Cochlear implantation in unilateral deafness (UD): Implications for speech understanding, self-perceived hearing improvement, tinnitus suppression and spatial acuity

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Abstract

Background
Patients with unilateral deafness (UD) lack the benefit of binaural hearing. As a result, they experience difficulties in understanding speech in the presence of background noise, in understanding speech that comes from the side of hearing loss (HL), as well as in localising the sound source. Furthermore, a large number of these patients suffer from tinnitus that cannot be treated by conventional masking devices. In the paediatric population, UD has been linked to delayed speech and language development, and poorer academic performance.

Aims

1. To conduct a pilot study to investigate the use of cochlear implant (CI) in patients with UD with and without associated tinnitus.

2. To evaluate the results from CI on speech understanding in the most challenging scenarios, on self-perceived improvement of hearing, and on tinnitus suppression in patients with UD.

3. To investigate the localisation abilities of unilaterally deaf patients who received CI.

4. To investigate the factors that influence the outcomes of CI in UD, with particular focus on deafness duration and speech coding strategies.

5. To investigate the benefits of CI in the paediatric population with UD.
Methods

Speech perception was assessed using the Bamford-Kowal-Bench adaptive speech-in-noise (BKB-SIN) test in three spatial configurations: (1) speech and noise from the front, (2) speech from the front and noise from the hearing ear, and (3) speech from the CI side and noise from the hearing ear. Subjective benefits were assessed using the Speech, Spatial and Qualities of Hearing (SSQ) questionnaire and the Abbreviated Profile of Hearing Aid Benefit (APHAB). Localisation abilities were also evaluated using the A§E localisation setup. Tinnitus was assessed using the Tinnitus Reaction Questionnaire (TRQ). Results were evaluated pre-operatively and at 3, 6, 12 and 24 months post-surgery.

Results

Aim 1 (Chapter 3)

- CI improves speech understanding in noise significantly and as early as 3 months post-operatively.
- CI decreases tinnitus perception significantly when the speech processor is on.
- CI improves self-perceived hearing performance.

Aim 2 (Chapter 4)

- CI improves speech understanding in the most challenging scenarios and the benefits remain stable in the long-term.
- CI significantly increases self-perceived improvement of hearing and remains stable in the long-term.
- CI suppresses tinnitus significantly in the short- and long-term.
- Outcomes are not influenced by duration of deafness.
- Age at implantation does not influence the outcomes.
**Aim 3 (Chapter 5)**

- Localisation ability improves significantly with CI on.
- Localisation ability is not influenced by age at implantation, duration of deafness or gender.

**Aim 4 (Chapters 6 and 7)**

- Patients with more than 25 years duration of deafness benefit from CI use for the treatment of UD.
- The speech coding strategy FS4-p is preferred by unilaterally deaf CI users.
- FS4-p is perceived as providing the most natural and pleasant signal when compared with the normal hearing ear.

**Aim 5 (Chapter 8)**

- Children with post-lingual UD experienced improvement in speech understanding in noise similar to the adult group.
- Children with congenital UD did not show any sign of binaural hearing benefits when implanted post-lingually.
- Children with congenital UD show signs of binaural hearing benefits when implantation occurs at a young age.

**Conclusions**

Cochlear implantation is a viable treatment option for UD and provides significant improvements in terms of hearing performance, localisation ability, lowering of tinnitus perception and high subjective acceptance of the implant. Age at implantation and duration of deafness do not influence the benefits from CI in patients with UD.
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Statement of candidate contribution

The biographical details of the published work of this thesis are outlined below.

Chapter 3

Dayse Távora-Vieira collected and interpreted the results. Writing up of the manuscript was performed by Dayse Távora-Vieira and co-authors.

Chapter 4

Dayse Távora-Vieira designed and performed the experimental work as well as collected and interpreted the results. Writing up of the manuscript was performed by Dayse Távora-Vieira and co-authors.

Chapter 5

Dayse Távora-Vieira designed and performed the experimental work as well as collected and interpreted the results. Writing up of the manuscript was performed by Dayse Távora-Vieira and co-author.
Chapter 6


Dayse Távora-Vieira and Isabelle Boisvert designed the experimental work.

Dayse Távora-Vieira collected and interpreted the results. Writing up of the manuscript was performed by Dayse Távora-Vieira and co-author.

Chapter 7


Dayse Távora-Vieira designed and performed the experimental work as well as collected and interpreted the results. Writing up of the manuscript was performed by Dayse Távora-Vieira and co-author.

Chapter 8


Dayse Távora-Vieira collected and interpreted the results. Design of the experimental work and writing up of the manuscript was performed by Dayse Távora-Vieira and co-author.
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<tr>
<td>ABR</td>
<td>Auditory brain responses</td>
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<tr>
<td>APHAB</td>
<td>Abbreviated Profile of Hearing Aid Benefit</td>
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<td>BAHA</td>
<td>Bone-anchored hearing aid</td>
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<tr>
<td>BKB-SIN</td>
<td>Bamford-Kowal-Bench adaptive speech-in-noise</td>
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<tr>
<td>CI</td>
<td>Cochlear implant</td>
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<tr>
<td>CROS</td>
<td>Contra-lateral routing of signal</td>
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<tr>
<td>dB</td>
<td>Decibel</td>
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<td>dB H</td>
<td>Decibel hearing</td>
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<td>dB HL</td>
<td>Decibel hearing loss</td>
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<tr>
<td>EAS</td>
<td>Electrical and acoustic stimulation</td>
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<td>GLM</td>
<td>General linear model</td>
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<tr>
<td>HHIA</td>
<td>Hearing Handicap Inventory for Adults</td>
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<tr>
<td>HL</td>
<td>Hearing loss</td>
</tr>
<tr>
<td>HUI-3</td>
<td>Health Utilities Index 3</td>
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<tr>
<td>ILD</td>
<td>Interaural level difference</td>
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<tr>
<td>IOI-HA</td>
<td>International Outcome Inventory for Hearing Aids</td>
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<tr>
<td>ITD</td>
<td>Interaural time difference</td>
</tr>
<tr>
<td>MCL</td>
<td>Most comfortable level</td>
</tr>
<tr>
<td>MRI</td>
<td>Magnetic resonance imaging</td>
</tr>
<tr>
<td>OAE</td>
<td>Otoacoustic emissions</td>
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<tr>
<td>PTA</td>
<td>Pure tone average</td>
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<tr>
<td>RMS</td>
<td>Route mean square</td>
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<tr>
<td>SNR</td>
<td>Signal-to-noise ratio</td>
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<tr>
<td>SPL</td>
<td>Sound pressure level</td>
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<tr>
<td>SSQ</td>
<td>Speech Spatial and Qualities of Hearing</td>
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<tr>
<td>TRQ</td>
<td>Tinnitus Reaction Questionnaire</td>
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<tr>
<td>UD</td>
<td>Unilateral deafness</td>
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<tr>
<td>UHL</td>
<td>Unilateral hearing loss</td>
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<tr>
<td>VAS</td>
<td>Visual Analogue Scale</td>
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1. Introduction

1.1. History of cochlear implantation

Experiments to apply electrical stimulation to the human auditory system date back to 1800 when Count Alessandro Volta tried to stimulate his own auditory system. Almost 150 years later, Lundberg claimed to have achieved direct stimulation of the auditory nerve of an individual during surgery (Niparko, 2009). In 1957, Djourno and Eyries implanted two deaf patients with an electrode placed on the auditory nerve, a small inductor coil in the temporalis muscle and an external stimulator. According to Epstein (1987), these patients were able to discriminate simple words and hence their device is generally considered to be the first CI.

The CI as it is known today took its form in the early 1960s with the work of surgeons WF House and John Doyle and the electronic engineer James Doyle. They implanted two patients with a single wire electrode inserted into the inner ear. However, the local skin reaction that followed implantation lead to the failure of this device (Niparko, 2009). In 1964, F.Blair-Simmons experimented by inserting six electrodes into the cochlea of a deaf adult who was subsequently able to recognise several tunes. Similar experiments were also performed at about the same time by RB Michelson and Pialaux (Niparko, 2009).

The first widely available CI was a single-channel device implanted in over 1,000 adult patients between 1972 and the mid-1980s. In 1980, children as young as 2 years of age could receive a CI. In 1975, 13 patients who had received a single-channel device implanted by WF House or RB Michelson were submitted for comprehensive evaluation by the National Institutes of
Health (NIH) in the US. It was concluded that a single-channel device could assist with lip reading and quality of life, but not with speech perception. From that time onwards, most research efforts were directed towards the development of a multi-channel CI (Niparko, 2009).

Meanwhile, multi-electrode CIs were being developed by Graham Clark’s group at Melbourne University, a group at the University of California, San Francisco (UCSF), and the Hochmairs in Austria. These were later commercialised as Cochlear Corporation’s Nucleus device, the Advanced Bionics’ Clarion device, and the Med-El system, respectively. In 1984, the first multi-electrode device (example shown in figure 1.1) was introduced and shown to provide enhanced speech perception compared to a single-channel device. This was possible due to the extensive research on how to represent an incoming sound, overcoming the limited stimulation of the implant electrode (Wilson et al., 1991). The multi-electrode CI received FDA approval for adult use in 1985 and for children aged from 2 years old in 1990 (Niparko, 2009).

During the following years, advances in the early diagnosis of deafness, progress of technology for in speech processing strategies, and the refinement of surgical techniques resulted in cochlear implantation evolving into the treatment of choice for bilateral, profound HL. Multiple studies have demonstrated the advantages of early CI for hearing rehabilitation in children with bilateral deafness (Dettman et al., 2007; Niparko et al., 2010; Gilley et al., 2008). Sharma and colleagues (2005; 2007; 2009) reported that there were critical periods in child development in which the CI was crucial for the development of normal speech and language. Recently, Leigh et al. (2013) compared the long-term follow-up of children who had received cochlear
implantation before 12 months of age compared to those who were implanted at age between 13-24 months. Children implanted before 12 months displayed a speech and language growth that was comparable to normal hearing children. Today, CIs are accepted as the only functional bionic device enabling children with bilateral deafness to achieve speech and language development within the normal range.

The success and advances in CI in post-lingual adults and congenitally deaf children resulted in a broadening of the indications. Patients with residual hearing but greater self-perceived handicap started to receive CIs (Cullen et al., 2004). Implant candidacy was also extended to patients with moderate to severe bilateral HL (Parkinson et al., 2002). Bimodal hearing, i.e. the combination of acoustic hearing in one ear and CI in the other ear, became the focus of several studies which showed superior results compared to monaural hearing through CI (Ching et al., 2004; 2005; 2006; Dunn et al., 2005).

Extending the candidacy criteria even further, patients with residual hearing in the low frequencies and profound HL in the high frequencies became a focus for further research. Combined electrical and acoustic stimulation systems (EAS) were developed. During the early 2000s, several studies reported the various benefits of EAS on speech perception scores compared to CI alone or to acoustic hearing alone (von Ilberg et al., 1999; Gantz and Turner, 2003; Gantz et al., 2005; Skarzynski et al., 2003; Kiefer et al., 2004; James et al., 2005; Gantz et al., 2006; Gstoettner et al., 2008).

1.2. Cochlear implantation for UD
With the expansion of CI candidacy criteria to include patients with moderate to severe HL and assisted hearing in the contra-lateral ear, the next research
focus became the use of CI for UD. The first report on CI for UD was published by Van de Heyning et al. (2008). A group of 22 adult patients with UD and highly disturbing tinnitus was included in their study. Based on previous literature reporting that CI had successfully suppressed tinnitus perception in patients with bilateral severe-profound loss (Ito, 1997; Ito and Sakakihara, 1994; Mo et al., 2002), the primary goal of Van de Heyning’s study was to investigate the effect of electrical stimulation on unilateral tinnitus. CI was found to significantly decrease tinnitus perception in these cases.

Figure 1.1. Cross-section showing a multi-channel modern CI in-situ (from Med-El)
A follow-up study designed to look at the hearing implications of CI in UD (Vermiere et al. (2009) reported the hearing outcomes for a group of 20 patients comprising of 11 subjects with normal hearing in the opposite ear and nine with some degree of HL. All patients reported substantial benefits
when bimodal was compared to monaural hearing. There were also significant improvements in self-perceived benefits of binaural hearing on speech understanding, quality of hearing and spatial hearing.

In 2010, two more studies on the implications of bimodal hearing in UD were published:

(1) Arndt et al. (2010) compared the outcomes from bone-anchored hearing aids (BAHA), conventional contra-lateral routing of signal (CROS) and CI in UD. Eleven patients received a CI after trialling the CROS and BAHA. Speech comprehension was measured in the unaided condition, aided with CROS, aided with BAHA, and 6 months post-CI surgery. Localisation, tinnitus disturbance and subjective improvement of hearing were also assessed. Speech comprehension was significantly improved with CI when speech was presented to the implanted side and noise to the normal hearing ear. There was also a significant difference between the treatment modalities when speech and noise were presented from the front. CI was significantly better than CROS and BAHA when noise was presented from the implanted side, but there was no improvement when compared to the unaided condition. Localisation error was significantly reduced with CI.

The subjective, self-rated hearing performance as measured by the SSQ questionnaire (Gatehouse and Noble, 2004) was significantly better with CI compared to unaided, CROS and BAHA for the speech and spatial subsections. The Health Utilities Index 3 (HUI-3) and the International Outcome Inventory for Hearing Aids (IOI-HA) questionnaires were also used to evaluate subjective improvement in daily life. HUI-3 scores for the CI condition were significantly better than BAHA and CROS. CI achieved
higher scores on all sub-domains of the IOI-HA compared to the other two devices. The sub-domain benefit, satisfaction and QoL all showed significant improvement with CI.

Tinnitus intensity was evaluated using a visual analogue scale (VAS) and the score was significantly reduced 6 months after implantation when the CI was on. Arndt et al. (2010) concluded from their study that CI was the better option for improvement of speech perception in noise and localisation compared to BAHA and CROS.

(2) A second study by Buechner et al. (2010) reported the outcomes for a group of 5 patients with UD and tinnitus. One patient judged that the benefits from CI did not outweigh the inconvenience of wearing the speech processor. Another patient could not wear the implant on a full-time basis due to debilitating tinnitus.

For the other three patients, CI resulted in significantly better speech perception scores when speech was presented from the front and noise from the normal hearing ear. When noise was presented from the front or from the implanted ear, CI did not provide any benefit. Buechner et al. (2010) concluded that although CI may be beneficial for UD, more research was necessary to establish the selection criteria.

1.3. Implications of UD
As discussed by Gray et al. (2009), the extent of the disability due UD is commonly underestimated, even ignored or frequently denied appropriate importance. This is usually due to the patient compensating for this disability by relying on their good hearing ear.
Binaural hearing is the ability of the auditory system to process and integrate inputs from both ears resulting in the precise perception of sound in space and time. It is well established that binaural hearing is essential for spatial hearing and for improved speech perception in noise (Arsenault and Punch, 1999; Bronkhorst and Plomp, 1988; Van Deun et al., 2009; Grothe et al., 2010). The advantages of binaural hearing relate to binaural summation, the head-shadow effect and the squelch effect (Dillon, 2002). Binaural summation refers to hearing the acoustic information with two ears. The redundant information arriving to both ears results in improved speech intelligibility (Bronkhorst and Plomp, 1990). In normal hearing subjects the head acts as a barrier and sounds from different directions reach each ear with different intensities, giving rise to the so called head shadow effect. Head shadow effect allows the individual to take advantage of 2 different signal-to-noise ratios and therefore helps in sound localisation and in understanding speech in noise. Finally, binaural squelch refers to the brain’s ability to separate signal and noise coming from two ears using interaural time latency, level and spectral differences (Carhart, 1965). Clearly, all of these mechanisms are missing when a patient loses their hearing on one side.

