SRT scores (**Figure 8.6 and 8.7**). Additionally, no statistical significant difference was found between good and poor performers on SRT_q and SRT_n with respect to finding it easy to recognize melody, nor rhythm.

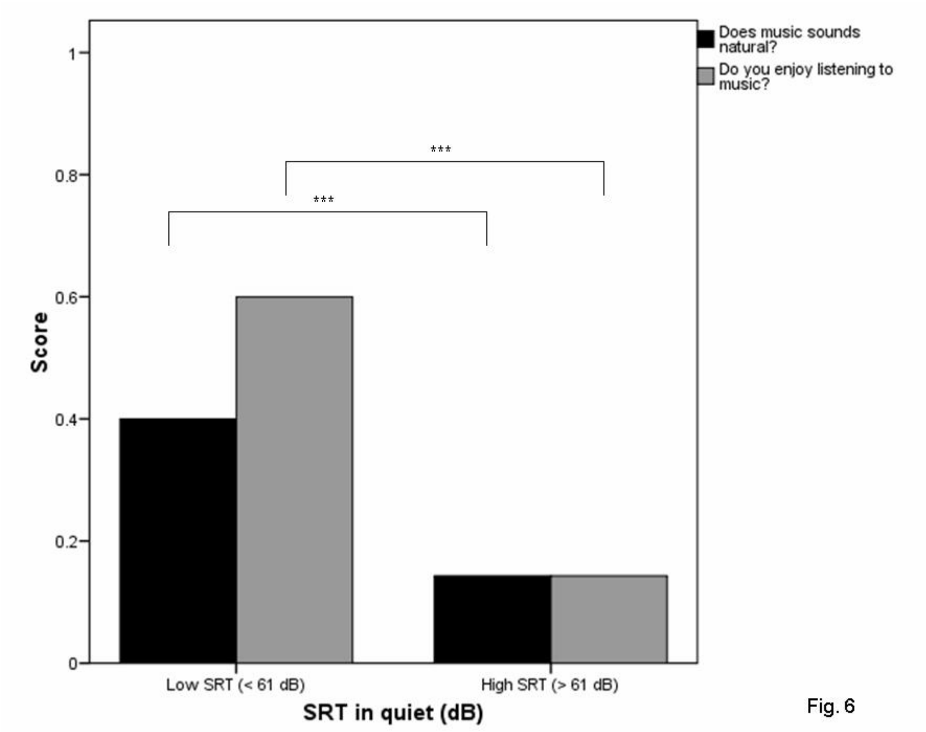

Figure 8.6 CI recipients with good SRT scores (meaning a SRT score lower than 61 dB) and CI recipients with poor SRT scores (meaning a SRT score higher than 61 dB) in quiet are depicted on the x-axis. The y-axis is the number of 'yes' answers for the CI recipients.
SRT = Speech Reception Threshold, *** p< 0.001

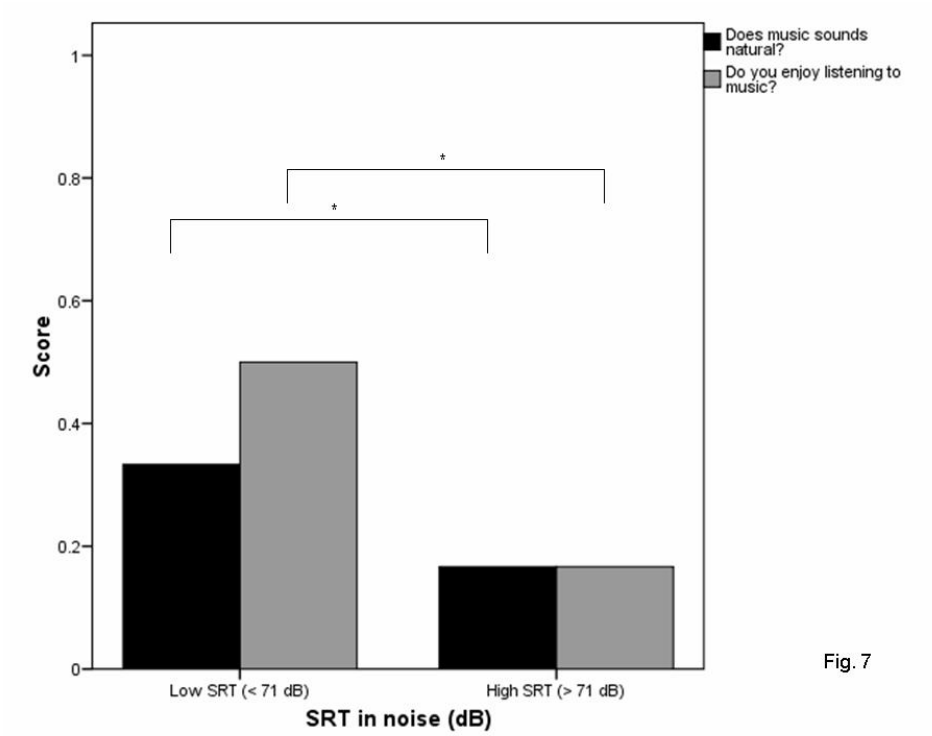

Figure 8.7 CI recipients with good SRT scores (meaning a SRT score lower than 71 dB) and CI recipients with poor SRT scores (meaning a SRT score higher than 71 dB) in noise are depicted on the x-axis. The y-axis is the number of 'yes' answers for the CI recipients.
SRT = Speech Reception Threshold, * p< 0.05

Nevertheless, not only patients with good SRT_q and SRT_n describe that music plays an important role in their lives, both with respect to their own well-being (F(1, 13) .270, p = .621) as with respect to their social life (F(1,13) = .025, p = .878). Furthermore, both good and poor performers emphasize the need of implementing music into rehabilitation (F(1,13) = .370, p = .554).

8.4 Discussion

8.4.1 Music after CI

In recent years, research has tend to focus on implant users' perception of non-speech sounds, especially music. Multiple studies have shown that CI recipients experience more difficulties in perceiving music than NH subjects or HA recipients (Kong et al, 2004, Gfeller et al, 2005, Cooper et al, 2008, Looi et al, 2007, 2008). Since music perception skills do not reflect music appraisal or enjoyment, some researchers have addressed this topic by means of questionnaires (Gfeller et al, 2000, Mirza et al, 2003, Lassaletta et al, 2007, 2008, Migirov et al, 2009, Veekmans et al, 2009, Looi and She, 2010). Not surprisingly, Gfeller and colleagues (2000), Lassaletta and colleagues (2008) and Looi and She (2010) report that postimplantation, people enjoy music less compared to before receiving their CI, which is confirmed in our study. Additionally, the extent to which music listening is satisfactory after implantation varies considerably among recipients. Also in accordance with Gfeller and colleagues (2000), results showed that general enjoyment of music was positively correlated with time spent in listening to music postimplantation. It remains unclear, however, whether more practice to music enhances listening to music or whether those who like listening to music are more likely to spend time in music listening.

A strong correlation was found between music enjoyment and speech perception scores, both in quiet and in noise, implying that recipients with better speech scores also report higher levels of music enjoyment and naturalness of music through their CI. These results are in accordance with the data from Looi and She (2010).

8.4.2 Music for special groups (bimodal and experienced listeners)

Bimodal listeners believe that the use of a contralateral HA enhances music listening, especially with respect to perceiving the melody. This finding corresponds to the results of

Gfeller and colleagues (2008, 2010) and Looi and She (2010). Also, in the CI-only condition, bimodal users did find music sounding less natural then in the bimodal condition and thus implying that the use of a HA enhances the naturalness of the perceived sound. Moreover, CI-only users emphasize the need of the development of new speech-coding strategies enabling them to improve listening to music, while bimodal listeners do not. These findings are consistent with other studies which have demonstrated the benefits of bimodal listening over the CI-only condition for a range of listening tasks (Leal et al, 2003, Kong et al, 2004, El Fata et al, 2009). Consequently, the importance of preserving and, if necessary, enhancing residual hearing, either in the contralateral ear by means of a HA or in the same ear by means of electroacoustic stimulation (EAS) is underlined. In patients with absence of usable acoustic hearing in the non-implanted ear, bilateral cochlear implantation (BICI) might be considered (Veekmans et al, 2009).