Binaural hearing (top panel) allows the head to act as a barrier and sounds from different directions will reach each ear with different intensities. In monaural hearing (lower panel) the sounds cannot be spatially separated, thus reducing the patients’ ability to discriminate speech and sound in space (Figure 1.2).
Figure 1.2. Schematic view of binaural versus monaural hearing (from Med-El)

Historically the handicap from UD was hidden by the ability of many patients to cope in the majority of listening situations by using several strategies. These include avoidance of noisy environments, positioning the good ear towards the target speech, and turning the head towards the sound of interest.
(Wie et al., 2010; Gray et al., 2009). In fact, several workers in the field pointed out that UD was also ignored by the hearing health professionals such as the audiologists and otolaryngologists (Northern and Downs, 1978).

In the 1980s, several studies were carried out to evaluate the implications of unilateral hearing loss (UHL) in children. UHL in children is linked to delayed speech and language development (McKay et al., 2008; Lieu et al., 2010) and with subsequent psychosocial problems (Tharpe and Sladen, 2008).

Shepard et al. (1981) reviewed the academic records of a large number of children with minimal or UHL. Children with UHL were found to have a reduced vocabulary and structure their sentences in a simpler form. Bess and Tharpe (1984; 1986) reviewed the medical and academic records of 60 children with UHL and reported that 35% had failed at least one grade and 20% had shown some kind of behavioural problems as reported by their teachers. Approximately 50% of these children needed academic assistance and they usually presented with poor performance on localisation ability and syllabic recognition tasks compared to their normal hearing peers.

Stein (1983) reported that 37% of children with UHL experienced difficulties with interpersonal and social functioning. The most common psychosocial impacts were withdrawal, aggression, frustration and isolation (Giolas and Walk, 1967). Borton et al. (2010) also found that children with UHL had significantly lower social functioning skills. In addition, their listening effort was significantly higher than for normal hearing children (Bourland-Hicks and Tharpe, 2002).
In adults, the two major consequences of UD are the lack of spatial hearing and reduced speech intelligibility in the presence of background noise and when speech is heard from the impaired ear (McLeod et al., 2008). These patients lack binaural squelch, i.e. the ability to separate speech from noise coming from a different direction. Priwin et al. (2007) reported on 57 subjects aged 3-80 years with UHL. They found that 14% of subjects had severe difficulty in a group conversation, while for 19% of subjects the ability to keep a conversation in the presence of traffic noise was severely affected. For 26% of participants, conversing in quiet was also difficult.

Recently, Vicci de Araujo et al. (2010) investigated the hearing handicap in 52 adults with varying levels of UHL using the Hearing Handicap Inventory for Adults (HHIA). In the emotional and social subscales, 73% reported some level of handicap, while 52% reported difficulty at the cinema and 40% reported difficulty on the phone.

1.4. Incidence of UHL

Incidence estimates of UHL is of great variety due to several factors such as study population, hearing assessment methods and the definition of HL, which commonly varies from study to study. The incidence estimates in newborns range 0.8-2.7 per 1000 (Dalzell et al., 2000; Johnson et al., 2005; Watkin and Baldwin, 1999; White et al., 1994). This increases substantially in school-aged children ranging from one to 56 per 1,000 (Bess et al., 1998; Niskar et al., 1998; Widen et al., 2000). There is evidence that the incidence increases to one in five adolescents as the children grow older (Mehra et al., 2009; Shargorodsky et al., 2010). This is probably related to lack of appropriate follow-up after newborn hearing screening programs.
Congenital UD is estimated to affect one in 8000 children (Mehl et al., 1992). There is no much literature reporting the incidence of UD in adult population. It is estimated that 60,000 people acquire UD in the United States per year, and in the UK, it is estimated that 9000 people develop profound UD each year (British Association of Otorhinolaryngologists, 2002).

1.5. Causes of UD
UD may be congenital or acquired at any stage of life. The most common etiologies of acquired UD include ototoxicity, inner ear infection, Meniere’s disease, inner ear otosclerosis, head trauma or trauma to the ear, circulatory disorders, genetic and acoustic neuroma (Schreiber et al. 2010). The most common causes of congenital UD are anomalies of the inner ear, cochlear nerve deficiency, mumps, cytomegalovirus infection, meningitis, auditory neuropathy spectrum disorder and unknown (Usami, 2014).

1.6. Management options for UD
Until recently, UD was not considered to be an impairment that warranted intervention. Following diagnosis and the exclusion of benefits from conventional hearing amplification, such patients were usually ignored or advised to use coping strategies. Gradually, more patients are being offered assistive listening devices such as FM systems or hearing devices that transfer the signal from the deaf ear to the better hearing side so called CROS-systems. Conventional CROS hearing aids comprises of a microphone placed in the deafened ear and a hearing aid placed in the hearing ear. CROS hearing aids have been proposed for the management of UD for over 40 years (Harold and Dodds, 1966; Green et al., 1967). Harold and Dodds (1966) reported that the success of CROS in UD is related to the patients’ motivation.
and the handicap imposed by the UD. Early studies demonstrated that CROS could improve speech discrimination in noise when speech was presented to the deafened ear but it was detrimental when speech was presented to the normal hearing ear and could not improve sound localisation (Lotterman and Kasten, 1971; Markides, 1979).

Among the reasons for patients’ dissatisfaction were the presence of the wire and the use of earpiece in the normal hearing ear (Hol et al., 2009). The improvement in technology has allowed CROS system to move away from the wired hearing devices to a wireless system and more refined sound processing (Figure 1.3). However, the patients still report discomfort, poor sound quality, and limited perceived benefit (Snapp et al., 2012). Although it helps patients to hear from the deaf side, it reportedly provides only limited improvement in hearing (Lin et al., 2006) and it cannot provide binaural hearing benefits including sound localisation (Kunst et al., 2008). CROS-aids are the easiest and cheapest rehabilitation device for UD and is thus routinely offered to patients prior to any implantable device.
Figure 1.3. Schematic view of conventional CROS system (from Phonak)

A transmitter sitting on the impaired will capture the sound and transmit it to the hearing aid sitting in the normal hearing ear. The patient relies on the hearing ear and binaural hearing cannot be achieved.

For more than a decade now, percutaneous bone conduction systems such as the BAHA (Cochlear Pty) or Ponto-system (Oticon Medical) have been used for rehabilitation of UD. More recently, new transcutaneous bone conduction hearing implants have been developed such the BoneBridge (Med-El), the Sophono and BAHA Attrack (Cochlear Pty). These bone conduction systems comprise of an internal and an external component which connects to each other through a percutaneous abutment (BAHA and Ponto) or through transcutaneous magnet attraction (BoneBridge, Sophono, BAHA Attrack). The external component captures the sound which is transmitted to the functional contra-lateral cochlear through the vibration of the bone (Figure 1.4).
Figure 1.4.  Schematic view of the bone conduction implant (from Cochlear)
Bone conduction implant transmits the sound captured from the impaired ear to the
normal contra-lateral cochlea.

Similarly to conventional CROS, these bone conduction hearing devices are
limited to capturing the sound from the deaf ear and transmitting it to the
contra-lateral functional inner ear via the vibration of the bone. BAHA has
merit for improving speech perception and for providing subjective benefits
(Yuen et al., 2009; Wazen et al., 2010; Linstrom et al., 2009; Hol et al., 2010;
House et al., 2010). However, binaural hearing and subsequent sound
localisation are not achievable with bone conduction or CROS devices
(Kunst, 2008, Hol et al., 2010).

Animal and electrophysiological models have indicated that the
pathophysiological mechanism of tinnitus comprises of auditory and
non-auditory centres. There is a general acceptance that tinnitus might be
triggered by injury to inner ear which leads to modification in the balance of
excitatory / inhibitory neuronal activities in the central auditory system (Kaltenbach, 2011). For patients with UD and symptomatic tinnitus, conventional CROS and bone conduction systems do not provide benefits for tinnitus control as they do not overcome auditory deprivation of the deaf ear.

1.7. Aims of this study
The most recent research suggests that CI is the most beneficial treatment option for UD. However, the literature on this subject is still quite sparse and more evidence is required. There is no published literature on the factors that could predict outcomes, the candidacy criteria remain to be defined, and there are no reports on the use of CI in the paediatric population with UD. It is also unknown whether the unilaterally deaf CI users require specific rehabilitation protocols. To address these issues, a prospective study was designed with the following aims:

1. To conduct a pilot study to investigate the use of CI in patients with UD with and without associated tinnitus. (Addressed in Chapter 3)

2. To evaluate the results from CI on speech understanding in the most challenging scenarios, on self-perceived improvement of hearing, and on tinnitus suppression in patients with UD. (Addressed in Chapter 4)

3. To investigate the localisation abilities of unilaterally deaf patients who received CI. (Addressed in Chapter 5)

4. To investigate the factors that influence the outcomes of CI in UD with particular focus on deafness duration and speech coding strategies. (Addressed in Chapters 6 and 7, respectively)
5. To investigate the benefits of CI in the paediatric population with UD.

   (Addressed in Chapter 8)
2. Materials and methods

2.1. Study population
Twenty-eight adult and four children with unilateral, sensorineural severe to profound HL [4-frequency pure tone average (PTA) 0.5-4kHz >70dB HL] were enrolled in this study, of which 13 adults had tinnitus. The better hearing ear had a PTA<sub>0.5-4kHz</sub> of ≤30dB. The detailed demographic data for these subjects is shown in Table 2.1. All subjects were implanted with a 31.5mm or 28mm electrode array and received an OPUS 2 speech processor (MED-EL GmbH, Innsbruck). The surgical procedure used was a hearing preservation technique as described by Rajan et al., (2012).

2.2. Pre-operative assessment
Standardised pre-operative evaluation of the subjects included high-resolution computed tomography (CT) and magnetic resonance imaging (MRI) of the temporal bones and brain to rule out the presence of any inner ear anomalies or cochlear nerve pathologies that might restrict or interfere with electrical stimulation by a CI.

2.2.1. Adult assessment
Audiological workup included immittance measures, audiometry, and speech discrimination in quiet using the Arthur Boothroyd (AB) word list (Boothroyd, 1968). A hearing aid was fitted to the poorer ear if any functional hearing was present. If this was unsuccessful the patient was considered for a CI. To participate in the study, a trial with a conventional CROS hearing aid and a BAHA were undertaken.
<table>
<thead>
<tr>
<th>Subject</th>
<th>Duration of deafness (years)</th>
<th>Age at implantation (years)</th>
<th>Ear</th>
<th>Aetiology</th>
<th>PTA: non-implanted ear * (dB)</th>
<th>PTA: implanted ear * (dB)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 (adult)</td>
<td>3</td>
<td>45</td>
<td>R</td>
<td>Sudden HL</td>
<td>10</td>
<td>74</td>
</tr>
<tr>
<td>2 (adult)</td>
<td>2</td>
<td>56</td>
<td>L</td>
<td>Post-traumatic</td>
<td>18</td>
<td>90</td>
</tr>
<tr>
<td>3 (adult)</td>
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<td>79</td>
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<td>84</td>
</tr>
<tr>
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</tr>
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<td>Post-traumatic</td>
<td>3</td>
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</tr>
<tr>
<td>8 (adult)</td>
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<td>40</td>
<td>R</td>
<td>Post-traumatic</td>
<td>34</td>
<td>&gt;110</td>
</tr>
<tr>
<td>9 (adult)</td>
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<td>46</td>
<td>R</td>
<td>ISSNHL</td>
<td>32</td>
<td>95</td>
</tr>
<tr>
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<tr>
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<tr>
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<tr>
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<td>R</td>
<td>Ototoxicity</td>
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<tr>
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<td>39</td>
<td>R</td>
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<tr>
<td>Subject</td>
<td>Age</td>
<td>PTA</td>
<td>Side</td>
<td>Diagnosis</td>
<td>Score</td>
<td>Handicap</td>
</tr>
<tr>
<td>---------</td>
<td>------</td>
<td>-----</td>
<td>------</td>
<td>-------------</td>
<td>-------</td>
<td>----------</td>
</tr>
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<td>&gt;110</td>
</tr>
<tr>
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<td>76</td>
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</tr>
<tr>
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<td>4</td>
<td>R</td>
<td>Unknown</td>
<td>19</td>
<td>&gt;110</td>
</tr>
<tr>
<td>24</td>
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<td>Unknown</td>
<td>21</td>
<td>&gt;110</td>
</tr>
<tr>
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<td>Adult</td>
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<td>R</td>
<td>Unknown</td>
<td>24</td>
<td>95</td>
</tr>
<tr>
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<td>L</td>
<td>ISSNHL</td>
<td>32</td>
<td>80</td>
</tr>
<tr>
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<td>ISSNHL</td>
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<td>92</td>
</tr>
<tr>
<td>28</td>
<td>Adult</td>
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<td>R</td>
<td>ISSNHL</td>
<td>21</td>
<td>87</td>
</tr>
<tr>
<td>29</td>
<td>Child</td>
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<td>R</td>
<td>Unknown</td>
<td>15</td>
<td>&gt;110</td>
</tr>
<tr>
<td>30</td>
<td>Child</td>
<td>4.5</td>
<td>R</td>
<td>Unknown</td>
<td>10</td>
<td>&gt;110</td>
</tr>
<tr>
<td>31</td>
<td>Child</td>
<td>6.9</td>
<td>R</td>
<td>Unknown</td>
<td>5</td>
<td>&gt;110</td>
</tr>
<tr>
<td>32</td>
<td>Child</td>
<td>0.1</td>
<td>R</td>
<td>Meningitis</td>
<td>13</td>
<td>&gt;110</td>
</tr>
</tbody>
</table>

ISSNHL = Idiopathic sudden sensorineural hearing loss  
PTA: at 0.5, 1, 2 and 4 kHz  
* Before cochlear implantation
2.2.2. Paediatric pre-operative assessment

The pre-surgical audiological evaluation included acoustic immittance measures, audiometry, otoacoustic emissions (OAE), and auditory brainstem responses (ABR). A trial with a conventional CROS hearing aid and a FM system were provided to all children. Detailed discussion about the risks and benefits of cochlear implantation in UD took place over several appointments with a multi-disciplinary team including a psychologist, audiologist, speech pathologist, and an otologist. Parents were provided with all of the recent literature on cochlear implantation for UD in adults and in children. The parents gave written informed consent for inclusion of the children’s data in this study.

The inclusion criteria for this study were: unilateral sensorineural severe to profound HL [4-frequency pure tone average (PTA) $0.5-4\text{kHz} > 70\text{dB HL}$]; normal hearing in the contra-lateral ear (PTA $< 20\text{dB HL}$); no history of delayed motor development, and normal cognitive and psychological development as assessed by a psychologist. It was a requirement that the child had an intact vestibulocochlear nerve as verified by MRI, and a strong family involvement in the rehabilitation process.