With respect to music experience before implantation, it was found that experienced listeners do not find music through their speech processor sounding worse in comparison with non-experienced listeners, neither did they had better speech perception scores.

8.4.3 Potential in rehabilitation

While the majority of published studies show that CI recipients are less accurate than NH and HA users in music perception tasks, some elements of music perception and appraisal can be enhanced by training and by increasing listening to music (Gfeller et al, 2010). Moreover, both bimodal and CI-only users believe music appreciation might be improved by training. Results showed that recipients with better speech perception scores in quiet and in noise report higher levels of music enjoyment and naturalness of music through their CI. This corresponds to other studies on music appraisal (Looi and She, 2010), but differs from studies who focus on correlating objective music perception tests and speech perception tests (Singh et al, 2009), and thereby hypothesize that music perception and speech recognition do involve two different processes. The present study does not aim to refute the above-mentioned hypothesis, but instead shows a shortcoming in current rehabilitation programs. Indeed, only 33% of the CI recipients states receiving, or having received some music training during rehabilitation. In other words, actual rehabilitation programs concentrate mainly on the optimization of speech perception. Only recipients who perform well in speech perception tasks (good performers), will move on to the next level within rehabilitation: listening to music. Because music is offered during therapy, these patients gradually become acquainted with the new input and musical appraisal might be enhanced. Consequently, good speech performers state having a higher level of enjoyment when listening to music in comparison with poorer speech performers. Furthermore, Gfeller et al (2008) found that no significant improvements in music appraisal can be established as a result of incidental exposure to music in everyday life as opposed to increasing scores on speech perception tasks over a 4 to 5 year period. Thus, improving music appraisal will be feasible only by implementing learning to listen to music within therapy.

8.5 Conclusions

These findings support the introduction of a music training program (MTP) in an early stage of rehabilitation. This early introduction might enhance enjoyment of music for both good and poor CI recipients in terms of speech perception.

Looi and She (2010) already listed some aspects that should be taken into account when designing a music training program for CI users (eg. low-frequency instruments, individual instruments). Nevertheless, music training should not be exclusively reserved for the good speech performers, since it is well known that music training can be a valuable aid in improving listeners' perception of language. Therefore, a MTP that consists of different difficulty levels should be developed. Hopefully, early implementation of MTP in rehabilitation programs can enable adult CI users to enjoy and appreciate music and to maximize their potential with commercially available technology.

Furthermore, results showed that bimodal users consider the bimodal stimulation to be the most enjoyable way to listen to music. Therefore, CI users with residual hearing in the contralateral ear should be encouraged to continue wearing their HA in that ear. El Fata et al (2009) proposed that a limit of 80 dB HL (0.125 kHz up to 1 kHz) would be useful for counseling patients on the likely benefit of bimodal listening to music. Within music therapy, these bimodal users should be wearing both devices en and not only their CI, which is often the case in speech therapy.

General discussion

Chapter 9

Discussion and future perspectives

9.1 Introduction

The CI has created a paradigm shift in the treatment of bilateral deafness. In less than thirty years, the CI progressed from a first attempt to elicit hearing via direct electrical stimulation of the auditory nerve to a commercially available device that has restored varying degrees of HL to tens of thousands of HI patients (Eisen, 2006). The earliest devices yielded benefits in the form of improved lip-reading, voice monitoring, awareness of environmental sounds, and closed-sets speech understanding. Present technology enables many implanted patients to achieve open-set speech understanding without visual cues. However, CI outcomes are characterized by wide inter-subject variability for both children and adults. Eisenberg states that the ability to understand and to reduce this variability remains a goal for researchers, implant manufacturers, and clinicians (Eisenberg, 2009).

Within this thesis speech perception outcomes of a contemporary group of pediatric and adult CI users were obtained. Furthermore, possible predictive variables that could administer a CI recipient’s speech perception outcome, were investigated.

9.2 Speech perception outcomes in the pediatric CI population

9.2.1 Impact of a universal newborn hearing screening program

A universal newborn hearing screening program (UNHSP) was implemented in Flanders from 1998 onwards and replaced the behavioral Ewing-screening performed at the child’s age of 9 months. Hence, Flanders was the first region in Europe that aimed to **diagnose hearing impairment in newborns before the age of 3 months** (Van Kerschaver et al, 2007).

The impact of this UNHSP on the outcome of HI children, was investigated in a multi-center study of 391 implanted children, out of which 195 were early screened (**Chapter 4**). This study revealed that, on average, early screened congenitally deaf children were detected by the age of 3 months as opposed to their late screened peers, who were on average 12 months old. This revelation has led to earlier clinical intervention and a steadily decreasing age of cochlear implantation. The study also observed that early screening and implantation were associated with better auditory receptive skills and speech intelligibility, which has been confirmed in several other large-scale studies (e.g. Svirsky et al, 2004). Further, research shows that early implanted children achieve significantly better language (e.g.

Artières et al, 2009, Boons et al, 2012), social-emotional (e.g. Moog et al, 2011, Theunissen et al, 2012) and academic skills (e.g. Venail et al, 2010).

The positive impact of early hearing screening is based on the assumption that rehabilitation and, eventually, cochlear implantation in children diagnosed as congenitally severely HI is initiated early on. A study in Flanders (2010) shows that 94% of the bilaterally deaf preschool population (2y6m-6y) receive a CI out of which 25% are bilaterally implanted (De Raeve and Lichtert, 2012). This implies that 6% of deaf preschool children is currently not implanted. Our clinical experience is that bilaterally deaf non-implanted children are mostly second-generation deaf children (deaf children from deaf parents), children from families with a low socioeconomic status (SES), and children from ethnic minorities.

Decision-making process

The **decision-making process** is often stressful for parents, especially for parents without hearing impairment navigating a world previously unknown to them. After the diagnosis of their child's deafness, parents are presented with a great deal of information about deafness and the education, communication, and technology options for their deaf children (Hardonk et al, 2011). Li and colleagues (2004) pointed out that, in making decisions for their deaf children, parents are often influenced by their beliefs, values, and attitudes as much as by the information provided by professionals. Archbold and co-workers (2006) showed that parents who placed the most importance on their child's learning to talk and participate in the hearing world, had less difficulty making the decision than parents who placed less importance on these skills and worried more about whether their child would be part of the deaf or hearing world.

Few studies have focused on the decision-making process of deaf parents – including but not limited to culturally Deaf parents. Hardonk et al (2011) performed a qualitative study which included six Flemish children aged 5-9 yrs with at least one deaf parent. One child received a CI, four were wearing a HA and one child did not use hearing devices. This study pointed out that deaf parents with deaf children attach less importance to professional advice regarding CI compared to hearing parents having a deaf child. Second, most deaf parents in this study indicated that they assigned limited priority to spoken-language development and oral communication skills. These findings have implications for the professional practice since deaf parents might not follow professionals' suggestions regarding early amplification. In addition, the stereotype of deaf parents always rejecting cochlear implantation is challenged by the findings from Obasi (2008) and Hardonk et al (2011).

Ethnic minorities

Research has demonstrated that children from higher-income families are implanted at significantly higher rates than children from families with lower incomes. Likewise, other research shows that deaf children from **ethnic minorities** are less implanted in the United States (e.g. Sorkin and Zwolan, 2008). Recently, Wiefferink and colleagues (2011) explored how counseling and rehabilitation of deaf children of Turkish-origin parents can be improved in the Netherlands. This qualitative study shows that, first, most of the parents initially did not believe their child was deaf and later on regretted the delayed start of rehabilitation. Second, the parents had little confidence in the national health system and sought a second opinion from a medical doctor of their own national origin. Third, the parents did not know how they could be actively involved in the care of their deaf child. Some implications for clinical practice were deducted from this study. First, anticipated regret might be used as a counseling technique to convince parents to start rehabilitation early. Second, giving parents the opportunity to talk with another family with a deaf child from the same ethnic group, or referring the parents to a physician of their ethnic group for a second opinio, might be helpful. Third, the use of an interpreter should be encouraged, because communication between these parents and health care professionals is not always optimal. Especially if only one of the parents (partially) masters the language spoken during counseling.