Different measures were adopted according to the child’s age. Speech perception in noise testing was performed using the BKB-SIN test (Bench et al., 1979) where possible. All the children were fitted with a FM system to assist with their auditory training at home, ensuring input from the CI alone.

2.2.3. Speech perception testing

Speech perception testing was performed using the adaptive (BKB-SIN) test. This investigates the signal-to-noise ratio (SNR) needed to achieve a score of
50% of words correct. Tests were performed in a free-field with the subject seated one meter away from loudspeakers located at angles of 0, -90 and +90 degrees (Figure 2.1). The spatial configurations used for speech testing were: S₀/N₀ (speech and noise presented from the front); S₀/Nₜₑ (speech presented from the front and noise to the normal hearing ear); and Sₕₑ/Nₜₑ (speech presented to the implanted ear and noise to the side of the normal hearing ear). The spatial configuration Sₕₑ/Nₜₑ was chosen on the basis of it being the most challenging situation, as described by the patients.

Figure 2.1. Spatial configuration of the speakers

2.2.4. Sound localisation
Localisation testing was performed using the A§E localisation test software. This presents a narrow band noise of 1/3rd octave centred around 4kHz simultaneously through two loudspeakers that were placed at -60 and 60 degrees from the listener. The noise from the speakers was correlated with an interaural time difference (ITD) of zero. The presentation level differs from each speaker simulating an interaural level difference (ILD), hence creating the illusion of a sound source being localised somewhere on the azimuth.
between the two loudspeakers, as described previously (http://www.otoconsult.com/asse/Localization.aspx).

Thirteen loudspeakers numbered -6 (left) to 6 (right) were set up in a semicircle and at 10 degrees interval. Using this arrangement, 11 loudspeakers were ‘sham’ loudspeakers (numbered from -5 to 5) and were placed in between two real loudspeakers, which were numbered as -6 and 6 (Figure 2.2). Testing was performed in a soundproof booth. The subject sat directly facing speaker number 0 and did not move their head.

For each presentation the software randomly selects an ILD from the following series: -30, -20, -10, -4, 0, +4, +10, +20, +30dB. The presentation level is 60dB HL at one loudspeaker and 60, 56, 50, 40 or 30dB at the other, depending on the ILD chosen. The number of test items is 33. Stimuli with intensity differences of -30, -20, -10, 10, 20 and 30dB are presented three times each, while stimuli with intensity differences of -4, 0 and 4dB are presented five times each.

Figure 2.2. Localisation testing setup
The participants were asked to state the speaker number they thought the sound was coming from. The subjects’ responses were entered into the computer and the software calculated the median values and the test error. The route mean square (RMS) error was used as a measure of the subjects’ localisation performance. A lower RMS error represents better localisation skills.

Each subject performed the localisation testing in two listening conditions – monaural hearing (normal acoustic hearing alone) and binaural hearing (acoustic hearing and CI activated). The order of the test was randomised. All subjects had at least 6 months’ experience with their CI. Testing was performed with the subjects’ everyday program settings, which accounts for subjective loudness balancing between the normal ear and the CI stimulation.

A normative study was carried out using 30 normal hearing subjects to obtain reference data. The subjects were recruited at the Eargroup (Antwerp, Belgium) and had an average age of 27 years (SD 6,8 years; range 15-41 years) with normal hearing (hearing thresholds of 20dB HL or better in the frequency range of 0.25 to 4kHz and no otological history. The subjects underwent the localisation test as described above. Each ILD thus yielded 30 responses, namely the loudspeakers as identified by the 30 listeners. The distribution of these responses is summarised by the average and standard deviation. Table 2.2 shows the normative data.

<table>
<thead>
<tr>
<th>ILD</th>
<th>-30dB</th>
<th>-20dB</th>
<th>-10dB</th>
<th>-4dB</th>
<th>0dB</th>
<th>4dB</th>
<th>10dB</th>
<th>20dB</th>
<th>30dB</th>
</tr>
</thead>
</table>

Table 2.2. Results of the A§E® localisation test in 30 normal hearing subjects

For each ILD (column headers), the average responses (number of the sham loudspeaker) are given, together with the lower (Min) and upper (Max) limit of the 95% confidence interval.
2.2.5. **Subjective testing – SSQ questionnaire**

Subjective assessment of hearing was determined using the standardised SSQ questionnaire (Gatehouse and House, 2004). Each question has a score of 0 to 10. A higher score on the SSQ indicates better subjective perception of hearing speech, spatial orientation and quality of hearing. All subjects were asked to complete the SSQ pre-surgically and at 3, 6, and 12 months post-CI. The long-term benefit was verified at 24 months.

<table>
<thead>
<tr>
<th>Min</th>
<th>-6,0</th>
<th>-6,0</th>
<th>-4,6</th>
<th>-2,6</th>
<th>-1,0</th>
<th>0,0</th>
<th>2,3</th>
<th>3,4</th>
<th>3,7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Av.</td>
<td>-5,5</td>
<td>-5,2</td>
<td>-3,2</td>
<td>-1,2</td>
<td>0,2</td>
<td>1,7</td>
<td>3,5</td>
<td>4,9</td>
<td>5,1</td>
</tr>
<tr>
<td>Max</td>
<td>-4,0</td>
<td>-3,3</td>
<td>-1,6</td>
<td>0,7</td>
<td>1,6</td>
<td>3,4</td>
<td>5,0</td>
<td>6,0</td>
<td>6,0</td>
</tr>
</tbody>
</table>

2.2.6. **Subjective testing – APHAB**

The APHAB is a self-assessment tool comprised of 24 questions that indicates the level of difficulty a person with a HL is faced with in various everyday listening situations. It aims to calculate the benefit of amplification, as perceived by the subject, by comparing the subjects’ reported difficulty in an unaided and aided condition. The questionnaire has four subscales – ease of communication, reverberation, background noise and aversiveness (Cox and Alexander, 1995). The APHAB was completed pre-surgically and at 12 months post-implantation with a CI to determine the subjective benefit from CI.

2.3. **TRQ**

Tinnitus disturbance was assessed using the TRQ [Wilson et al., 1991]. The TRQ is composed of 26 questions designed to establish the effects of tinnitus over the past week on lifestyle and general well-being using a scale from 0 (not at all) to 4 (almost all of the time). The final total score can range from 0 through to 104. All subjects were asked to complete the TRQ pre-surgically.
and at 3, 6, and 12 months post-CI activation. The long-term effect was also measured at 24 months post-implantation.

### 2.4. Subjective evaluation of speech coding strategy

Three months post-CI activation, after giving written consent to participate in this study, 13 adult subjects were provided with two maps called program 1 (P1) and program 2 (P2) that differed only in the coding strategy, FS4 or FS4-p, respectively. The subjects were blind to the different settings P1 and P2, and were not provided with any information regarding the differences between the two programs. The subjects were requested to alternate between the two maps daily for two consecutive weeks and to complete a non-standardised 10 point scale questionnaire at the end of each day, and return it to the audiologist. The patients’ scores were averaged for each question for each program (P1 and P2). The questionnaire comprised of five questions:

1. How similar is the sound from the CI to the other ear? In this question the scale varied from 1 (“very different”) to 10 (“very similar”).

2. How is the clarity of sounds? The scale varied from 1 (“very unclear”) to 10 (“very clear”).

3. How easy is it to hear in quiet? The scale varied from 1 (“very difficult”) to 10 (“very easy”).

4. How easy is it to hear in noise? The scale varied from 1 (“very difficult”) to 10 (“very easy”).

5. How do you like the sound? The scale varied from 1 (“not at all”) to 10 (“very much”).
Speech perception in noise test was performed at the end of the two weeks. The sequence of the test (P1/P2) was randomised. The audiologist performing the test was blind to the settings of the speech processor.

2.5. Auditory training
Intensive auditory training was provided for all participants. The rehabilitation methods were thoroughly discussed with each individual prior to surgery and enrolment in this study in order to ensure good compliance and strong commitment to the training. Participants agreed to commit 30-60 minutes per day to auditory training. Auditory training started at week 2 or 3 after the CI was switched on.

All subjects were provided with an audio cable that connected the speech processor to an audio source. A special map – named the direct audio input map – was created for each subject at week 2 or 3, depending on the individual’s progress regarding the tolerance of the electrical stimulation. To establish the most comfortable levels (MCLs) of the direct audio input map, a series of progressive maps was created. The speech processor was then connected to an audio player and the subject was asked to listen to a speech signal bilaterally using a standard earphone in the good hearing ear. The subject would select the map that best matched the loudness level of the contra-lateral good hearing ear.

Once the MCLs were established the subjects were provided with pre-recorded material comprising of meaningful English words, such as numbers, colours, animals, fruits and vegetables. A printed copy of the material was also provided. The subjects were asked to work on the material
during the week. When the subjects were able to discriminate the words, a list of pre-recorded meaningful sentences was provided.

The next step in the auditory training was to listen to audiobooks. These varied in difficulty of the vocabulary, sentence length and reading speed rate. The subjects’ books were selected based on their ability to discriminate sounds. Those with greater difficulties were provided with books with easy vocabulary and known themes such as nature, the planets, etc; and with a reading speed rate as low as 40 words per minute. Books with a more complex vocabulary and higher reading speed rate were provided to subjects as their speech discrimination scores improved. Visual cues were allowed at the beginning of the program but removed as the subjects progressed. To ensure that all subjects received similar auditory training, a computer was available at the implant centre for those who did not have access to a computer or CD player at home.

A support group was established by the implant audiologists to allow recipients to share their experiences and difficulties and to help manage possible problems. The subjects meet every 3 months.

2.6. Statistical analyses
Repeated measure ANOVA was used to determine the effect over time of the three spatial setups of the adaptive BKB-SIN test, the three SSQ subscales, and the effects of tinnitus on the TRQ. Post-hoc pairwise comparisons using paired sample t-test was performed to determine the effect between test intervals in the BKB-SIN, the SSQ, and the TRQ. Paired sample t-tests were also performed to analyse for differences between pre-operative and 12 months post-operative testing on the APHAB subscales and the Global scale.
The independent-samples t-test was used to determine a significant effect of age at implantation, gender and the duration of deafness between age groups. Three-way repeated measure (RM) ANOVA was used to determine the effect of time, age at implantation, and duration of deafness as well as their interaction on the three spatial setups of the adaptive BKB-SIN test, the three SSQ subscales, and the TRQ scores. To test a possible influence of age and duration of deafness as well as their interaction at the single tested intervals, a multivariate general linear model (GLM) was performed for all outcome variables. All p-values are results of 2-sided tests. Paired sample t-tests were used to examine differences between CI off and CI on in localisation. Stratified analyses using paired sample t-test were performed to test the influence of gender, age (36-55 years vs 56-73 years), and the duration of deafness (3 months-10 years vs 10-39 years) on localisation. The Kolmogorov-Smirnov test was used to determine the data distribution before data analyses. The independent-samples t-test was used to investigate for significant effects of age, gender and duration of deafness between age groups. Wilcoxon signed-rank test was used to determine the difference between the coding strategies for all five questions of the questionnaire. A p-value of <0.05 was considered statistically significant. IBM SPSS Statistics 19 (IBM, Armonik, New York) software was used for the data analyses. Graphs were created using Microsoft Office Excel 2010 (http://www.microsoft.com).
3. Cochlear implantation for UD with and without tinnitus – a case series


3.1. Abstract
This pilot study aimed to investigate the benefits of cochlear implantation in patients with UD with and without tinnitus.

Nine post-lingually deafened adult subjects with UHL with and without tinnitus ipsilaterally, and functional hearing in the contra-lateral ear were enrolled in this study. Speech perception in noise was tested using the BKB-SIN presented at 65dB SPL (sound pressure level). The SSQ scale was used to evaluate the subjective perception of hearing outcomes and the TRQ assessed the effect on tinnitus.

All patients were implanted with the Med-El Flex soft electrode. CI was successful for all nine patients, with improvement of speech recognition in noise, self-perceived improvement of hearing and for tinnitus control.

3.2. Introduction
Patients with UHL who cannot benefit from hearing aids have been historically left without the benefit of binaural hearing. They usually report difficulties understanding conversation coming from the poorer-hearing side, especially in noisy environments and they have no ability to localise sounds. These difficulties experienced by subjects with UD are due to the lack of binaural effects (head shadow, squelch and summation effect) as described by Vermeire & Van de Heyning (2009). CROS and bone-conduction hearing
device, such as BAHA, are the current established management options for UD. Although these treatments options have their merits in improving speech perception and providing subjective benefits (Yuen et al., 2009; Wazen et al., 2010; Linstrom et al., 2009), binaural hearing is not achievable (Hol et al., 2010), and sound localisation remains poor (Wazen et al., 2010). For patients with UD and symptomatic tinnitus, conventional CROS and BAHA systems do not provide benefits for tinnitus control as they do not overcome the auditory deprivation of the deaf ear.

The use of cochlear implantation in patients with UD has been reported recently. Van de Heyning et al. (2008) used CI to treat individuals with UD who had severe unilateral tinnitus as the chief complaint. It was found that CI was successful in decreasing or totally inhibiting tinnitus perception. Vermeire & Van de Heyning (2009) reported that cochlear implantation in patients with UD and tinnitus also provided substantial improvement in speech understanding and subjective improvement in daily life. Positive outcomes were also reported by Buechner et al. (2010). A comparison of outcomes achieved with traditional CROS hearing aids, BAHA, and CI in the rehabilitation of UD was recently reported by Arndt et al. (2010), with a significantly superior improvement of hearing performance of the CI. However, several questions remain when it comes to understanding the effects of cochlear implantation for UD.

In patients with bilateral severe to profound sensorineural HL or deafness, the duration of the HL/deafness is considered as a predictive factor for implant performance, and patients with shorter duration tend to achieve better outcomes (Green et al., 2007). Is this also applicable for UD? We subsequently present a case series of nine unilaterally deaf patients with
various periods of deafness duration who received a CI for UD with or without tinnitus.

3.3. **Materials and methods**
The population, audiological and otological evaluation of the patients, statistical analysis are described in Chapter 2.

3.4. **Results**
This report includes nine subjects with post-lingual UHL/deafness characterised by a pure-tone average (0.5, 1, 2 and 4kHz) of 70dB or greater. Duration of deafness varied from 6 months to 40 years. Contra-lateral hearing was normal or close to normal with a group pure-tone average of 16dB.