All parents of children referred by the UNHSP receive information regarding the options available to their HI child. Currently, clinical care and rehabilitation is insufficiently tailored to the needs of deaf parents, parents with a low socioeconomic status, and parents form ethnic minorities. **In future**, deaf children coming from these groups should be given specific attention respect to decision-making and rehabilitation in order to maximize their communication possibilities in daily life.

Academic achievement after UNHS

The implementation of the UNHSP in Flanders has led to a significantly lower average age of implantation and resulted in a better speech perception and production outcome. Given that the UNHSP was implemented in Flanders only from 1998, the cohort of early diagnosed children is currently younger than 15 yrs. For many years, outcome evaluation has been focused on speech and language performance. The impact of the implementation of this UNHSP on academic skills such as **reading, writing, and academic achievement**, is a topic for further research. Until now, few researchers have focused on these skills because, in most countries, early screened (and implanted) children haver not yet reached secondary grade level. Research compared e**mergent literacy skills,** such as phonological awareness and executive functioning in early implanted children, HI children, and normal hearing children after they finished kindergarten (Nittrouer et al, 2012). Results showed that the implanted children scored roughly 1 SD below the mean performance of normal hearing children on most tasks, except for syllable counting, reading fluency, and rapid serial naming. Thus, even deaf children that have benefitted from early identification and implantation have a higher risk of problems with emergent literacy. Geers and Hayes (2011) investigated reading, writing, and phonological processing skills of adolescents with at least 10 yrs of CI experience. They assessed 112 implanted high-school adolescents aged between 15.5 and 18.5 yrs on a battery of reading, spelling, expository writing, and phonological processing skills. The study group had participated in a previous battery of reading assessment during early elementary grades, between the ages of 8.0 to 9.9 yrs. A group of normal hearing peers served as control group. Results revealed that on reading tests many of the implanted adolescents performed within or even above the average range of results for the hearing peers. Importantly, when compared to their own results obtained during elementary grade, results showed that good early readers were also good readers in high school. Moreover, the majority of implanted adolescents maintained their reading levels over time compared with the hearing peers, indicating that the gap in performance did not widen for most implanted adolescents. Unlike reading skills, written expression, and phonological processing skills posed a great deal of difficulty on the implanted adolescents. Overall, the results revealed significant delays in spelling, expository writing, and phonological processing skills. Phonological processing skills also appeared to be a critical predictor of second grade literacy skills, accounting for 39% of variance remaining even after discounting child, family, and implant characteristics. Thus, phonological processing skills should be assessed and enhanced during rehabilitation. It should be noted that the averaged age at implantation was 3.5 yrs in Geers and Hayes' study. Implantation age below 2 yrs, which is common within Flanders since the implementation of the UNHSP, may have an impact on reading and writing. The impact is unknown and warrants **further investigation**.

In 2002, Govaerts and colleagues showed that in Flanders, integration into mainstraim school tend to decrease with increasing age at implantation. They showed that only 33% of children implanted after the age of 4 years are likely to be integrated in mainstraim schools by the age of 6 or 7 years, whereas 67% of children implanted before their first birthday attend mainstraim school at the age of 3 years. De Raeve and Lichtert compared integration data in mainstraim schools in Flanders from 1990-1991 and 2009-2010 and showed a significant increase of hard-of-hearing children attending mainstraim schools. Furthermore, their data showed that today, only 25% of implanted children enters preschool (2yr6m-6yr) in a regular school. The number of enrolled implanted children in regular schools increases to 46% in primary school and 66% secondary school (De Raeve and Lichtert, 2012). The possible difference between the Govaerts and the De Raeve and Lichtert study might be that Govaerts and co-workers excluded children with severe mental retardation and cochlear malformations, while no excusion criteria were used in the study of De Raeve and Lichtert.

The full impact of early implantation on **academic achievement**, especially after secondary school, remains unknown. Studies examining the impact of early implantation on outcomes, such as academic achievement, may be tainted, for example, by excluding low performers or non-users, or by excluding longitudinal data (Beadle et al 2005). In **future**, tracking the academic progress of a cohort of early implanted Flemish children from preschool over primary school and secondary school into high school, should be considered.

9.2.2 Impact of additional disabilities

The success rate of CIs on congenitally deafened, early implanted children, has led to the broadening of initially restrictive CI candidacy criteria to include a myriad of children affected by SNHL, including **children with multiple disabilities**. Hence, the number of HI children with additional disabilities that receive CIs has increase. A decade ago, the percentage of children with additional disabilities that receive CI was reported at 5 to 8%, because clinical researchers were still exploring the possible benefit of cochlear implantation in multiple handicapped children (e.g. Pyman et al, 2000, Waltzman et al, 2000). Nowadays, the proportion of implanted children with additional disabilities has increased to approximately 15 to 45% of currently implanted children (e.g. Filipo et al, 2004, Edwards et al, 2007, Wiley et al, 2008, Meinzen-Derr et al, 2009, Birman et al, 2012).

Our study showed that early screening has a positieve impact on speech perception and production skills, while additional impairments have a negative impact (**Chapter 4**). Birman and colleagues (2012) pointed out that, 12-months postoperative, only 52% of children with additional disabilities achieve a good CAP score (at least 5), compared to 96%of children without disabilities. We should be prudent in the prediction of outcome after implantation in very young children, given that a number of additional disabilities such as attention deficit hyperactive disorder (ADHD) and autism, cannot be diagnosed in very young children. Today, up to 45% of implanted children have additional disabilities. These children are often difficult to test with the standard speech perception and production tests currently available in Dutch. Thus, developing specific speech perception and production test materials that can be applied to these implanted children with special needs, should be considered. Current rehabilitation materials also need to be reconsidered, in order to provide these children with the opportunity to achieve their full potential with CI. **Future research** in this population is critical to enable appropriate parental and familial counseling and decision-making with realistic (postoperative) expectations.

9.2.3 Impact of congenital cytomegalovirus

Early detection of HL and additional disabilities are certainly two important factors that influence the outcome of cochlear implantation. Children deafened by cCMV are at risk for these factors, given that progressive and/or delayed-onset SNHL occurs in 18 to 50% of cCMV cases (Fowler et al, 1997, Fowler and Boppana, 2006) and a large portion of them suffers from additional disabilities such as mental retardation (2-45%) or chorioretinitis (1-15%) (Dahle et al, 2000, Fowler and Boppana, 2006).

Rosenthal and colleagues (2009) focused on **delayed SNHL** in 580 cCMV infected infants. They showed that asymptomatic cCMV infants with normal hearing at 6 months of age developed HL at the rate of nearly 1% per year (until age 8), whereas in symptomatic infants developing HL, the rate increased to 4% per year. Hence, their cumulative risk by age 8 was substantial: respectively 6.9% and 33.7% for populations of asymptomatic and symptomatic children. Cumulative risk of delayed SNHL may even be higher, as these numbers did not include children whose newborn hearing test was normal but developed HL within the first six months.

Due to this progressive and/or delayed-onset nature of cCMV SNHL, the UNHSP will not identify a significant proportion of cCMV-related HI children. Fowler and colleagues (1999) pointed out that the UNHSP will not identify approximately half of SNHL that may impair speech and language development in the cCMV population. Since 2007, audiological, neurological, and ophthalmological data of Flemish cCMV-positive newborns, has prospectively been registered over a 6-yr follow-up period in several hospitals. Thus, infants diagnosed as cCMV positive are assessed audiologically to detect, and subsequently manage progressive or delayed-onset SNHL. The hearing status is closely monitored through the Flemish CMV protocol (**Figure 9.1**).

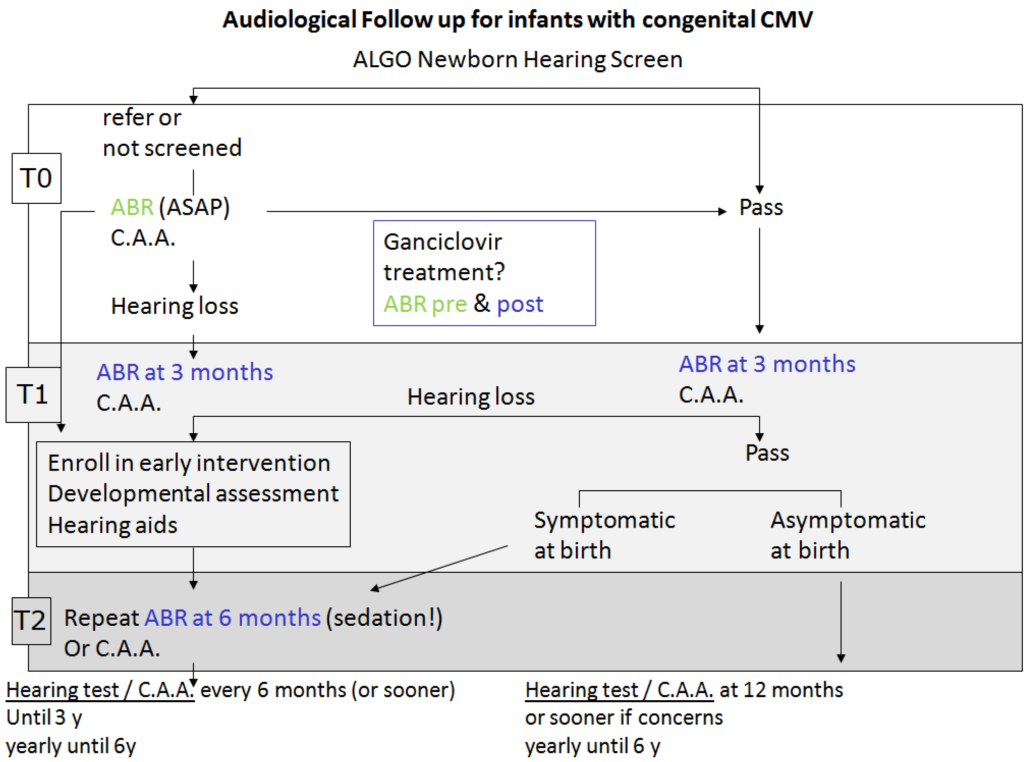

Figure 9.1 Flemish CVM-protocol.
ABR= Auditory Brainstem Respons, y= year

The unknown cCMV positive status can hamper early diagnosis of hearing impairment (Fowler et al, 1999). Research has studied the implementation of a screening program to diagnose cCMV during pregnancy (e.g. Adler, 2011, Johnson and Anderson, 2013, Walker et al, 2013) or in newborns (e.g. Barbi et al, 2006, Grosse et al, 2009). National organizations currently recommend against routine screening during pregnancy or in newborns, because more evidence is needed about the safety, reliability and validity of screening tests (Leung et al, 2012, Johnson and Anderson, 2013). Until more-effective interventions are available, it will be important to provide women of child-bearing age with better education, including behavioral intervention to reduce exposure to infected body fluids of young children (Cheeran et al, 2009, Adler, 2011).

Besides a possible delay in the detection and management of HL, **cCMV** children also risk having **additional disabilities**. The coexistence of symptomatic cCMV and additional disabilities is well-known, as these children may suffer from mental retardation, cerebral palsy, seizures, and visual impairment (e.g. Pass et al, 1980, Conboy et al, 1987, Bale et al, 1990, Boppana et al, 1992). Recent research has focused on additional psycho-neurological disorders in both symptomatic and asymptomatic cCMV children. Yamashita et al (2003) and Yamazaki et al (2012) demonstrated that additional psycho-neurological disorders affect the outcome of CI in cCMV children. The Yamazaki et al (2012) study included 11 cCMV-CI and 14 genetic-CI children. Nine of the 11 cCMV-CI infants suffered from psycho-neurological disorders such as ADHD (N=1), pervasive developmental disorder (PDD, N=2) and mental retardation (MR, N=6). Outcome after implantation was investigated by testing four different aspects of auditory perceptual ability, with increasing degree of difficulty (hearing threshold, closed-set discrimination, open-set discrimination, language development). The results showed comparable hearing thresholds in both groups, with lower scores on tasks requiring higher levels of auditory processing and the severity of impairment being determined by the type of psycho-neurological disorder (ADHD<MR<PDD). Overall, the presence of additional disabilities could explain a portion of the large inter-individual variability in cCMV-CI children.

Our study revealed that over a 5-yr period, cCMV-CI infants and their Cx26 counterparts, progress equally. However, cCMV-CI children with abnormal MRI results tend to progress slower on speech production tasks. The significance of the observed abnormal MRI results needs further clarification (**Chapter 5**).

Further, long-term follow-up of several outcome variables, after CI in implanted cCMV children is required including social-emotional, reading and motor skills. To this end the UZ Ghent is conducting a long-term follow-up study aiming to evaluate these skills in HI cCMV-infants.

Awareness of the possible impact of cCMV on vestibular function and balance has grown only recently. We found that four out of six cCMV children had absent cVEMP responses prior to implantation, as opposed to their Cx26 counterparts. A longitudinal study on balance and vestibular function in HI children, and more specifically in children with cCMV, is actually being conducted at our clinic. The results of this study will deepen the understanding of the impact of cCMV, not only on the cochlea, but also on the labyrinth *in toto*.

9.3 Speech perception outcomes in the adult CI population

9.3.1 Channel interaction in adult CI users

The outcome of cochlear implantation in adults is highly variable. This variation might be caused by a lack of selectivity of stimulation due to a degree of interaction that occurs when overlapping subsets of nerve fibers are stimulated by various electrodes of a multi-electrode array (Abbas et al, 2004). In addition to psychophysical measurements, channel interaction can be assessed through the use of the Electrically evoked Compound Action Potential (ECAP).

For this purpose, our study included data from 20 postlingually deafened adult CI users and aimed to investigate a possible relationship between channel interaction and speech perception (**Chapter 6**). First, it was hypothesized that subjects with smaller widths at ECAP profiles should have better speech perception scores because narrow spatial profiles would allow for more independent information channels. Novel in our study, compared to prior research was the use of a thorough speech perception test battery, including relative information transmission scores. However, neither our data, nor the data from prior studies confirmed this hypothesis (Hughes and Abbas, 2006b; Hughes and Stille, 2008; Tang et al 2011; van der Beek et al 2012). In addition, we found that 47% of ECAP derived spatial profiles had a displaced peak. Few studies have investigated the impact of displaced peaks on speech perception performance. It was hypothesized that recipients without displacements would achieve better scores on speech perception tests. No statistical significant difference between recipients with or without displaced peaks was found. Results tentatively suggest that the presence of displaced peaks, assessed by means of the clinically available spatial measurement techniques, do not equate to lower speech perception scores. Hughes and Stille (2008) hypothesized that the presence of displaced peaks could be compensated by the use of higher-level auditory mechanisms. However, in our study, the closed-set identification test of consonants, which makes use of lower-order auditory and linguistic mechanisms, did not show a significant difference between recipients with or without displaced peaks. When accounting for duration of deafness prior to CI, the results show that all but one recipient (meningitis) with displaced spatial profiles had a longer duration of deafness prior to CI compared to recipients without displacements. First, spiral ganglion loss might be accounted for the presence of displaced peaks given that a longer duration of deafness is sometimes characterized by a lower degree of spiral ganglion survival. Secondly, the absence of differences in speech perception between recipients with and without peak displacements might be due to temporal and/or intensity cues. Recipients with lower spectral integration might compensate for this impairment by having higher temporal en/or intensity integration cues.

The clinical application of currently available spatial measurement techniques warrants **further examination**. Channel interaction remains a significant challenge. To this end, it is first critical to understand the distribution of the electric field in the cochlea and the interaction between the electrical stimulation and the auditory nerve, since these factors might play a key role in determining the outcome after cochlear implantation (Tang et al, 2011). Additional research is necessary to determine the role of objective measurements, such as ECAP spatial profiles, as a means for predicting speech perception in CI users. The assessment of spectral, temporal and intensity integration cues in a large group of CI recipients will benefit the exploration of possible compensation mechanisms within these integration cues, leading to better speech perception scores. In addition, as suggested by Undurraga (2013), ECAPs might be used to gain insight in the degree of degeneration of the peripheral process along the cochlea.

9.3.2 Cochlear implantation in early-deafened, late-implanted adults

Without doubt, **duration of deafness** is an important determinant influencing speech perception outcomes after CI. One specific group of CI recipients suffering from an extensive period of auditory deprivation are early-deafened, late-implanted subjects. These subjects are generally considered poor CI users. However, recent studies have shown improved speech perception outcome after CI in some early-deafened, late-implanted users (e.g. Van Dijkhuizen et al, 2011, Caposecco et al, 2012). Thus, this population also shows a high inter-subject variability.