All subjects displayed an improvement of speech perception in noise in the “CI on” condition in comparison to the “CI off” condition in at least 3 of the spatial configurations, more evident at the $S_0/N_0$ and $S_{CI}/N_{HE}$ spatial settings (Table 3.1). It is important to note that there was no deterioration of the SNR loss when the noise was presented to the CI side for seven of the nine subjects. The data represent the results achieved at 3 months post-cochlear implantation.
Table 3.1. BKB-SIN SNR values pre-, and 3 months post-cochlear implantation, the mean and SD for all spatial condition

<table>
<thead>
<tr>
<th>ID</th>
<th>S0/N0 SNR (dB) Baseline</th>
<th>3 months</th>
<th>Improvement</th>
<th>S0/NCI SNR (dB) Baseline</th>
<th>3 months</th>
<th>Improvement</th>
<th>S0/NHE SNR (dB) Baseline</th>
<th>3 months</th>
<th>Improvement</th>
<th>SCI/NHE SNR (dB) Baseline</th>
<th>3 months</th>
<th>Improvement</th>
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<tr>
<td>S1</td>
<td>6</td>
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<td>3.5</td>
<td>-3</td>
<td>-5</td>
<td>2</td>
<td>-1</td>
<td>-4.5</td>
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<td>3</td>
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</tr>
<tr>
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<td>4</td>
<td>-3</td>
<td>7</td>
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<td>-5</td>
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<td>0</td>
<td>3</td>
<td>0</td>
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<td>0</td>
<td>3</td>
<td>5</td>
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<td>5</td>
</tr>
<tr>
<td>S6</td>
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<td>0.5</td>
<td>5</td>
<td>-1.5</td>
<td>6.5</td>
<td>3</td>
<td>-4.5</td>
<td>7.5</td>
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<tr>
<td>S7</td>
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<td>7</td>
<td>3.5</td>
<td>3.5</td>
<td>-4.5</td>
<td>-4</td>
<td>-0.5</td>
<td>-3</td>
<td>-3.5</td>
<td>0.5</td>
<td>3</td>
<td>-2</td>
<td>5</td>
</tr>
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<td>S9</td>
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<td>-1</td>
<td>0</td>
<td>-4</td>
<td>-3.5</td>
<td>-0.5</td>
<td>-2</td>
<td>-4.5</td>
<td>2.5</td>
<td>-1.5</td>
<td>-2.5</td>
<td>1</td>
</tr>
<tr>
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<td>3</td>
<td>-3</td>
<td>-3</td>
<td>1</td>
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<td>2</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>
It was surprising for the investigators that even subject 1, who proceeded with CI purely because of the severity of his tinnitus, achieved an overall improvement of his hearing abilities. All patients are full-time wearers and say that the implant provides them with what they describe as “full hearing”.

All subjects accepted the sound quality and reported they could integrate the sound from the implant with the hearing from the better hearing ear. This acceptance occurred within the first 4 weeks of switch on. All nine patients reported that the implant improved their hearing in the most challenging situations. This subjective improvement of hearing was clearly demonstrated in the SSQ scores at 3 months compared to the pre-surgical scores. According to the results of Wilcoxon signed rank-test there was a significant improvement from pre-operative scores to the 3 months follow-up for all three subscales of SSQ: speech hearing (p=0.008); spatial hearing (p=0.001), and quality of hearing (p=0.009). Figure 3.1 displays the mean, median and minimum to maximum values of each subscale.

The lateralisation test setup in this study was quite limited as only two speakers positioned at +90 and -90 degrees were used. However, based on subjects’ reports and on the scores of the SSQ (spatial section), there is improvement in the ability to localise the sound source. Six patients could confidently identify the side of the stimulus presentation close to 100% of the time for 10 random presentations. Ability to localise sounds is currently under investigation.
Figure 3.1. Results of SSQ subscales (n=9)

“Box and Whisker” plots display the data set of each group from minimum to maximum values. In each box the mean (black quadrants), the median (horizontal lines) and the 25th and 75th percentiles are shown.

Two patients (S6 and S9) had no tinnitus either before or after surgery. All seven subjects, who had tinnitus before the operation, reported reduction in tinnitus distress when the processor was activated. This reduction varied from 77% to 100% (Figure 3.2). To date, there is no report of increase in tinnitus perception after surgery.

Figure 3.2. Change in TRQ scores from pre- to post-CI (n=9)
This group of patients was particularly very committed to the rehabilitation program and all of them dedicated between 1-3 hours a day to work on the rehabilitation material for the first 3 months. It was interesting to note that six out of nine patients were able to discriminate 100% of speech material presented to the implanted ear through direct audio input, 3 months after surgery.

3.5. Discussion

The results of this case series report are in line with previous findings and indicate that cochlear implantation is an option for hearing and tinnitus rehabilitation for a well selected group of patients with unilateral profound deafness with or without tinnitus.

An interesting finding is that in our case series even patients with long-standing deafness (S3, S5, S6 and S9) were able to rapidly integrate the electric stimulation into their acoustic normal hearing, and to improve their speech perception scores in complex listening conditions as early as at 3 months post-implantation.

The mechanisms underlying these outcomes are not understood, and several factors may be involved. Even though this case series has limited number of participants and the significance of our results was not calculated, considering the results obtained so far, it could be speculated that the frequency and intensity of rehabilitation has an impact on speed of progress and the learning curve. This has also been noted by Buechner et al. (2010).

This then leads to the crucial patient factors such as motivation and discipline as demonstrated by this particular group of subjects. An intensive discussion
on expectations took place before each surgery, which may have played a key role. It is also important to consider surgical techniques and technological factors involving electrodes design and coding strategies.

Other possibilities that need to be investigated are that some minimal amount of residual hearing in the ‘deaf’ ear may be enough to maintain a baseline auditory stimulation, which the electrical stimulation of the implant can utilise. Boisvert et al. (2011) discussed that in case of bilateral severe to profound HL, residual hearing in one ear is sufficient to maintain the metabolic activity in the auditory cortex bilaterally. Is that applicable for UD?

It is known from studies with bilateral deafened CI users that prolonged usage of the implant leads to better performance (Geers et al., 2008). This needs to be investigated in the unilateral profound HL cases.

This result opens to discussion the idea that central processes of auditory information play a more important role than the peripheral system deprivation itself. It is likely that the cortical representation of phonological information may also be relevant. These previous points should be the object of further investigation. A further argument is that consistent and continuous electric stimulation of the deprived ear could incite reorganisation of the auditory cortex leading to better acceptance and usage of the implant. How long it takes to establish remains an important question.

Like Buechner et al. (2010) and Arndt et al. (2010), we did not find deterioration in our subjects’ speech-in-noise performance in any of the spatial configurations tested. This differs from Vermiere and Van de Heyning (2009) who reported that their patients with normal hearing in the opposite
ear performed worse in the aided condition when the signal was presented from the front and the noise was presented from the implant side.

Another finding differing from previous reports, is that our patients showed an improvement in speech perception in noise when speech and noise were presented from the front indicating that the summation effect is likely to occur with electric-acoustic hearing, as early as 3 months post-implantation. More data is needed to validate this hypothesis. It is relevant to note that our group of patients had a mean score of 6dB SNR in the \( S_0/N_0 \) setup pre-operatively, which is poorer than those reported by Vermiere and Van de Heyning (2009). This may be due to the contra-lateral mild high frequency HL presented by subjects 4-7. This is even more relevant for subject 5 who presented with a high SNR value pre-operatively. Difference on the level of difficulty between the speech-in-noise tests should also be considered.

Van de Heyning et al. (2008) reported that cochlear implantation in patients with UD and tinnitus was successful for tinnitus loudness reduction for 95% of their subjects. Arndt et al. (2010) also reported that 80% of their patients achieved total or partial subjective residual inhibition of tinnitus. Our group of patients achieved total or partial suppression of tinnitus and no patient reported an increase in tinnitus perception. The patients who report higher levels of tinnitus suppression use the implant for longer periods on a daily basis.

Amoodi et al. (2011) discussed the benefits of cochlear implantation in patients with speech discrimination scores exceeding the current selection criteria. It was found that there is a discrepancy between the objective and subjective impact of cochlear implantation for those patients and
recommends that those patients should be considered for CI on an individual basis. This discrepancy is also evident for UD, which may be due to lack of an adequate objective assessment. This finding was also reported by Stelzig et al. (2011). The outcomes of our group of UD patients suggests that subjective assessment of the hearing impairment may be a crucial tool for selection criteria for UD cochlear implantation and should be carefully studied.

Arndt et al. (2010) compared the outcomes of traditional CROS hearing aids, BAHA, and cochlear implantation in the rehabilitation of UD, and reported that the improvement in hearing performance obtained with CIs was superior and should therefore be considered an alternative. Their results revealed that CI provided a statistically significant improvement in speech perception when speech was presented to the deaf ear and the noise was presented to the normal hearing ear. Our patients also achieved higher improvement in the speech perception test when noise was presented to the normal hearing ear indicating that CI can add an important positive effect in these listening situations.

Buechner et al. (2010) also investigated the use of CI to treat UD associated with tinnitus in five subjects, and found that CI may be beneficial for some patients and recommended case-to-case assessment. Our findings are in accordance with their results and support that expectations and motivation should be carefully assessed in the pre-CI work up. This group was highly compliant to the rehabilitation program and had better acceptance of the device from switch on. They were able to integrate the sound more rapidly, regardless of deafness duration. This subjective acceptance facilitates auditory training, which in turn increases their self-motivation. More data, which is necessary for a valid statistical analysis, is being collected as a
follow-up to this report. A larger number of subjects are needed to allow general conclusions.

3.6. Conclusion
This case series shows that there a clear tendency that CI should be considered as a rehabilitation option for people with UD with or without tinnitus. The patients in this group achieved a substantial benefit in speech understanding in noise, tinnitus control and a significant improvement in SSQ-scores. Although general conclusions cannot be drawn due to the lack of statistical significance calculation, this report brings to the attention that deafness duration, age of deafness onset, the presence of residual hearing, patient motivation, self-perceived handicap and the commitment to the rehabilitation program need to be further investigated in order to understand their impact on performance after implantation.
4. Impact of cochlear implantation on speech understanding, subjective hearing performance, and tinnitus perception in patients with unilateral severe to profound HL

The work described in this chapter was published in:

4.1. Abstract
Objectives: This study aimed to determine the impact of cochlear implantation on speech understanding in noise, subjective perception of hearing and tinnitus perception of adult patients with unilateral severe to profound HL, and to investigate the factors that might influence the outcomes. In addition this paper describes the auditory training protocol used for unilaterally deaf patients.

Design: This is a prospective study of subjects undergoing cochlear implantation for UD with or without associated tinnitus.

Methods: Speech perception in noise was tested using the BKB-SIN test presented at 65dB SPL. The SSQ scale and the APHAB were used to evaluate the subjective perception of hearing with a CI and quality of life. Tinnitus disturbance was measured using the TRQ. Data were collected prior to cochlear implantation and 3, 6, 12 and 24 months post-implantation.

Results: 28 post-lingual unilaterally deaf adults with or without tinnitus were implanted. There was a significant improvement in speech perception in noise over time in all spatial configurations. There was an overall significant improvement on the subjective perception of hearing and quality of life.
Tinnitus disturbance reduced significantly over time. Gender, age at implantation and duration of deafness did not influence the outcomes significantly.

Conclusion: Cochlear implantation provided a significant improvement in speech understanding in challenging situations, subjective perception of hearing performance, and quality of life. Cochlear implantation also resulted in reduced tinnitus disturbance. Age at implantation and duration of deafness did not appear to influence the outcomes.

4.2. Introduction

The use of cochlear implantation in subjects with UD was first reported in the literature in 2008 by Van de Heyning et al. (2008). Although that study was designed to investigate the benefits of cochlear implantation for tinnitus inhibition, it was found that speech perception in noise scores were also enhanced (Vermeire & Van de Heyning 2009). Since then, several other studies have investigated the benefits of cochlear implantation in subjects with UD. Buechner et al. (2010) reported that CI decreased tinnitus annoyance and improved speech understanding in some of the subjects. Arndt et al. (2011) conducted a study to compare the outcomes of the available treatment options for UD and found that CI users demonstrated significant improvement in localisation and speech understanding. Other authors have reported similar positive hearing benefits from CI use in small cohorts of patients with UD (Stelzig et al. 2011; Hansen et al. 2013; Firszt et al. 2012).

The main disabilities that affect adults with UD are lack of spatial hearing, impaired speech intelligibility in the presence of background noise and
reduced speech understanding when speech is presented to the impaired ear (McLeod et al. 2008). In a study by Priwin et al. (2007) 14% of the subjects with UHL had severe difficulty in a group conversation, 19% found that the ability to keep a conversation in presence of traffic noise was severe affected and even conversation in quiet was difficult for 26% of the participants. Vicci de Araujo et al. (2010) showed that 73% of patients with UHL reported some level of handicap, 52% reported difficulty at the cinema and 40% reported difficulty on the phone.

Subjects with UD are deprived of binaural hearing which is the ability of the auditory system to process and integrate inputs from both ears. Binaural hearing is essential for spatial hearing and it improves speech perception in noise (Arsenault et al. 1999; Bronkhorst et al. 1988; Van Deun et al. 2009; Grothe et al. 2010). The benefits of binaural hearing through bimodal amplification and bilateral CIs in adults have been well documented (Ching et al. 2004; Nopp et al. 2004; Grantham et al. 2007; Litovsky et al. 2009). Therefore, it might be expected that patients with UD would attain additional benefit from adding a CI to their unilaterally deafened ear, to be used together with their contra-lateral normal-hearing ear. However, to the best of our knowledge, there is no literature reporting the selection criteria and the key factors that might influence the outcomes. This study was designed to investigate whether duration of deafness or age at implantation has any impact on the outcomes. We hypothesised that in subjects with post-lingual UD, duration of deafness and age at implantation would not play a major role on the outcomes of cochlear implantation. Duration of deafness has been used on studies with bilateral severe-to-profound HL as a predictive factor for the outcomes of cochlear implantation in post-lingually deaf patients (Dunn et
al., 2008; UK Cochlear Implant Study Group, 2004). However, recent investigation has shown that this is particularly true if analysing the duration of deafness in the better hearing ear (Boisvert et al., 2011). It appears that if the contra-lateral ear has good speech understanding, good outcomes of implantation are obtained from the ear with long duration of deafness (Boisvert et al., 2011). Thus, subjects with long-term UD should not be denied a CI based solely on this criterion.

Age at implantation is well accepted as another key factor that will determine the outcomes of CI in congenital bilateral deafness (McConkey et al., 2004; Dettman et al., 2007). In the contrary, it seems that this is not a determinant factor for CI in adults with post-lingual deafness (Herzog et al., 2003; Olze et al., 2012).

The preliminary results of our first nine patients, with a follow-up to 3 months after cochlear implantation, have been previously published. This study adds to the existing literature by incorporating a larger number of subjects and investigating: (1) the impact of CI use in speech intelligibility in noise; (2) the long-term impact of CI use on the subjective perception of hearing; (3) if the use of the CI decreases tinnitus perception in the long-term; and (4) if the duration of deafness and age at implantation affect the outcomes.

4.3. Methods

The population, audiological and otological evaluation of the patients, statistical analysis are described in Chapter 2.
4.4. Results

4.4.1. Speech perception in noise

The speech perception results in noise of the adaptive BKB-SIN test in the three spatial setups are shown in Figure 4.1. Mean scores for the $S_0/N_0$ spatial test set up ranged from 4.47 (±SD=1.796) pre-operatively to 1.90 (±SD=1.917) 12 months post-operatively. Mean scores for the $S_0/N_{\text{HE}}$ spatial test set up ranged from 0.14 (±SD=3.681) pre-operatively to -1.57 (±SD=3.217) 12 months post-operatively. Mean scores for the $S_{\text{CI}}/N_{\text{HE}}$ spatial test set up ranged from 5.11 (±SD=3.722) pre-operatively to -0.80 (±SD=3.262) 12 months post-operatively.