We conducted a study to assess and predict speech perception outcomes in a group of 19 early-deafened, late-implanted adults (**Chapter 7**). In general, our data demonstrate that early-deafened, late-implanted adults progress beyond their preimplant state on sound detection and consonant identification tasks. However, their results on more demanding measurements (words and sentences in noise) are limited. In addition, we investigated the possible predictive value of speech production. In this regard, our study showed that one single factor, errors in speech production (ErrSP), accounted for 78% of the variance of postimplantation speech perception (NVA70). Thus, participants with fewer ErrSP achieve better scores on speech perception. Counseling and candidate selection will therefore be facilitated byusing ErrSP s a predictor in this particular patient population given that ErrSP does not depend on patients self-reported factors, such as age at onset of deafness or age at first HA. Based on our study, the used speech production test was included in the early-deafened, late-implanted UZ Ghent counseling protocol. As a clinical rule of thumb, future early-deafened, late-implanted CI candidates achieving -0.20 on the ErrSP factor, should be expected to achieve some degree of open-set speech understanding.

However, some caution must be made when generalizing the results presented, for the following three reasons: First, the **future** inclusion of a larger number of patients, might alter the results obtained, since the wide confidence limits reported are of concern (**Figure 7.6**). Second, speech perception tests were only obtained one year after cochlear implantation and a longer follow-up period is therefore possibly required for early-deafened, late-implanted patients to achieve their maximal speech performance scores. Third, early-deafened, late-implanted recipients achieving lower speech perception scores are not necessarily unsatisfied with their outcomes, given that the majority of these subjects experience a substantial increase in quality of life. Therefore, CI candidates achieving an ErrSP score below -0.20 should not be refused to be implanted, but special attention should be given during preoperative counseling to the possibly limited benefit with regard to speech perception outcome after implantation.

9.3.3 Impact of multiple variables in postlingually deafened adults

In postlingually deafened implanted adults, a number of prior studies looked at predictive variables for speech perception outcome (e.g. Blamey et al, 1996, Holden et al, 2013). In 1996, a retrospective analysis of speech recognition in quiet for 808 postlingually deafened adults described five clinical predictors accounting for 21% of variance in CI performance, i.e. in order of relative importance: duration of severe to profound HL, age at implantation, age at onset of severe to profound HL, duration of CI experience and etiology (Blamey et al, 1996). The factor accounting for the largest proportion in variance was duration of deafness, with longer duration of deafness negatively influencing speech perception outcome. Since many factors have changed since 1996, including the implantation of CI candidates with residual hearing, an updated version of the 1996 study was conducted in 2011, including 2251 postlingually deafened adult CI recipients (Blamey et al, 2013). In this study, duration of CI experience was the most important factor, followed by age at onset of severe to profound SNHL, duration of severe profound SNHL and etiology. Together, these factors accounted for 10.5% of the variance in CI performance in quiet. The differences between both studies are likely due to relaxed patient selection criteria, improved clinical management of HL, modification of surgical practice, and improved devices. The difference in total variance explained in these two studies, as indicated by Blamey et al (2013), might be found in the greater random variance in the 2011 study, due to the inclusion of more centers. Nevertheless, a large proportion of residual variance remains unexplained, possibly due to the fact that many other potential factors were not included in either of these two studies. Therefore, Lazard et al (2013) included 15 new factors, accessible from clinical information routinely collected, possibly accounting for a portion of this residual variance and conducted several new statistical analyses on the dataset from the 2011 study. Four out of these 15 factors (duration of moderate HL, HA use, CI brand, and percentage of active electrodes) were found to have a significant influence on CI outcome. However, an analysis of the five factors of the 2012 study as well as these four factors of the 2013 stuyd, did not modify the relative importance of any of the main factors described earlier (Blamey et al, 2013). This new model accounted for 22% of the variance in quiet as well as in noise, still leaving 78% unexplained. Possibly, the impact of higher order cognitive reorganization might account for a portion of the currently unexplained variance (e.g. Lazard et al, 2010, Strelnikov et al, 2010).

To date, research has mainly focused on gaining insight in the peripheral auditory system and nervous pathway. **In future**, moving from a 'bottom-up' to a 'top-down' approach might reveal a portion of the unexplained variability in outcome after implantation, given thatcentral auditory pathways, including cortical processing and cortical plasticity, are critical factors in the CI success (Cosetti and Waltzman, 2011).

9.3.4 Music appreciation in adult CI recipients

While open-set speech perception in quiet is an achievable goal for the majority of postlingually deafened adult CI users, the ability to perceive and **enjoy music** remains a challenge (e.g. Gfeller et al, 2005). The majority of studies examining music perception have examined accuracy for pitch, rhythm, timbre and melody. In general, CI users' skills for these key structures of music are poor, except for rhythm (e.g. McDermott, 2004). Compared to normal-hearing (NH) listeners, CI users are significantly less accurate in identifying direction of pitch change, musical timbre and familiar melodies (for a review, see McDermott, 2004, Looi et al, 2012). For example, when asked to recognize "real-world" musical excerpts in an open-set task, a group of 79 CI subjects yielded a mean score of 16% correct answers, compared to a score of 55% for NH listeners (Gfeller et al, 2005).

While perceptual accuracy for music is important, appreciation and enjoyment also warrant research as both also contributes to perceived benefits (Looi et al, 2012). Enjoyment of music is one of the most common self-reported hearing difficulties by CI users (Zhao et al, 2008). Over the past decade, studies investigating music appreciation in unilaterally implanted adult CI users have only shown a limited number of factors contributing to better music appraisal. First, a study by Gfeller and colleagues (2009) examining music appraisal in 209 adult CI users showed that neither device type nor processing strategy predicted music appraisal. Secondly, research by Looi and She (2010), Gfeller et al (2000) and Lassaletta et al

(2008) showed that recipients can enhance their listening enjoyment to some extent by controlling their listening environment (e.g. quiet, non-reverberant room, good quality sound equipment, appropriate volume etc.) and using visual cues (e.g. watching the performer, or following a score or lyrics). In addition, recipients reported that their music experience was enhanced by being selective in their choice of music, such as listening to music that they are familiar with, choosing less complex music with a strong beat, and/or choosing music with lyrics.

The current trend to extend indications for cochlear implantation, together with the improvement in soft surgery and electrode design, will lead to more CI users with residual hearing in one or both ears. The benefit of bimodal stimulation (CI in one ear and HA in the opposite ear) has already been shown in terms of speech discrimination, especially in noise, and to a lesser extent sound localization (e.g. Ching et al, 2006). However, the possible benefit of bimodal hearing on music appreciation is unclear.

Therefore, a study was conducted to investigate the impact of bimodal hearing on music appreciation (**Chapter 8)**. First, our study showed that bimodal listeners believe that the use of a contralateral HA enhances music listening, especially with respect to melody perception. This finding corresponds with the results of Gfeller and colleagues (2008, 2010) and Looi and She (2010). Secondly, bimodal users find that music sounds less natural in the CI-only condition than in the bimodal condition, thus implying that the use of a HA enhances the naturalness of the perceived sound. Moreover, CI-only users emphasize the need to develop new speech-coding strategies enabling them to improve listening to music, while bimodal listeners do not express such need. These findings are consistent with other studies that have demonstrated the benefits of bimodal listening over the CI-only condition for a range of listening tasks (Leal et al, 2003, Kong et al, 2004, El Fata et al, 2009). Consequently, the importance of preserving and, if necessary, enhancing residual hearing, either in the contralateral ear by means of a HA or in the same ear by means of electroacoustic stimulation (EAS) is underlined.