Figure 4.1. BKB-SIN test results over time (n=28)

Median values are displayed as a horizontal line, mean values as black squares. Asterisks denote outliers.

Time (pre-operatively, 3, 6 and 12 months post-operatively) had a significant main effect on the $S_0/N_0$ spatial test outcome ($F(3,42)=21.878; p<0.001$), on the $S_0/N_{\text{HE}}$ spatial test outcome ($F(3,42)=7.226; p=0.003$), and on the $S_{\text{CI}}/N_{\text{HE}}$ spatial test outcome ($F(3,42)=74.326; p<0.001$). There was no significant interaction between age at implantation, or duration of deafness across time,
nor any of their possible interactions across time. There was no significant
interaction between age at implantation, or duration of deafness, at the
individual time intervals tested; nor any of their possible interactions at the
individual time intervals tested. Tables 4.1, 4.2, and 4.3 show the p-values for
the three spatial configuration.

Table 4.1.  BKB-SIN S₀/N₀ results of a multivariate GLM

<table>
<thead>
<tr>
<th>Dependent variable</th>
<th>df</th>
<th>F-value</th>
<th>p-value 2-sided</th>
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<tbody>
<tr>
<td>Age</td>
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</tr>
<tr>
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Table 4.2.  BKB-SIN S₀/N_H results of a multivariate GLM

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<td>Duration of deafness</td>
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Table 4.3.  BKB-SIN S_C/N_H results of a multivariate GLM

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<th>p-value 2-sided</th>
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<td>.516</td>
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4.4.2. Subjective testing

The results of the three subscales of the SSQ are shown in Figure 4.2. Mean scores for the subscale speech ranged from 4.69 (±SD=1.827) pre-operatively to 7.65 (±SD=1.105) 24 months post-operatively. Mean scores for the subscale Spatial ranged from 2.61 (±SD=1.604) pre-operatively to 7.37 (±SD=1.204) 24 months post-operatively. Mean scores for the subscale quality of hearing ranged from 6.16 (±SD=1.797) pre-operatively to 8.15 (±SD=0.947) 24 months post-operatively.

![Speech: Spatial Quality Questionnaire [SSQ]](image)

**Figure 4.2.** SSQ subscales results over time (n=28)

Median values are displayed as a horizontal line, mean values as black squares. Asterisks denote outliers.

Time had a significant main effect on the speech subscale (F(1.4; 12.7)=6.051; p=0.021), on the spatial subscale (F(1.6; 14.7)=24.160; p<0.001), and on the quality of hearing subscale (F(1.5; 13.3)=6.326; p=0.017). There was no significant interaction between age at implantation, or duration of deafness across time, nor any of their possible interactions across time. There was no significant interaction between age at implantation, or duration of deafness, at the individual time intervals tested; nor any of their possible interactions at the individual time intervals tested. Tables 4.4, 4.5 and 4.6 show the p-values for the three SSQ subscales.
Table 4.4. SSQ subscale speech – results of a multivariate GLM

<table>
<thead>
<tr>
<th>Dependent variable</th>
<th>df</th>
<th>F-value</th>
<th>p-value (2-sided)</th>
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</thead>
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<tr>
<td>Age</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>pre-op</td>
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<td>2.828</td>
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<td>3 months post-op</td>
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<td>.991</td>
<td>.358</td>
</tr>
<tr>
<td>6 months post-op</td>
<td>1</td>
<td>2.497</td>
<td>.165</td>
</tr>
<tr>
<td>12 months post-op</td>
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</tr>
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<td>.393</td>
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<td>Duration of Deafness</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>pre-op</td>
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<td>.309</td>
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<td>.953</td>
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<td>12 months post-op</td>
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<td>2.294</td>
<td>.181</td>
</tr>
<tr>
<td>24 months post-op</td>
<td>1</td>
<td>.446</td>
<td>.529</td>
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</table>

Table 4.5. SSQ subscale spatial – results of a multivariate GLM

<table>
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<th>Dependent variable</th>
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<td>6 months post-op</td>
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<td>.090</td>
<td>.774</td>
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<td>24 months post-op</td>
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<td>.001</td>
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<td>Duration of Deafness</td>
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<td>24 months post-op</td>
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<td>.552</td>
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Table 4.6. SSQ subscale quality of hearing – results of a multivariate GLM

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<th>F-value</th>
<th>p-value (2-sided)</th>
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</thead>
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<tr>
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<tr>
<td>pre-op</td>
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<td>.445</td>
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<td>6 months post-op</td>
<td>1</td>
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<td>12 months post-op</td>
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<td>24 months post-op</td>
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</tr>
<tr>
<td>pre-op</td>
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<td>3 months post-op</td>
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<td>12 months post-op</td>
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<tr>
<td>24 months post-op</td>
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<td>1.009</td>
<td>.354</td>
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</tbody>
</table>
The results of the three APHAB subscales are shown in Figure 4.3. Mean scores on the EC subscale were 33.6 (±SD=14.912) at pre-operative testing and 14.4 (±SD=12.602) at 12 months post-operatively. Mean scores on the subscale RV were 46.6 (±SD=17.570) at pre-operative testing and 23.2 (±SD=12.984) 12 months post-operatively. Mean scores on the BN subscale were 63.6 (±SD=14.212) at pre-operative testing and 32.1 (±SD=12.576) at 12 months post-operatively. Mean scores on the Global scale were 47.2 (±SD=13.797) at pre-operative testing and 22.9 (±SD=11.802) 12 months post-operatively.

![APHAB Scales](image)

**Figure 4.3.** APHAB subscales and Global scale results between pre-operative testing and 12 months post-operative testing (%) (n=28)

Median values are displayed as a horizontal line, mean values as black squares. Asterisks denote outliers.

The subjective perception of EC improved significantly between pre-operative testing and 12 months post-operatively (t=4.97; p<0.001). The subjective perception of RV improved significantly between pre-operative testing and 12 months post-operatively (t=5.38; p<0.001). The subjective perception of BN improved significantly between pre-operative testing and 12 months post-operatively (t=6.45; p<0.001). There was no significant
interaction between age at implantation, or duration of deafness, pre-operatively or 12 months post-operatively; nor any of their possible interactions, pre-operatively or 12 months post-operatively (p=0.090 to p=0.968; see Supplemental Digital Content 7, 8 and 9, which show the p-values for the three APHAB subscales).

The Global scale improved significantly between pre-operative testing and 12 months post-operatively (t=6.15; p<0.001). There was no significant interaction between age at implantation, or duration of deafness, pre-operatively or 12 months post-operatively; nor any of their possible interactions, pre-operatively or 12 months post-operatively (p=0.122 to p=0.543) Table 4.7 shows the p-values for the APHAB Global scale.

<table>
<thead>
<tr>
<th>Dependent variable</th>
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<th>F-value</th>
<th>p-value (2-sided)</th>
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<td>Age</td>
<td></td>
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</tr>
<tr>
<td>pre-op</td>
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<tr>
<td>12 months post-op</td>
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<td>2.911</td>
<td>.122</td>
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<tr>
<td>Duration of deafness</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>pre-op</td>
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<td>12 months post-op</td>
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<td>.225</td>
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### 4.4.2 Tinnitus disturbance

The results of the TRQ are shown in Figure 4.4. Mean scores for the TRQ were 48.8 (±SD=27.150) at pre-operative testing and 1.75 (±SD=4.200) 24 months post-operatively. Time had a significant main effect on the TRQ F(1.3; 7.8)=10.098; p=0.011). There was no significant interaction between age at implantation, or duration of deafness across time, nor any of their possible interactions across time.
Figure 4.4. TRQ results over time (n=13)
Median values are displayed as a horizontal line, mean values as black squares.

There was no significant interaction between age at implantation, or duration of deafness, at the individual time intervals tested; nor any of their possible interactions at the individual time intervals tested (p=0.170 to p=1.000).

Table 4.8 shows the p-values for the TRQ scores.

Table 4.8. TRQ results of a multivariate GLM

<table>
<thead>
<tr>
<th>Dependent variable</th>
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<th>F-value</th>
<th>p-value (2-sided)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
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</tr>
<tr>
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<td>3 months post-op</td>
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<td>.000</td>
<td>1.000</td>
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<td>6 months post-op</td>
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<td>.006</td>
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<td>12 months post-op</td>
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<td>24 months post-op</td>
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<td>pre-op</td>
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<td>.645</td>
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<tr>
<td>Duration of deafness</td>
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</tr>
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<td>3 months post-op</td>
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<td>.565</td>
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<td>.541</td>
</tr>
<tr>
<td>24 months post-op</td>
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<td>.018</td>
<td>.906</td>
</tr>
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</table>
4.5. **Discussion**

Recent literature has demonstrated cochlear implantation to be a suitable hearing rehabilitation option for adults with unilateral profound deafness. A CI is the only option that provides ear specific information and thus potentially the benefits of binaural hearing. There are known benefits to listening with two ears as reviewed in the “International consensus on bilateral CIs” which suggested that speech recognition in noise is better under binaural listening conditions than under binaural deprivation (Offeciers *et al.*, 2005). Litovsky *et al.* (2006) have shown that using bilateral implants a 12-33% improvement in speech perception can be achieved by deaf adults. Bilaterally implanted deaf children perform better than children with a unilateral CI in speech in noise testing (Kuhn-Inacker *et al.*, 2004). However, the benefits of CI in UD and the improvement on speech performance in noise have been addressed by very few groups over the past few years (Buechner *et al.*, 2010; Arndt *et al.*, 2011; Stelzig *et al.*, 2011; Hansen *et al.*, 2013; Firszt *et al.*, 2012). To date, the selection criteria for cochlear implantation in UD are not clearly established and factors that might determine the outcomes are unknown. This study addressed this issue by analysing the potential influence that age at implantation and duration of deafness might have on the CI outcomes.

This study investigated the hearing benefits of CI in terms of speech perception, self-assessment of hearing performance, and tinnitus disturbance, in a cohort larger than 25 subjects, with long-term follow-up. It is the first to investigate whether duration of deafness, age at implantation and gender has an effect on the outcomes. Our results showed a significant improvement in speech perception in noise scores when speech and noise are presented from
the front \((S_0/N_0)\), and when speech is presented from the front \((S_0/N_{\text{HE}})\), or from the CI side \((S_{\text{CI}}/N_{\text{HE}})\) with the noise presented to the normal hearing ear. These results indicate that patients with UD may be able to make use of some of the benefits associated with bilateral hearing.

Both SSQ and APHAB showed a significant subjective improvement of hearing with CI use. The analysis of the SSQ scores confirmed that the improvement over time was significant for all three subscales – speech, spatial hearing and qualities of hearing. Similarly, Arndt et al. (2011) reported a significant improvement in the speech and spatial hearing subscales while Vermeire and Van de Heyning (2009) found an improvement in all three subscales, indicating that the subjective perception of hearing improves with CI use in UD subjects. The SSQ questionnaire was repeated at 24 months post-CI and the scores showed that the subjective benefit remained stable in the long-term. The results of the APHAB were in line with the SSQ scores, with an improvement in the subscales – ease of hearing, reverberation, and background noise. The scores of the aversiveness subscale did not change significantly before and after surgery. This demonstrates that CI in UD enhances patients’ self perception of hearing performance and does not create any tolerance issues.

The disturbance caused by tinnitus also decreased for the subjects. A further follow-up at 24 months showed that the improvement was stable in the long-term. A significant positive effect on tinnitus has been reported previously (Van de Heyning et al., 2008; Buechner et al., 2010; Ramos et al., 2012). In accordance with the data herein, Punte et al. (2011) reported a significant decrease in tinnitus distress in 26 subjects who received a CI for
UD associated with tinnitus. Thus, it appears that CI can be used as an effective means of reducing tinnitus disturbance.

Our results showed that, for post-lingual unilateral deaf adult, age at implantation or duration of deafness did not have a significant effect on the speech perception in noise scores, subjective hearing performance as measured by the SSQ and APHAB and on the improvement of tinnitus confirming our initial hypothesis that these factors would not play a key role in the rehabilitation of post-lingual UD using CI. A more robust sample size would be beneficial to corroborate these findings.

There was a concern that subjects with normal contra-lateral hearing would have difficulties integrating the acoustic and electric signals. In contrast to standard CI recipients who had to rely on some kind of amplification, subjects with UD might be expected to give up using their CI if their progress was too slow or if the obvious benefits did not overcome the challenges associated with CI use. Studies have demonstrated the benefits of active auditory training for patients using auditory rehabilitation (Fu et al., 2007; 2004). In an earlier UD case study by Nawaz et al. (2014) one subject received training to stimulate both ears simultaneously and promote binaural integration with success. In our study, intensive auditory training was provided which aimed to facilitate acclimatisation to the CI. Further investigation is necessary to establish the exact contribution of auditory training. Cumulatively these results along with device acceptance suggest that subjects with UD are probably able to combine the acoustic and electrical signals.
4.6. Conclusion
The results of the present study indicate that in subjects with UD speech perception in noise is improved with CI use. CI use improves the subjective perception of hearing in UD subjects and decreases the disturbance of tinnitus, which may in turn contribute to an overall positive subjective impression of benefits in this group of subjects. It appears that age at implantation and duration of deafness do not affect the outcome.
5. **Localisation acuity is improved in CI users with a UD**

The work described in this chapter was published in:

5.1. **Abstract**

Objectives: One of the major complaints of people with a single-sided deafness is the inability to localise sound sources. Evidence suggests that subjects with a HL can benefit from the use of a CI in sound localisation. This study aimed to determine the effect of CI use on localisation ability in unilaterally deafened subjects.

Design: Sixteen adult subjects with post-lingual UD, fitted with a CI on the deaf side, were included in this study. The auditory speech sounds evaluation (A§E®) localisation test was used to determine localisation with a CI on (binaural) and a CI off (monaural). The RMS error was used as a measure of the subjects’ localisation performance. Stratified analyses were performed to test the influence of gender, age of implantation (<55 years and >55 years), and the duration of deafness (<10 years and >10 years) on localisation ability.

Results: Subjects with a CI on localised significantly better than without a CI. Gender, age, and the duration of deafness had no effect on the localisation ability of the subjects.

Conclusions: Cochlear implantation is effective in improving localisation abilities in subjects with UD. The RMS error dropped significantly with binaural hearing compared to monaural hearing.
5.2. Introduction

Binaural hearing provides the benefit of sound localisation in the normal auditory system and allows better understanding of speech in noisy situations when the source of speech and the noise are presented from different locations in the horizontal plane (reviewed by Grothe et al. 2010 and Litovsky et al. 2012).

Sound localisation in the horizontal plane relies on two binaural cues – ILD and ITD. The aforementioned cues help to localise high frequency and low frequency sounds respectively (Grothe et al. 2010). Several studies have demonstrated that CI users rely mostly on Interaural Level Differences (ILDs) to localise sounds (e.g. Laback et al. 2004, Senn et al. 2005, Grantham et al. 2007). It is conceivable that this is also true for unilaterally deafened CI users. In view of that, it was thought that a localisation test that only uses ILDs as a cue would be suitable to verify whether unilaterally deafened CI users can integrate the two, acoustic and electrical, signals in order to identify the sound location. This study then was performed using the auditory speech sounds evaluation (A$E®) localisation test (PJ Govaerts, Antwerp, Belgium) that creates an ILD in an artificial way.

Numerous reports over the last decade have argued that one of the benefits of bilateral cochlear implantation over unilateral usage is sound localisation in both paediatric and adult populations (e.g. Van Hoesel & Tyler 2003; Nopp et al. 2004; Grantham et al. 2007; Litovsky et al. 2009).

Cochlear implantation in the treatment of unilaterally deafened subjects is a relatively new treatment modality (Arndt et al. 2010). Traditionally patients were offered either no treatment, a conventional CROS, or BAHA (Arndt et
al. 2010). Our clinical experience with unilateral deafened patients is that the possibility of acquiring localisation skills can be the driving force that triggers some of them to explore the possibility of cochlear implantation. For this group of patients, BAHA or CROS may not be satisfactory options, as both devices are limited to transferring the signal of the deaf ear towards the good hearing ear, therefore not providing binaural advantage for localisation ability.

Arndt et al. (2010) compared the outcomes of CI, CROS and BAHA in a group of 11 subjects. Localisation tests were performed using seven loudspeakers positioned at intervals of 30 degrees between -90 and 90 degrees. Localisation results showed a significant improvement with CI when compared with the unaided condition, and also when compared to performance with the CROS or BAHA devices.

Recently published reports include data on a very small number of CI subjects with UD of short duration – Firszt et al. (2012) reported that localisation was significantly better with bilateral hearing in all three subjects included in her study. Likewise, Hassepass et al. (2013) showed improved localisation skills in two out of three children, 12 months after cochlear implantation. To our knowledge there is no literature that solely addresses the localisation ability in a large number of participants with UD and CI. A novelty of this study is the inclusion of patients with long-deafness duration.

that cochlear implantation could re-establish the benefits of binaural hearing in post-lingual UD, significantly improving the patients’ ability to localise sound sources. The present study addresses this hypothesis by examining the localisation ability in 16 adult subjects with post-lingual UD who received a CI within the last 18 months.

Duration of deafness has been used in studies on bilateral severe-to-profound HL as a predictive factor for the outcomes of cochlear implantation in post-lingually deaf patients (Dunn et al. 2008, UK Cochlear Implant Study Group 2004). We have previously looked at a small cohort of five patients with more than 25 years of deafness duration and did not find that deafness duration was a determinant factor in the CI outcomes in post-lingual UD (Távora-Vieira et al. 2013). There is some evidence that if UD occurs after maturation of bilateral pathways the lateralisation of contra-lateral activation of the auditory areas is not altered (Langers et al. 2005) and thus it was thought that localisation could be achievable by this group of subjects if cochlear implantation was provided to the deaf ear.

Younger age at implantation has been well accepted as essential for improved outcomes of cochlear implantation of children with congenital bilateral deafness (e.g. Van Deun et al. 2009, Dettman et al. 2007, McConkey et al. 2004). For adult with post-lingual bilateral deafness, there is no evidence that age of implantation is a key factor for CI outcomes (Herzog et al. 2003, Olze et al. 2012). However, it is possible that older adults with UD would find more difficult to adapt to a new combined acoustic and electrical inputs.
This study also aimed to investigate whether deafness duration and age at implantation played a role in the localisation performance of unilaterally deafened CI users.

5.3. Methods
The methods used in this chapter were described in Chapter 2.

5.4. Results
Figure 5.1 shows the localisation test results of the subjects (n=16) tested with their CI on and with their CI off. The difference in localisation ability, as determined using the A§E, was significantly better with the CI on (mean RMS error=22.8; SD=11.58) than the CI off (mean RMS error=48.9; SD=11.86) (Paired samples t-test: t=6.979; df=15; p<0.001).

Individual RMS data is presented in Figure 5.2. The data shows that subjects S2 and S5 did not show any improvement in localisation ability.

![Localization Chart]

**Figure 5.1.** Mean RMS error with CI on and CI off (n=16)

Lower RMS represents better localisation skills. Median values are displayed as horizontal line, mean values as black squares. Length of the whiskers corresponds to the range of the data. The black asterisk represents an outlier (1.5 to 3 x box height above the 75th percentile).
In female participants the localisation ability with the CI on was significantly better than with the CI off ($p=0.003$). In male participants localisation ability with the CI on was significantly better than with the CI off ($p=0.001$). There was no significant difference with the CI on between female subjects and male subjects (Independent-samples $t$-test: $t=.377; df=14; p=0.712; CI: -10.547-15.047$). Also no significant difference between genders was found for CI off ($t=-.389; df=14; p=0.703; CI: -15.473-10.723$).

Localisation ability with the CI on was significantly better than with the CI off in subjects <55 years of age ($p<0.001; n=10$), and in subjects >55 years of age ($p=0.012; n=6$). There was no significant difference with the CI on ($t=.357; df=14; p=0.727; CI: -11.023-15.423$) between subjects <55 years of age and subjects >55 years of age. Likewise, there was no significant difference with the CI off ($t=-.058; df=14; p=0.955; CI: -13.965-13.232$) between subjects <55 years of age and subjects >55 years of age.

Localisation ability with the CI on was significantly better than with the CI off in subjects implanted after a duration of deafness of <10 years ($p=0.001; n=9$), and in subjects with a duration of deafness >10 years ($p=0.010; n=7$).
There was no significant difference with the CI on (t=.375; df=14; p=0.713; CI: -15.153-10.645) or with the CI off (t=1.187; df=14; p=0.255; CI: -5.651-19.651) between subjects with duration of deafness <10 years and subjects with a duration of deafness >10 years.

Additionally, the effect of gender, duration of deafness and age at implantation on the difference between the CI off and CI on conditions was examined. No significant difference was found between female and male subjects (Independent-samples t-test: t=.606; df=14; p=0.554; CI: -20.993-11.743), between subjects <55 years and those >55 years of age (Independent-samples t-test: t=.323; df=14; p=0.752; CI: -19.628-14.495), and between subjects with <10 years of duration of deafness and those >10 years of duration of deafness (Independent-samples t-test: t=-1.252; df=14; p=0.231; CI: -6.594-25.102).

5.5. Discussion
This is the first study to solely investigate the localisation ability in a large number of unilaterally deafened CI users, including subjects with long-term deafness duration (up to 39 years). The results confirmed our hypothesis that localisation would improve significantly after cochlear implantation. This study also investigated some factors that potentially could influence the outcomes.

This study investigated localisation skills in unilaterally deafened CI users using A§E test localisation software, which simulates an ILD, therefore generating the impression of the sound originating from the azimuth between the two loudspeakers. This software was developed to assess subjects using
bilateral CIs and is used for the first time in this study to determine localisation ability in UD.

Paired sample t-tests showed that in the binaural condition (CI on) subjects performed significantly better on the A§E localisation test than in the monaural condition (CI off). The mean RMS error dropped from 49 degrees (CI off) to 22.8 degrees (CI on). If S2 and S5, the poorer performers on the localisation test, were excluded from the analysis the mean RMS error would be 20.0 degrees. Based on the normative data, the RMS error for 30 normally hearing adults using A§E ranged from 5-11 (median=8) degrees. The RMS error for the group of subjects in this study ranged from 11 to 47 (median=17.5) degrees in the binaural condition. Although the results shown by the unilaterally deafened group is worse than the normal hearing subjects tested in the same condition, the outcomes are very encouraging. While normal hearing individuals make use of interaural matched stimuli to localise sound sources, unilaterally CI users combine two distinct inputs (acoustic and electrical) and consequently their brain needs to create a spatial map with a substantial interaural dissimilarity.

There are very few studies that investigated the localisation ability of unilaterally deafened adults CI recipients (all with duration of deafness <10 years). Arndt et al. (2010) investigated the localisation ability of 11 adult with UD and CI using a setup of seven loudspeakers positioned at intervals of 30 degrees between -90 and 90 degrees. Speech was used as the stimuli. The localisation error was significantly reduced with CI on (median=15.0 degrees) compared with the CI off condition (median=33.9 degrees). This study showed that the RMS error in the CI off and CI on conditions were higher than what was reported by Arndt et al. (2010). This is true even if we
included only the patients with less than 10 years of deafness. It is possible that, as observed with bilateral CI users, the use of speech signal as stimuli (Verschuur et al. 2005, Grantham et al. 2007) and fewer loudspeakers facilitated sound localisation in that study. Similarly to our results, Firszt et al. (2012) reported that a small group of three adults performed significantly better in the binaural condition with the RMS error being 18, 19 and 25 degrees. Although a speech signal was used as the stimuli, localisation testing was performed using 15 loudspeakers located 10 degrees apart making the setup more comparable to ours.

Several studies of children and adults with bilateral deafness have demonstrated the superiority of localisation skills with bilateral CIs over unilateral (e.g. Van Hoesel & Tyler 2003; Nopp et al. 2004; Grantham et al. 2007; Litovsky et al. 2006a,b; 2009). Litovsky et al. (2009) reported the localisation test results of 17 adult bilateral CI users. The mean RMS error was 56.6-60.4 degrees with monaural hearing and 28.4 degrees with binaural hearing. Grantham et al. 2007 reported that the average RMS localisation error of 30.8 degrees for 22 CI users compared to 6.7 degrees for normal hearing subjects. This study showed that the RMS error in the CI off and CI on conditions were lower than what was found with bilateral CI users in monaural and bilateral listening conditions. Although bilateral CI users can localise sound significantly better when using both devices, they perform worse than normal hearing individuals. This is explained on the basis that they rely on a binaural input that is likely to be dissimilar due to potentially different innervation pattern of the cochlea, and different depth of electrode insertion with consequent mismatch of anatomical place of stimulation (Kan et al. 2013). The challenge for the unilaterally deafened CI users is not lesser
as their brain works with an interaural divergence in order to create a spatial map as mentioned earlier.

It was thought that the ability to achieve good binaural integration might be time dependent, linked to (1) age at implantation, (2) duration of deafness or (3) the age at onset of deafness. Age of implantation and duration of deafness were not supported by the results of our study. The age at onset of deafness was not analysed in the study.

When looking if age at the time of implantation had an influence on the results, the difference between CI off and CI on was significant for both age groups. This finding is in line with several studies showing that the hearing outcomes achieved by the elderly CI users do not differ from the younger population (Herzog et al. 2003, Noble et al. 2009, Olze et al. 2012). Nevertheless, the sample size of both age-stratified groups was relatively small. It is possible that a larger sample would more reliably reflect the population mean and yield a different result. Additionally, gender did not influence the results.

Previous literature has reported only the outcomes of patients with less than 10 years of UD duration. As the patients in this study had between 3 months to 39 years of deafness duration, it was decided to stratify the listeners into two groups with deafness duration > or <10 years. The difference between CI off and CI on was significant for both groups and there was no significant difference between the groups in each listening condition. It is important to note, however, that from the six patients with >10 years of deafness of duration, four lost their hearing after age of 12. Normal binaural hearing during the development of bilateral auditory pathways may explain why these
patients had an improvement in spatial hearing even after a prolonged period of hearing deprivation (Keating & King 2013, Grothe et al. 2010, Litovsky et al. 2010). Electrophysiological studies have demonstrated that if UD occurs after maturation of bilateral pathways the lateralisation of contra-lateral activation of the auditory areas is not altered (Langers et al. 2005) supporting the idea that localisation may be improved even if cochlear implantation occur after a long-term UD.

In the present study, two subjects (out of the 16 that participated) did not demonstrate any improvement in localisation ability with CI use. Interestingly, both subjects had a long duration of deafness before implantation and had lost their hearing at 6 and 7 years of age, the youngest ages of deafness onset in the group. Similarly, Nopp et al. (2004) found that two patients who lost their hearing in early childhood (one at birth and one at the age of 6 years) did not show improvement in localisation skills. Johnstone et al. 2010 reported that localisation ability was only improved in children with UHL who received a hearing aid before the age of 5. In the last few years, human and animal studies have demonstrated that there is a critical period for bilateral auditory pathway development explained by neuronal reorganisation of the auditory brain following prolonged monaural hearing input in bilateral congenital profound deafness (e.g. Gordon et al. 2011, 2013, Kral et al. 2013). Keating and King (2013) also suggested that spatial hearing might be affected permanently if an extended period of asymmetrical hearing input occurs at a young age. Age of onset of deafness was not analysed in the study due to small sample size. However, it is conceivable that the two poorer performing patients in our study were unilaterally deafened prior to binaural hearing maturation. It is important to note that both subjects used their speech
processor on a full-time basis and reported a substantial benefit. The subjects also reported that they could not localise sounds precisely in an everyday listening environment, but that they were able to lateralise correctly most of the time.

Litovsky et al. (2006b) reported that sound localisation in children improves over time. It is thought that children develop experience-dependent skills because of auditory plasticity. It is possible that there are differences in the ability of children and adults with CIs to improve their localisation ability, which may depend on auditory plasticity. However, a long-term follow-up is needed to investigate if localisation ability improves over time in the adult population.

Training probably facilitates adults’ ability to adapt to new spatial information (Nawaz et al. 2014). Further research is needed to determine whether appropriate training is enough to re-establish the auditory pathways related to localisation skills in post-lingually unilaterally deafened individuals and whether the subjects that did not perform well in the present study would develop strategies to localise sound better with practice.

5.6. Conclusion
The data herein suggests that cochlear implantation is effective in improving localisation abilities in unilaterally deafened subjects. This improvement occurred within the period from 6-18 months after cochlear implantation and the RMS error dropped significantly with binaural hearing compared to monaural hearing. Deafness duration and age at implantation do not seem to influence the results.
6. Successful outcomes of cochlear implantation in long-term UD – brain plasticity?

This work was published in:

6.1. Abstract
Objectives: to investigate the implications of deafness duration in the rehabilitation of UD utilising cochlear implantation.

Methods: out of the ongoing prospective cochlear implantation in UD study, we looked at five adults who received a CI for long-term UD. Speech perception in noise and subjective evaluation of the benefits of cochlear implantation were measured at 3, 6 and 12 months post-implantation. The results were analysed and compared with published data from normal hearing people and adults using CIs bilaterally.

Results: analysis of speech perception in noise revealed significant improvement for three spatial configurations: speech and noise from the front (S₀/N₀; p=0.003); speech from the front and noise from the normal hearing ear (S₀/N_HE; p=0.001); and speech from the implanted ear and noise from the normal hearing ear (S_CI/N_HE; p<0.001). The scores obtained at 12 months post-surgery improved towards values similar to those obtained by individuals with normal hearing. Results of subjective measures showed significant improvement of hearing over time, towards the scores obtained by individuals with bilateral CIs and those with normal hearing.

Conclusions: in this study, older adults with more than 25 years of UD obtained scores in speech perception testing and in subjective evaluation that
are similar to those attained by subjects with normal hearing and/or with bilateral CIs. Therefore, patients with post-lingual UD should not be excluded as CI candidates on the basis of long deafness duration.

6.2. **Introduction**

CIs are widely accepted as the most appropriate hearing devices for individuals with severe to profound bilateral HL and poor aided speech recognition. However, in recent years, patients with more residual hearing, asymmetrical HL and those with UD (having normal hearing in one ear) have also benefited from cochlear implantation (Buechner *et al.*, 2010; Vermiere & Van de Heyning, 2009; Firszt *et al.*, 2012; Tavora-Vieira *et al.*, 2013).