The Belgian health care system grants adult CI users a maximum of four years of multidisciplinary therapy after implantation. In addition, the Belgian health care system also requires a long-term follow-up after implantation. However, De Raeve and Wouters (2013) reported that there are currently no published data available on the demand for, and consumption of, rehabilitation sessions after cochlear implantation in adults. Furthermore, their study reports that some adults receive no or limited therapy after implantation. Possibly, these CI recipients consider that they achieve sufficient speech understanding or do not recognize the benefit of speech therapy. As demonstrated by research of Boothroyd (2010) rehabilitation is likely to be more effective when the individual is interested and motivated to participate. In addition to results obtained by Looi and She (2010), our study also revealed that a large portion of the current adult CI users believes that music training would be helpful in maximizing their CI benefit. Thus, the findings of our study support the introduction of a music training program (MTP) in an early stage of rehabilitation as this might enhance enjoyment of music for both good and poor CI recipients. Indeed, several prior studies also showed showed that music training is effective in improving perception of various aspects of music listening for individuals with CIs, although the degree of improvement varies depending on a number of factors such as age of participant, life experiences, and stimuli used in training (e.g. Gfeller et al, 1998, Gfeller et al, 2002, Fu et al, 2007, Driscoll, 2012). Nevertheless, music training should not be exclusively reserved to the good speech performers, since it is well known that music training can be a valuable aid in improving listeners' perception of language. In sum, we hope that the early implementation of MTP in rehabilitation programs can allow adult CI users to increase their enjoyment and appreciation of music and to maximize this potential with commercially available technology.

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Curriculum Vitae

Birgit Philips werd geboren op 16 november 1982 te Leuven.

Na het afronden van haar middelbare studies Wiskunde-Wetenschappen aan het Heilig-Hartinstituut te Heverlee in 2000, startte ze aan aan de opleiding Logopedische en Audiologische Wetenschappen aan de Katholieke Universiteit Leuven. In 2004 behaalde ze met grote onderscheiding het diploma van licentiaat in de Logopedische en Audiologische Wetenschappen, afstudeerrichting logopedie.

Nadien vervolledigde ze aan diezelfde universiteit haar licentiaatsdiploma tot geagreggeerde in het secundair onderwijs (2005, grote onderscheiding).

In 2006 behaalde ze, eveneens met grote onderscheiding, het masterdiploma in de Logopedische en Audiologische Wetenschappen, afstudeerrichting audiologie, aan de Universiteit Gent.

Oral presentations

1. **8th European Investigators' Conference Advanced Bionics**, 7-8.03.2008, Marrakech, Marokko.
   Philips B, Vinck B, De Vel E, Dhooge I. 'Defining the success rate in spatial selectivity measures using RSPOM.'
2. **Newborn Hearing Screening Congress**, 19-21.06.2008, Cernobbio, Italy.
   Philips B, Corthals P, De Raeve L, Vinck B, Dhooge I. 'Outcome of children fitted with cochlear implants. Comparing data of high and low performing children.'
3. "***Koninklijke Belgische Vereniging voor ORL, Gelaat- en Halschirurgie***", 28-29.11.2008, Antwerpen.
   Philips B, Corthals P, De Raeve L, Vinck B, Dhooge I. 'Outcome of children fitted with cochlear implants. Comparing data of high and low performing children.'
4. **9th European Symposium on Pediatric Cochlear Implantation** (ESPCI), 14-17.05.2009, Warsaw, Poland**.**
   Philips B, De Leenheer E, De Vel E, Dhooge I. 'Cochlear implantation in infants deafened by cCMV.'
5. **CORA-CI**, 15.12.2009, Brussel.
   Philips B, 'Cochlear implantation in infants deafened by cCMV.'
6. "***Studiedag CMV***", 05.03.2010, Sint-Lievenspoort Gent.
   Philips B. "*Cochleaire implantatie bij cCMV kinderen.*"
7. **9th European Investigators Meeting**, 12-13.03.2010, Amsterdam, The Netherlands.
   Philips B, Dhooge I. 'Characteristics and determinants of music appreciation in adult CI users.'
8. **Intensive program on technical innovations in audiology**, 11.04.2011, Arteveldehogeschool Gent (invited speaker).
   Dhooge I, Philips B. 'Cochlear implantation: breaking the barrier of silence.'
9. **10th European Symposium on Pediatric Cochlear Implantation** (ESPCI), 12-15.05.2011, Athens, Greece.
   Philips B, Dhooge I. 'Cochlear implants in children deafened by cCMV.'
10. **10th European Symposium on Pediatric Cochlear Implantation** (ESPCI), 12-15.05.2011, Athens, Greece (round table, invited speaker).
    Philips B, De Bruyn L, Leman M, Dhooge I. 'Music appreciation in adults and children with CI.'
11. **11th European Federation of Audiology Society** (EFAS) Congress, 19-22.06.2013, Budapest, Hungary.
    Philips B, Baudonck N, Dhooge I. 'Assessing and predicting speech perception in early-deafened, late-implanted adults.'
12. **ORL-NKO Annual Congress**, 15-16.11.2013, Brussels, Belgium.
    Philips B, Baudonck N, Dhooge I. 'Assessing and predicting speech perception in early-deafened, late-implanted adults.'

Poster presentations

1. **Conference on Auditory Implantable Prostheses** (CIAP), 12-17.07.2009, Lake Tahoe, CA, USA.
   Philips B, Corthals P, De Vel E, Dhooge I. 'Spatial spread measurements in HR 90K users.
2. *"**Wetenschappelijk Onderzoek Logopedie en Audiologie Vlaanderen** (WOLAV)",* 11.12.2009, Antwerpen.
   Philips B, De Leenheer E, Dhooge I. 'Cochlear implantation in infants deafened by cCMV.'
3. **9th European Investigators Meeting**, 12-13.03.2010, Amsterdam, The Netherlands.
   Philips B, De Vel E, Dhooge I. 'Test-retest reliability within spatial measurement techniques in adult CI users.'
4. **11th International Conference on Cochlear Implants and Other Auditory Implantable Technologies**, 30.06-03.07.2010, Stockholm, Sweden.
   Philips B, Dhooge I. 'Characteristics and determinants of music appreciation in adult CI users.'
5. "***Wetenschapsdag UZGent***" 7.4.11, Gent
   Philips B, Dhooge I. 'Characteristics and determinants of music appreciation in adult CI users.'
6. **Conference on Auditory Implantable Prostheses** (CIAP), 24-29.07.2011, Asilomar, CA, USA.
   Philips B, Dhooge I. 'Correlation between spread of excitation measurements and speech perception in adult cochlear implant users.'
7. **ESPO**, 21-23.05.2012, Amsterdam, The Netherlands.
   Philips B, Dhooge I. 'Cochlear implants in children deafened by cCMV.'
8. **Objective Measures in Auditory Prostheses**, 19-21.09.2012, Amsterdam, The Netherlands
   Philips B, Dhooge I. 'Feasibility and long-term stability of spread of excitation measurements in adult cochlear implant users.'

Attended conferences

1. **8th European Investigators' Conference** Advanced Bionics, 7-8.03.2008, Marrakech, Marokko
2. **Newborn Hearing Screening Congress**, 19-21.06.2008, Cernobbio, Italy
3. "***Studiedag SIG, Vroegdetectie gehoorverlies***", 27.10.2008; Destelbergen
4. "***Koninklijke Belgische Vereniging voor ORL, Gelaat- en Halschirurgie***", 28-29.11.2008, Antwerpen.
5. **Children's Audiology Course** (CMV-ANSD-Autism-AVS), 3-6.03.2009, Nottingham, UK
6. **Auditory Neuropathy Spectrum Disorder** The Ear Foundation, 12.03.2009, Mechelen
7. **9th European Symposium on Pediatric Cochlear Implantation** (ESPCI), 14-17.05.2009, Warsaw, Poland.
8. **9th Bionics European Research Group Meeting**, 24.06.2009, Cambridge, UK
9. **Conference on Auditory Implantable Prostheses** (CIAP), 12-17.07.2009, Lake Tahoe, CA, USA. **Student travel aid acknowledgment.**
10. **Cochlear Implantation in Ethnic Minority Families**, 9.12.2009, The Ear Foundation, Mechelen
11. *"**Wetenschappelijk Onderzoek Logopedie en Audiologie Vlaanderen** (WOLAV)",* 11.12.2009, Antwerpen.
12. "***Studiedag CMV***", 05.03.2010, Sint-Lievenspoort Gent.
13. **10th Bionics European Research Group Meeting**, 11.03.2010, Amsterdam, The Netherlands
14. **9th European Investigators Meeting**, 12-13.03.2010, Amsterdam, The Netherlands
15. **11th International Conference on Cochlear Implants and Other Auditory Implantable Technologies**, 30.06-03.07.2010, Stockholm, Sweden
16. **6th International Symposium on Objective Measures in Auditory Implants**, 22-25.09.2010, St. Louis, MI, USA
17. "***Wetenschapsdag UZGent***" 7.4.11, Gent
18. **Intensive program on technical innovations in audiology**, 11.04.2011, Arteveldehogeschool Gent.
19. **10th European Symposium on Pediatric Cochlear Implantation (ESPCI)**, 12-15.05.2011, Athens, Greece!i
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21. **Objective Measures in Auditory Prostheses**, 19-21.09.2012, Amsterdam, The Netherlands
22. "***Evenwicht en muziekperceptie bij kinderen met een gehoorstoornis***", 20.10.2013, Ghent, Belgium
23. **11th European Symposium on CI in Children** (ESPCI), 22-26.05.2013, Istanbul, Turkey
24. **11th European Federation of Audiology Society** (EFAS) Congress, 19-22.06.2013, Budapest, Hungary
25. **"*Opgroeien met een cochleair implantaat*",** 21.09.2013, Leuven, Belgium
26. **"*Van bionisch naar bimodaal horen met een CI*",** 9.11.2013, Ghent, Belgium
27. **ORL-NKO Annual Congress**, 15-16.11.2013, Brussels, Belgium.

A1 Publications

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5. Philips B, Corthals P, De Raeve L, D'Haenens W, Maes L, Bockstael A, Keppler H, Swinnen F, De Vel E, Vinck BM, Dhooge I. Impact of newborn hearing screening: comparing outcomes in pediatric cochlear implant users. *Laryngoscope*. 2009;119(5):974-979.
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Appendix

Universitair Ziekenhuis Gent

UNIVERSITEIT GENT

Vragenlijst muziekappreciatie bij CI-patiënten

A. Algemene vragen over het **huidige spraakverstaan** met uw implantaat.

1) Kruis telkens 1 vakje aan.

	Nooit	Moeilijk	Soms	Vaak	Zeer vaak	n.v.t.
Kunt u in een rustige omgeving een gesprek voeren met één persoon?	☐	☐	☐	☐	☐	☐
Verstaat u de nieuwslezer op tv bij een normaal volume *met* ondertiteling?	☐	☐	☐	☐	☐	☐
Verstaat u de nieuwslezer op tv bij een normaal volume *zonder* gebruik te maken van ondertiteling?	☐	☐	☐	☐	☐	☐
Wanneer u een rustig gesprek voert met uw arts in een onderzoekskamer, kunt u de conversatie dan vlot volgen?	☐	☐	☐	☐	☐	☐
Kunt u in een drukke winkel de verkoper verstaan?	☐	☐	☐	☐	☐	☐
Kunt u iemand die u in een drukke straat aanspreekt verstaan?	☐	☐	☐	☐	☐	☐
Kunt u iemand die u aanspreekt op een receptie verstaan?	☐	☐	☐	☐	☐	☐
Wanneer u met meerdere mensen aan tafel zit en er is vrij veel lawaai op de achtergrond (bv. op restaurant), kunt u dan een gesprek met de personen in uw onmiddellijke omgeving volgen?	☐	☐	☐	☐	☐	☐
Herkent u verschillende familieleden aan hun stem zonder dat u ze ziet?	☐	☐	☐	☐	☐	☐
Kunt u op straat het geluid van een auto onderscheiden van dat van een bus?	☐	☐	☐	☐	☐	☐
Kunt u mannen- en vrouwenstemmen van elkaar onderscheiden?	☐	☐	☐	☐	☐	☐

2) Hieronder volgen enkele stellingen over uw luistergewoontes voor en na implantatie. Duid aan wat voor u van toepassing is.

<u>Voor</u> uw implantatie

	Nooit	**Zelden**	**Soms**	**Vaak**	**Zeer vaak**	**n.v.t.**
Ik luisterde naar muziek *<u>vóór</u>* mijn implantatie.	☐	☐	☐	☐	☐	☐
Ik luisterde naar klassieke muziek (waar voornamelijk gebruik gemaakt wordt van klassieke instrumenten zoals piano, viool, fluit…).	☐	☐	☐	☐	☐	☐
Ik luisterde naar populaire muziek (popmuziek, R&B, schlagers…).	☐	☐	☐	☐	☐	☐
Ik luisterde naar muziek waar voornamelijk gebruik gemaakt wordt van elektrische gitaren (rock, heavy metal…).	☐	☐	☐	☐	☐	☐
Ik luisterde naar andere muziek: …………………………………………………………….	☐	☐	☐	☐	☐	☐
Ik luisterde naar muziek op de radio.	☐	☐	☐	☐	☐	☐
Ik luisterde naar muziek op plaat, cd, cassette en/of mp3/i-pod.	☐	☐	☐	☐	☐	☐
Ik ging naar live concerten.	☐	☐	☐	☐	☐	☐
Ik genoot van het luisteren naar muziek.	☐	☐	☐	☐	☐	☐

<u>Na</u> uw implantatie

	Nooit	**Zelden**	**Soms**	**Vaak**	**Zeer vaak**	**n.v.t.**
Ik luister *<u>nu</u>* naar muziek.	☐	☐	☐	☐	☐	☐
Ik luister naar klassieke muziek (waar voornamelijk gebruik gemaakt wordt van klassieke instrumenten zoals piano, viool, fluit…).	☐	☐	☐	☐	☐	☐
Ik luister naar populaire muziek (vb. popmuziek, R&B, schlagers…).	☐	☐	☐	☐	☐	☐
Ik luister naar muziek waar voornamelijk gebruik gemaakt wordt van elektrische gitaren (vb. rock, heavy metal…).	☐	☐	☐	☐	☐	☐
Ik luister naar andere muziek: ………………………………………………………..	☐	☐	☐	☐	☐	☐
Ik luister naar muziek op de radio.	☐	☐	☐	☐	☐	☐
Ik luister naar muziek op plaat, cd, cassette en/of mp3/i-pod.	☐	☐	☐	☐	☐	☐
Ik ga naar live concerten.	☐	☐	☐	☐	☐	☐
Ik geniet van het luisteren naar muziek.	☐	☐	☐	☐	☐	☐

	Nooit (0 uur)	Zelden (1-2 uur)	Soms (3-5 uur)	Vaak (6-8 uur)	Zeer vaak (9 uur of meer)
Hoe regelmatig luisterde u per dag naar muziek *<u>vóór uw implantatie?</u>*	☐	☐	☐	☐	☐
Hoe regelmatig luistert u per dag naar muziek *<u>na uw implantatie</u>?*	☐	☐	☐	☐	☐

B. Vragen over uw **muzikale achtergrond**.

1) **Muzikale training <u>voor uw implantatie:</u> duid aan wat van toepassing is.**
(meerdere antwoorden zijn mogelijk)

☐ Ik heb officiële muzieklessen gevolgd: algemene muzikale vorming (AMV).
☐ Ik heb officiële muzieklessen gevolgd: algemene muzikale cultuur (AMC).
☐ Ik maakte deel uit van een koor of muzikaal ensemble.
☐ Geen van de voorgaande mogelijkheden.

2) **Duid aan wat van toepassing is.**
(slechts één antwoord is mogelijk)

☐ Ik heb geen muzikale training genoten, heb weinig kennis betreffende muziek en ik heb weinig ervaring in het luisteren naar muziek.

☐ Ik heb geen muzikale training genoten, heb weinig kennis betreffende muziek maar ik heb wel ervaring in het luisteren naar muziek.

☐ Ik ben een zelfgeleerde muzikant en nam/neem deel aan muzikale activiteiten.
☐ Ik heb enige muzikale training genoten, heb een basiskennis van de muzikale terminologie en nam/neem deel aan muzieklessen of ik maak(te) deel uit van een muzikale groep op een (hoge)school en/of de universiteit.

☐ Ik heb verschillende jaren muzikale training genoten, heb een extensieve kennis van muziek en maak(te) deel uit van één (of meerdere) muzikale groep(en).

3) **Bespeelde u een muziekinstrument <u>vóór uw implantatie</u>**
☐ Ja. Welk(e) instrument(en) bespeelde u?……………………………………………………………………
☐ Neen

4) **Bespeelt u een muziekinstrument <u>na uw implantatie</u>?**
☐ Ja. Welk(e) instrument(en) bespeelt u?..
☐ Neen

5) Heeft u tijdens de logopedische therapie ooit geoefend op het luisteren naar muziek?