While candidacy criteria continuously expand and evolve towards considering non-traditional patients for implantation, the rate of expansion is intrinsically related to the rate of available evidence that supports the higher benefit patients can derive from implantation over other hearing interventions or devices.

For patients with UD, hearing disabilities have been particularly associated with the lack of binaural benefits. This causes great difficulties understanding speech in the presence of noise, especially when the speech is perceived from the affected side (Tavora-Vieira *et al.*, 2013; Arndt *et al.*, 2010).

The enhancement of speech understanding when listening with two ears compared to one has been demonstrated in several studies of bimodal amplification (i.e. a CI in one ear and a hearing aid in the other (e.g. Ching *et al.*, 2007) and bilateral implantation (Litovsky *et al.*, 2006; Dunn *et al.*, 2008). In the case of UD, the benefits of implantation have been compared with those of a CROS hearing aid, where the signal on the side of the deaf ear
is directed towards the good hearing ear, a BAHA, which uses vibration of the skull to stimulate both cochleae similarly. In most cases, cochlear implantation is found to be an effective treatment option to gain bilateral hearing with UD (Firszt et al., 2012; Arndt et al., 2010). However, the advantages of cochlear implantation may be masked by the inadequacy of the speech testing set-up (Stelzig et al., 2011).

Based on studies with bilateral severe-to-profound HL, duration of deafness and presence of residual hearing have largely been used as predictive factors for cochlear implantation outcomes in post-lingually deaf patients (Dunn et al., 2008; UK Cochlear Implant Study Group, 2004). In particular, the duration of profound deafness in the ear to be implanted seems to be the most clinically accepted predictor of outcomes in a majority of CI centres. Accordingly, cochlear implantation in UD has predominantly been performed in cases of short-duration deafness: 18-62 months (Buechner et al., 2010); 4-110 months (Arndt et al., 2010); and 11-96 months (Stelzig et al., 2011). However, recent data suggest that good speech understanding with one ear might be sufficient to obtain good outcomes of implantation in any of the ears (Boisvert et al., 2012). This opens up a discussion about whether duration of deafness is the most significant predictor of outcomes in cases of unilateral long-term deafness. While candidacy criteria for cochlear implantation are being defined for UD, to date, no study has investigated the implications of long deafness duration.

As speech perception outcomes of cochlear implantation are most strongly related to the duration of significant deafness in the better ear (Boisvert et al., 2011), then adults with post-lingual unilateral severe to profound HL (i.e. with normal hearing in one ear) should be amongst the best performers with a
CI. This paper addresses this hypothesis by examining outcomes of cochlear implantation in five adults with over 25 years of severe to profound UHL, and comparing this with published data from normal hearing participants and adults with bilateral CIs.

6.3. Methods
The methods used in this chapter were described in Chapter 2.

6.4. Results
Five adults (three males, two females) with UHL greater than a 4-frequency PTA0.5-4kHz of 70dB HL and duration of deafness >25 years were identified out of the patient cohort of the cochlear implantation in UD study. The average duration of deafness was 35 years (range of 27-40 years) and the mean age at implantation was 55 years (range of 48-68 years). All participants had either normal hearing or a mild loss in the better hearing ear, with a mean PTA0.5-4kHz <30dB H.

6.4.1 BKB-SIN sentences
Group results are presented in Figure 6.1. Repeated measure ANOVAs show that a significant improvement was reached for all three spatial configurations: S0/N0 (F (3,12)=8.382; p=0.003), S0/NHE (F (3,12)=10.924; p=0.001), SCL/NHE (F (3,12)=25.221; p<0.001).
Figure 6.1. Group results for the three presentation configurations of the BKB-SIN sentence recognition test over time (n=5)

Mean values are depicted as black squares, median values as horizontal lines and asterisks describe extreme values, i.e. outliers.

Figure 6.2 illustrates the individual scores obtained on the BKB-SIN test before and 1 year after cochlear implantation for the five participants with long duration UD. This is compared with results obtained by traditional CI users with a bilateral HL and adults with normal hearing [adapted data from 7, 17, 18]. This suggests that before implantation, individuals with UD obtained BKB-SIN scores between those obtained by CI users with bilateral HL and individuals with normal hearing. One year after implantation, the scores obtained by individuals with UD improved towards values similar to those obtained by individuals with normal hearing.
Figure 6.2. Individual scores obtained on the BKB-SIN test before and 1 year after CI

The arrows illustrate the changes in SNR required to obtain 50% on a sentence recognition task before and 12 months after implantation, in three presentation conditions, for the five participants with long duration of single-sided deafness. Lower SNR values = higher score. Patterned areas represent, as a comparison, the mean and standard deviation obtained on the BKB-SIN test in other studies.

6.4.2 SSQ subscales

According to the results of repeated measure ANOVAs, a significant improvement over time for speech ($F(3,12)=19.631; p<0.001$), for spatial ($F(3,12)=30.772; p<0.001$), and for qualities of hearing ($F(3,12)=10.696; p=0.001$) was reached (Figure 6.3).
Figure 6.3. Results of the SSQ scales measured pre-operatively, at 3 months, 6 months and 12 months post-operatively (n=5)
Mean values are depicted as black squares, median values as horizontal lines, and asterisks describe extreme values, i.e. outliers.

Figure 6.4 presents the individual scores obtained by the five participants with long duration UD, on the SSQ questionnaire before and 1 year following implantation, in comparison to scores obtained by individuals with bilateral CIs and individuals with normal hearing. This comparison suggests that before implantation, individuals with long-term UD had poorer hearing abilities than individuals with bilateral CIs. Following implantation, their abilities on the sections of the SSQ improved towards the scores obtained by individuals with bilateral CIs and those with normal hearing.

6.4.3 CUNY sentences in quiet presented through direct audio input
According to the results of a repeated measure ANOVA, a significant improvement for the CUNY sentences was also reached over time ($F(3,12)=24.446; p<0.001$) when using the implanted ear alone (Figure 6.5).
Figure 6.4. Individual scores obtained by the five participants with long duration of deafness, on the SSQ questionnaire before and 1 year following implantation

The arrows illustrate the scores in the three SSQ sections before and 12 months after implantation, for the 5 participants with a long duration of single-sided deafness. Higher scores indicate greater ability. Patterned areas represent the mean and standard deviation obtained on the SSQ questionnaire from previous publications (adapted from Ponton et al. (2001) and O’Neil et al. (2010).

Figure 6.5. Results of City University of New York (CUNY) sentence recognition scores presented via direct audio input over time (%) (n=5)

“Box and Whisker” plots display the data set at each testing time. Mean values are depicted as black squares, median values as horizontal lines.
6.5. Discussion

The use of a CI in unilateral profound HL or deafness is a new hearing rehabilitation option. Although its acceptance among the otolaryngologists and audiologists is growing, there is limited information regarding the factors that may affect outcomes. Currently cochlear implantation for long-term UD is highly controversial. However, neurophysiological and clinical research data suggest that monaural auditory stimulation may be sufficient to maintain the integrity of the auditory pathways bilaterally.

Neurophysiological changes have been demonstrated at multiple levels of the auditory pathways following unilateral and bilateral deafness in animals [see Demeester et al., 2012 for a review]. While the neurophysiological changes occurring with unilateral ablation in animal models are more extensive when the asymmetry occurs early in life (Noble et al., 2009; Shepherd et al., 2001), acquired long-term UD in humans can also evoke changes in the symmetry of cortical activity (Moore, 1990). In addition, a study in congenitally deaf cats suggests that monaural stimulation with a CI in case of bilateral deafness may "partially salvage[d] auditory nerve synapses on the unimplanted side" (Kral et al., 2013). In the present study, the good outcomes obtained by the five participants with long-term UD ear may be explained by: 1) the limited changes occurring in the auditory pathways following a post-lingual HL; and/or 2) the binaural convergence in the auditory system occurring via the hearing ear which may have maintained the functional integrity of the deaf ear.

The results of the present study showed that CI recipients with more than 25 years of UD duration can achieve a significant improvement in speech discrimination in noise in the three most challenging spatial configurations –
S₀/N₀, S₀/N_HE and S_CI/N_HE. The improvement was also evident in the results of the SSQ questionnaire assessing self-perceived daily hearing abilities, and with the sentence recognition test when using the implanted ear alone. The improvement was measurable despite the previous suggestion that speech tests measuring the benefits of implantation in UD may not be sensitive enough to show significant improvement as patients with monaural hearing may present with a low mean SNR (Vermiere & Van de Heyning, 2009). It is assumed that those with longer duration of deafness may have even greater abilities to hear speech in noise as they may have developed mechanisms to compensate for some of the difficulties imposed by UD.

This study used adaptive speech in noise testing to assess outcomes in order to overcome the risk of a ceiling effect. It also compared the results of implantation in long-term unilaterally deaf patients to the studies of bilateral implantation and normal hearing individuals to appreciate the magnitude of hearing abilities gained with the CI. As shown on Figure 6.2, Donaldson et al. (2009) reported that normal hearing individuals scored on average -1.6dB SNR on the BKB-SIN test when speech and noise are presented from front (S₀/N₀) while Etymotic Research (2005) indicated an average of -2.5dB SNR. In comparison, our group of five subjects needed much higher SNRs (3 to 9dB) before implantation, but their SNRs required to recognise speech in noise decreased (3.5 to -1dB) towards normal hearing values following implantation. As expected, our group with UD demonstrated substantially lower SNRs than CI users with bilateral HL (Litovsky et al., 2006; Donaldson et al., 2009).

The improvement in speech perception was substantially greater when noise was presented to the hearing-ear and the speech was presented from the front (S₀/N_HE) or from the CI side (S_CI/N_HE). These configurations are considered
the most challenging scenarios by this population, leading to positive subjective outcomes. With the SSQ questionnaire, improvement was most notable for spatial hearing abilities. Nonetheless, in the three conditions, participants with UD had poorer abilities than individuals with bilateral CIs and their abilities improved following implantation towards values obtained by individuals with normal hearing (Figure 6.4).

Although this study is limited by the small number of participants, it provides a valid argument that patients with post-lingual UD should not be excluded as CI candidates on the basis of long deafness duration. Further investigation is warranted and may support the studies demonstrating that residual hearing in one ear leads to better outcomes with an implant placed in either ear (Boisvert et al., 2011).

Furthermore, the impact of factors such as patient motivation, rehabilitation technique in combination with emerging speech-coding strategies warrant further investigation in order to identify predicting factors for outcomes of cochlear implantation in UD.

6.6. Conclusion
This study shows that deafness duration should not be considered the sole determinant factor for success of cochlear implantation in patients with post-lingual UD. The patients, in this study, with more than 25 years of UD obtained significant improvement of speech perception scores and subjective evaluation of hearing. Speech perception scores improved towards values presented by people with normal hearing. Furthermore, the scores of subjective evaluation of hearing improved towards those reported by bilateral CIs users and those with normal hearing.
7. Assessment of fine structure processing strategies in unilaterally deafened CI users

This work was published in: Távora-Veira D, Rajan, GP. Assessment of fine structure processing strategies in unilaterally deafened cochlear implant users. Int J Otol Head Neck Surg 2014.

7.1. Abstract

This study aimed to investigate the speech perception and subjective preference of unilaterally deafened CI users for two different speech coding strategies. Thirteen subjects who received a CI were provided with two maps that differed in the speech coding strategy, FS4 or FS4-p (MED-EL). Subjects were requested to alternate between the two maps daily for two weeks and to complete a questionnaire daily. Speech perception testing was performed using the adaptive BKB-SIN after 2 weeks of alternating FS4/FS4-p use.

The subjective benefit of FS4-p was significantly greater than the subjective benefit of FS4 on all five questions of the questionnaire. There was a significant improvement in speech perception scores over time under the S₀/N₀, S₀/N₁HE, S₁CI/N₁HE test conditions. There were no significant difference between the speech perception scores obtained with FS4 and FS4-p coding strategies. For this group of CI recipients, assessment of the subjective preference for the speech coding strategy is likely to enhance motivation, compliance and consequently, outcomes.

7.2. Introduction

The technology and surgical techniques for auditory implants have advanced rapidly in recent years enabling us to treat various types and degrees of HL. In addition, there is a continuous interest in improving signal processing and
speech coding strategies. The improvements are designed not only to improve CI users’ speech understanding in quiet and in noise, but to enhance appreciation of music, and speech understanding in tonal languages.

Various speech coding strategies have been developed, all of which aim to provide CI users with the clearest and most natural sound possible given the constraints of the limited stimulation representation of the implant electrodes. Envelope representation of an incoming sound signal is a common theme in many speech-encoding strategies, examples of which include Continuous Interleaved Sampling (CIS) (Wilson et al., 1991), HiResolution (HiRes) (Koch et al., 2004), and Advanced Combination Encoder (ACE) (Kiefer et al., 2001). One of MED-EL’s (Innsbruck, Austria) coding strategies, called Fine Structure Processing (FSP) (Zierhofer, 2003), uses envelope representation (High Definition-CIS) and low frequency temporal information to improve subtle pitch discrimination and temporal cues in the low frequencies (Muller et al., 2012; Moore et al., 2008).

There are reports of a general subjective preference for FSP over CIS+ coding strategies as well as a better appreciation of music using FSP (Lorens et al., 2010). The presentation of the fine structure is thought to enhance CI users’ music appreciation, and speech understanding in noise (Vermiere et al., 2010). Arnoldner et al. (2007) reported that speech and music perception was improved with FSP when compared to CIS in the early stage of the study, but this improvement was not statistically significant at the 12 month follow-up (Riss et al., 2008). FS4 and FS4-p, developments of the FSP coding strategy, both have fine structure information delivered to designated low-frequency apical channels which can span 70-950Hz. While FS4 can stimulate just one low-frequency fine structure channel at any point in time, FS4-p can
simultaneously stimulate two of the four fine structure channels at any given
time and can thus provide the temporal code specific to each of the two
channels with higher accuracy. Recently, Riss et al. (2014) compared FS4
and FS4-p with FSP in terms of speech perception, sound quality and
subjective preference. It was found that there was no significant difference
among the three strategies for speech performance in noise. At the end of the
study, 20 out 33 participants chose FS4 or FS4-p over FSP.

In the last few years, several studies have investigated the benefits of cochlear
implantation in individuals with UD. There is a growing literature
demonstrating that cochlear implantation decreases tinnitus disturbance
associated with UD, improves speech understanding in noise, enhances
localisation ability and improves patients’ self perception of hearing
performance (Van de Heyning et al., 2008; Vermeire & Van de Heyning,
2009; Buechner et al., 2010; Arndt et al., 2010; Stelzig et al., 2011; Firszt et
al., 2012; Hansen et al., 2013; Tavora-Vieira et al., 2013).

Unilaterally deafened subjects commonly expect to match the hearing from
the CI to their normal hearing in the contra-lateral ear. However, to best of
our knowledge, there are no studies that address whether unilaterally
deafened CI users demand different mapping techniques or modification of
map parameters.