☐ Ja
☐ Neen

6) Heeft u thuis ooit geoefend op het luisteren naar muziek?

☐ Ja
☐ Neen

C. Vragen naar uw **beoordeling van muziek.**

1) Hoe beoordeelt u muziek *met uw implantaat*? Duid aan wat voor u van toepassing is.

	Helemaal niet akkoord	Niet akkoord	Neutraal	Akkoord	Helemaal akkoord	n.v.t.
Ik kan het luisteren naar muziek via het implantaat waarderen.	☐	☐	☐	☐	☐	☐
Ik hou van de klank van muziek via het implantaat.	☐	☐	☐	☐	☐	☐
Muziek via het implantaat klinkt natuurlijk.	☐	☐	☐	☐	☐	☐
De melodielijn van een muziekstuk is gemakkelijk te volgen via het implantaat.	☐	☐	☐	☐	☐	☐
Ik kan het ritme van een muziekstuk gemakkelijk volgen via het implantaat.	☐	☐	☐	☐	☐	☐
Het luisteren naar muziek via het implantaat geeft mij veel voldoening.	☐	☐	☐	☐	☐	☐
Muziek klinkt via mijn implantaat beter dan met mijn vroegere hoortoestel.	☐	☐	☐	☐	☐	☐

2) In vergelijking met *voor uw implantatie*…

	Ik kan het niet waarderen	Niet zo goed	Matig	Behoorlijk goed	Zoals voorheen	Beter	n.v.t.
…hoe ervaart u muziek nu?	☐	☐	☐	☐	☐	☐	☐
…hoe ervaart u het bespelen van een instrument nu?	☐	☐	☐	☐	☐	☐	☐
…hoe ervaart u het zingen nu?	☐	☐	☐	☐	☐	☐	☐
…hoe ervaart u het dansen nu?	☐	☐	☐	☐	☐	☐	☐

3) **Geef aan wat voor u van toepassing is.**

Na … maanden	3 maanden	6 maanden	9 maanden	12 maanden of meer	n.v.t.
Sinds mijn implantaat vond ik muziek aangenamer na…	☐	☐	☐	☐	☐
Sinds mijn implantaat verstond ik songteksten beter na…	☐	☐	☐	☐	☐
Sinds mijn implantatie luisterde ik opnieuw naar muziek na…	☐	☐	☐	☐	☐

4) **Bij welke gemoedstoestanden luisterde u voor en na uw implantatie naar muziek?**
(meerdere antwoorden zijn mogelijk)

	Voor implantatie	Na implantatie
Blij	☐	☐
Droevig	☐	☐
Boos	☐	☐
Van streek zijn	☐	☐
Gefrustreerd	☐	☐
Bij verveling	☐	☐
Opgewonden	☐	☐
Depressief	☐	☐

5) **Duid links de vakjes aan die voor u van toepassing zijn en duid dan voor elk geselecteerd onderdeel aan hoe/waar u het meeste luistert.**
(meerdere antwoorden zijn mogelijk)

Sinds ik mijn implantaat heb, luister ik meestal naar muziek in de volgende situaties:

☐ In de kerk luister ik …
- ☐ … naar een koor
- ☐ … naar het orgel

☐ Naar de radio luister ik …
- ☐ … in de auto
- ☐ … op het werk/thuis
- ☐ … via directe input

☐ Een live concert beluister ik …
- ☐ … in een zaal/tent/concertgebouw
- ☐ … in openlucht.

☐ Achtergrondmuziek beluister ik …
- ☐ … op tv
- ☐ … in een film
- ☐ … op café, recepties…

☐ Naar cd's, cassettes, mp3 luister ik …
- ☐ … in de auto
- ☐ … via luidsprekers
- ☐ … via directe input

☐ Ik maak muziek door …
- ☐ … zelf te zingen
- ☐ … zelf een instrument te bespelen

6) Ik kan muziek op tv/een concert beter volgen wanneer ik de zanger(es) zie.

☐ Ja
☐ Neen

7) Hoe belangrijk is muziek voor u? Beantwoord volgende vragen.

Hoe belangrijk is…

	Helemaal niet belangrijk	**Weinig belangrijk**	**Een beetje belangrijk**	**Belangrijk**	**Zeer belangrijk**	**n.v.t.**
…genieten van muziek voor u?	☐	☐	☐	☐	☐	☐
…muziek voor uw sociale leven?	☐	☐	☐	☐	☐	☐
…het voor u om een instrument te bespelen?	☐	☐	☐	☐	☐	☐
…dansen voor u?	☐	☐	☐	☐	☐	☐

8) Mocht er een implantaat bestaan dat muziek perfect zou kunnen doorgeven, zou u hier dan voor kiezen?

☐ Ja, omdat…………………………………………………………………………………………………..
☐ Neen, omdat………………………………………………………………………………………………..

9) Wat zijn uw verwachtingen van het implantaat in verband met muziek?

	Helemaal niet akkoord	Niet akkoord	Neutraal	Akkoord	Helemaal akkoord	n.v.t.
Als ik naar muziek luister, ben ik het liefst alleen.	☐	☐	☐	☐	☐	☐
Ik zou graag naar muziek willen luisteren die speciaal gemaakt is voor gebruikers van een implantaat.	☐	☐	☐	☐	☐	☐
Ik beluister graag muziek in een rustige omgeving.	☐	☐	☐	☐	☐	☐
Ik zou graag naar muziek willen luisteren die ik zelf gemaakt heb.	☐	☐	☐	☐	☐	☐
Luisteren naar muziek met mijn implantaat is moeilijk.	☐	☐	☐	☐	☐	☐
Ik denk dat leren luisteren naar muziek tijdens (logopedische) therapie zinvol is.	☐	☐	☐	☐	☐	☐
Ik luister graag naar muziek samen met vrienden en familie.	☐	☐	☐	☐	☐	☐

D. Persoonlijke vragen.

1) Heeft u een beroep dat gelinkt is aan muziek? Zoja, wat is uw beroep en welke invloed heeft uw implantaat hierop?

………

2) Droeg u <u>voor</u> uw implantatie een hoortoestel aan uw <u>geïmplanteerd</u> oor?

☐ Ja
☐ Neen

3) Droeg u <u>voor</u> uw implantatie een hoortoestel aan uw <u>niet-geïmplanteerde</u> oor?

☐ Ja, ga naar vraag 4
☐ Neen, ga naar vraag 5

4) Draagt u <u>nu</u> nog steeds een hoortoestel aan uw <u>niet-geïmplanteerde</u> oor?

☐ Ja, ga naar vraag 6
☐ Neen, ga naar vraag 7

5) Draagt u <u>nu</u> een hoortoestel aan <u>niet-geïmplanteerde</u> oor?

☐ Ja, ga verder naar vraag 6
☐ Neen, ga verder naar vraag 7

6) Duid aan wat voor u van toepassing is.

	Helemaal niet akkoord	Niet akkoord	Neutraal	Akkoord	Helemaal akkoord
Wanneer mijn hoortoestel uitgeschakeld is, terwijl mijn implantaat ingeschakeld is, ondervind ik problemen om een gesprek in rumoerige omstandigheden te volgen.	☐	☐	☐	☐	☐
Wanneer mijn hoortoestel uitgeschakeld is, terwijl mijn implantaat ingeschakeld is, ondervind ik problemen om mannen- en vrouwenstemmen van elkaar te onderscheiden.	☐	☐	☐	☐	☐
Mijn hoorapparaat biedt een meerwaarde bij het beluisteren van muziek.	☐	☐	☐	☐	☐

7) **Heeft u op uw implantaat een speciaal muziekprogramma?**
(slechts één antwoord is mogelijk)

☐ Ja, ik gebruik het.
☐ Ja, maar ik gebruik het niet.
☐ Neen, ik heb het niet nodig.
☐ Neen, maar ik zou het wel willen.
☐ Ik weet het niet.

8) Indien u nog opmerkingen/suggesties omtrent deze vragenlijst, muziekbeleving via uw implantaat, … hebt, kan u deze hieronder formuleren.

……

……

……

Alvast bedankt voor uw deelname!