Unilaterally deafened CI users are in the unique position of being able to
assess and compare the quality of speech coding strategies directly with the
correlating sound percepts of their normal hearing ear. In this study, it was
proposed that subjective evaluation of sound quality, and ease/effort of
listening should also be explored, as the speech perception tests in isolation
are insufficient to address these dimensions of hearing. Therefore, the present study set out to evaluate how unilaterally deafened CI users subjectively perceive and rate sound when using two different speech coding strategies. It aimed to investigate if the differences between FS4 and FS4-p had a subjective benefit for unilaterally deaf CI users. We expected subtle differences between FS4 and FS4-p, and therefore formulated an open questionnaire that aimed to obtain information regarding the subjective perception.

7.3. Methods
The methods used in this chapter were described in Chapter 2.

7.4. Results
Figure 7.1 illustrates the difference between FS4 and FS4-p for each question presented to the unilaterally deafened CI users. The speech coding strategy FS4-p scored significantly higher than FS4 for all five questions. The results are shown in Table 7.1.

Figure 7.1. Group results for each of the five questions presented to the unilaterally deafened CI users (n=13)
FS4 is shown in grey boxes. FS4-p is shown in diagonally lined boxes. Mean values are depicted as black squares, median as horizontal lines, and asterisks are the outliers (calculated as 1.5 to 3 times box height above the 75th percentile).

Table 7.1. Wilcoxon signed-rank test results for questions 1-5

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<tr>
<td>p-value (2-sided)</td>
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<td>0.008</td>
<td>0.057</td>
<td>0.010</td>
<td>0.004</td>
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There was a significant improvement across all test intervals in the BKB-SIN under the $S_0/N_0$ test condition ($p<0.001$) (Figure 7.2). The improvement in the BKB-SIN under the $S_0/N_0$ test condition was significant between pre-operative testing and the P1 program test ($p=0.001$); and significant between pre-operative testing and the P2 program test ($p=0.001$). There was no significant difference between P1 and P2 in the BKB-SIN under the $S_0/N_0$ test condition ($p=1.000$).

There was a significant improvement across all test intervals in the BKB-SIN under the $S_0/N_{HE}$ test condition ($p=0.002$) (Figure 7.2). The improvement in the BKB-SIN under the $S_0/N_{HE}$ test condition was significant between pre-operative testing and the P1 program test ($p=0.005$); and significant between pre-operative testing and the P2 program test ($p=0.007$). There was no significant difference between the P1 and P2 in the BKB-SIN under the $S_0/N_{HE}$ test condition ($p=0.763$).

There was a significant improvement across all test intervals in the BKB-SIN under the $S_{CI}/N_{HE}$ test condition ($p<0.001$) (Figure 7.2). The improvement in the BKB-SIN under the $S_{CI}/N_{HE}$ test condition was significant between pre-operative testing and the P1 program test ($p=0.002$); and significant between pre-operative testing and the P2 program test ($p=0.002$). There was no significant difference between the P1 and P2 in the BKB-SIN under the
$S_{CI/N_{HE}}$ test condition ($p=0.157$). The majority of the patients (10 out of 13) kept either FS4 or FS4-p at the end of the study.

Figure 7.2. Results of the BKB-SIN ($n=13$)

Speech presented from the front and noise presented from the front ($S_0/N_0$); speech presented from the front and noise presented from the side of the normal hearing ear ($S_0/N_{HE}$), and; speech presented from the side of the CI and noise presented from the side of the normal hearing ear ($S_{CI}/N_{HE}$).

Mean values are depicted as black squares, median values as horizontal lines, and dots signify outliers (1.5 to 3 x box height above the 75th percentile).

7.5. Discussion

Several studies have demonstrated that cochlear implantation is a suitable hearing rehabilitation option for adults with unilateral profound deafness. Among the hearing devices used in the rehabilitation of UD subjects, CI is the only option that provides ear specific information and thus potentially the benefits of binaural hearing. The studies have investigated the effects of cochlear implantation on tinnitus, subjective perception of improvement, speech understanding in noise and localisation ability (Van de Heyning et al., 2008; Vermeire & Van de Heyning, 2009; Buechner et al., 2010; Arndt et al., 2010; Stelzig et al., 2011; Firszt et al., 2012; Hansen et al., 2013; Tavora-Vieira et al., 2013). A review of the literature by Vlastarakos et al.
(2013) has emphasised that self-assessment questionnaires were commonly used to assess the patients’ perception of improvement in daily listening conditions. In fact, Stelzig et al. (2011) reported that the subjective rating of outcomes tended to be more positive than the objective measures which could be related to an inadequacy of speech perception test for the unilaterally deafened CI users.

To date, there is no literature that addresses whether the patients with UD demand any different mapping strategies or mapping parameters. This study addressed this issue by comparing the speech coding strategies FS4 and FS4-p in terms of patients’ performance in the adaptive speech in noise test and the patients’ responses to a non-standardised questionnaire. The questionnaire was developed by the authors and aimed to determine the subjective perception of sound from the CI.

The results showed a significant improvement in speech perception in noise scores when speech and noise are presented from the front ($S_0/N_0$), and when speech is presented from the front ($S_0/N_{HE}$), or from the CI side ($S_{CI}/N_{HE}$) with the noise presented to the normal hearing ear. This was true for both speech coding strategies. Riss et al. (2014) found that there was no significant difference among the three strategies FSP, FS4 and FS4-p using an adaptive sentence test in noise. Similarly, in this study, there was no significant difference between FS4 and FS4-p in the speech understanding measures. These finding were expected as the difference between FS4 and FS4-p is subtle.

Outcome performance studies (which investigated the superiority of one speech coding strategy over another) have predominantly focused on speech
perception, for which objective speech perception testing is appropriate. Unilaterally deafened CI users are in the unique position of being able to assess and compare the quality of speech coding strategies directly with the correlating sound percepts of their normal hearing ear. Therefore, it was proposed that subjective evaluation of sound quality and ease/effort of listening should be added to the evaluation protocol with the final objective to facilitate patients’ acclimatisation to electrical stimulation.

The results indicated that FS4-p was rated significantly superior to FS4 in all five questions answered by the unilaterally deafened CI users. As per question 1, it appears that unilaterally deafened CI users perceived that FS4-p mimicked the sound quality of the normal hearing contra-lateral ear significantly better than FS4. This was reinforced by the rating in question 5, since the patients reported to like the sound provided by the CI more when using FS4-p. The explanation for these results is not clear. As the patients alternated between the two strategies daily, they had the same experience with both settings and thus it unlikely that the patients have acclimatised to one setting in particular. To avoid any bias, the audiologist performing the speech in noise test was blind to which speech coding strategy was being used.

The FSP strategy with its fine structure coding (Schatzer et al., 2010) aims to improve pitch perception, which is thought to improve speech discrimination, sound localisation, and music appreciation (Lorens et al., 2010). The original FSP strategy provides fine structure processing in one to three apical channels up to 470Hz. The newer developed FS4 and FS4-p provide it to the four most apical low-frequency channels. FS4 stimulates the apical channels sequentially, while FS4-p simultaneously stimulates two of the four
designated low frequency apical channels from 70-950Hz, and this is thought to further enhance temporal information. This may explain why CI users preferred FS4-p to FS4.

The subjective results in this study differ from those reported by Riss et al. (2014) since our group of unilaterally deafened subjects rated FS4-p superior to FS4. This difference might be linked to the variability of the subjects between the studies. The subjects with UD might have rated the sound in comparison to normal acoustic hearing, while patients with bilateral HL may rate the sound quality based solely on their auditory memory and/or an input from acoustic amplification on the contra-lateral ear.

The results of this study need to be interpreted with caution, as it used a non-validated questionnaire and a small number of subjects. However, it is the first to provide some insights about the mapping strategies to be considered for patients with UD. Combined with speech perception testing, assessment of the patients’ subjective preference for a specific speech coding strategy may assist in the rehabilitation program for UD, enhancing patients’ motivation and compliance with CI use.

7.6. Conclusion
There were no significant differences in the speech perception in noise scores between FS4 and FS4-p. The FS4-p fine structure was rated higher subjectively than FS4 in the present study. Subjective evaluation may assist in the rehabilitation program of unilaterally deafened CI users potentially enhancing motivation, compliance, and, consequently, outcomes.
8. Cochlear implantation in children with congenital and non-congenital UD – a case series

This work was published in:

8.1. Abstract

Objectives: Cochlear implantation is rapidly gaining acceptance as the most effective treatment for adult patients with UD. The benefits for the paediatric population remain to be investigated. This study aimed to investigate the implications of cochlear implantation in children with congenital and non-congenital UD.

Design: Four children, three with congenital and one with a sudden UD, were studied after implantation. The children were aged 17 months, 4.5 years, 6.8 years and 9 years old at the time of implantation. Speech perception in noise, and sound localisation ability were evaluated using age appropriate materials.

Results: The child with post-lingual UD rapidly integrated the normal acoustic hearing with the electrical signal from the CI and showed binaural benefits, as indicated by the localisation ability and the improvement of speech perception in noise scores. The younger child with congenital UD showed some clinical evidence of binaural integration and the two older children with congenital deafness have not yet indicated signs of binaural benefits.

Conclusion: It appears that cochlear implantation in children with congenital UD may provide some of the benefits of binaural hearing if implantation occurs within the critical period for bilateral auditory development.
8.2. Introduction

CIs are well accepted as the most suitable hearing device that enables children with bilateral deafness to achieve speech and language development within the normal range. Multiple studies have demonstrated the advantages of early cochlear implantation in hearing rehabilitation of children with bilateral deafness (Dettman et al., 2007; Niparko et al., 2010; Gilley et al., 2008; McConkey et al., 2004).

It is thought that poor speech and language outcomes of cochlear implantation in older children and adults with prelingual deafness are the result of cross-modal plasticity involving the auditory pathway. This has been demonstrated in animal and human studies (e.g. Park et al., 2010; Lomber et al., 2010; Lee et al., 2001; 2003; Kral & O’Donoghue, 2010; Kral & Sharma, 2012). These changes can potentially become permanent in children with congenital deafness if there is a long delay before cochlear implantation (Sharma et al., 2005; Kral et al., 2006; Gordon et al., 2005) and it is well accepted that there are critical periods in child development in which the benefits from cochlear implantation are more likely to be optimal (Sharma et al., 2005; 2007; 2009). Recently, Leigh et al. (2013) reported on the long-term follow-up of a group of children who received a CI prior to 12 months of age and compared the results to CI recipients implanted at age 13-24 months and normal hearing children. The results showed that the children implanted before 12 months showed language growth rates equivalent to their hearing peers.

Gordon et al. (2013a) argued that unilateral development of the brainstem for more than 2 years promotes asymmetrical bilateral auditory pathways. Investigations of the benefits of early bilateral cochlear implantation in deaf
children showed that one CI is enough to promote speech and language development. However, the outcomes of binaural hearing are superior (Chadha et al., 2011), particularly if implantation occurs simultaneously or with a short inter-implant interval (Gordon et al., 2011; 2013a; Gordon & Papsin, 2009).

In recent years, several studies have investigated the benefits and implications of cochlear implantation for UD in adults (Vermeire & Van de Heyning, 2009; Arndt et al., 2010, Buechner et al., 2010; Stelzig et al., 2011; Firszt et al., 2012; Távora-Vieira et al., 2013a; 2013b). It appears that there is an increasing awareness and acceptance that UD warrants intervention.

Cochlear implantation for UD was first introduced to treat tinnitus (Van de Heyning et al., 2008). Cochlear implantation for unilateral post-lingual deafness is currently the focus of various studies, which demonstrate significant improvement of speech understanding in noise, subjective measures of hearing improvement, tinnitus suppression, and enhancement of quality of life (Vermeire & Van de Heyning, 2009; Arndt et al., 2010, Buechner et al., 2010; Stelzig et al., 2011; Firszt et al., 2012; Távora-Vieira et al., 2013a; 2013b; Punte et al., 2011; Ramos et al., 2011). As a result, various countries have approved CI as treatment of UD and tinnitus. In the paediatric population, UHL has been linked to delayed speech and language development (Bess & Tharpe, 1986; McKay et al., 2008; Lieu et al., 2010) with subsequent psychosocial problems and poorer academic performance (Tharpe et al., 2008; Lieu et al., 2012). Attempts to provide hearing rehabilitation via bone conduction devices or conventional CROS devices are of limited value and difficult to implement in children under 5 years (Christensen et al., 2010). While options such as CROS hearing aids and
bone-conduction hearing implants can improve overall hearing, a CI is the only option that can potentially provide the benefits of binaural hearing. Consequently, the number of parents of unilaterally deafened children seeking advice regarding CI is increasing rapidly. Unfortunately, there is limited literature regarding cochlear implantation for hearing rehabilitation of unilaterally deafened children.

Therefore, the present study was initiated to investigate the implications of cochlear implantation in children with UD. This report presents the preliminary results of a group of four children; including three children with congenital UD treated with CI.

8.3. Methods
The methods used in this chapter were described in Chapter 2.

8.4. Results
Four children were included in this preliminary report.

Subject 1
Subject 1 (S1) was aged 17 months and had congenital HL. The subject wears the speech processor on a full-time basis. S1 accepted the speech processor within the first 2 weeks of initial stimulation, only removing it in the presence of loud sound. S1 had weekly visits to the speech therapist over the first 3 months followed by fortnightly sessions. However, S1 was too young to be assessed with the formal measures.

Subject 2
Subject 2 (S2) was aged 4.5 years and had congenital HL. S2 had difficulty accepting the speech processor and he is currently a non-user. Although, the
child wore the speech processor for approximately 7 weeks no audiological evaluation measures were obtained.

Subject 3

Subject 3 (S3) was aged 6.8 years and had congenital HL. The subject wears her speech processor on a full-time basis. Several trials of speech recognition in the CI-only condition were performed with S3 using either direct audio input or the FM system. S3 was only able to perceive that a sound was presented through the CI alone, but could not discriminate between different sounds. Auditory stimulation was perceived as vibration only. Speech perception in noise scores with the CI on did not differ to those with the CI off (Figure 8.1 – 1A). There was no difference in the RMS error between CI on and CI off conditions at 6 and 12 months post-CI, indicating that S3 did not achieve any improvement in localisation ability (Figure 8.2 – 2A).
Figure 8.1. Speech perception in noise scores

Subject 4

Subject 4 (S4) was aged 9 years and had sudden UD post-meningitis. The subject continues to wear his speech processor on a full-time basis 12 months post-implantation. S4 showed a substantial improvement in speech recognition in noise in all three spatial configurations as shown in Figure 8.1 – 1B. S4 achieved a remarkable improvement in sound localisation performance as shown in Figure 8.2 – 2B.
8.5. Discussion

Very little is known regarding the outcomes of cochlear implantation in children with UD, and this study is to our knowledge the first to report the outcomes of congenital UD and CI.

In our study, S4 was the only post-lingual CI recipient. S4 was implanted 5 weeks after losing his hearing unilaterally due to meningitis. S4 achieved substantial improvement in speech perception in noise scores and localisation ability from pre- to post-CI. Similarly, Hassepass et al. (2013) presented the results of three children who received a CI as a treatment for post-lingual UD. The patients were aged 4, 10, and 11 years, and two of the patients had a short
duration of deafness of 5 and 18 months. All three children were reported to wear the speech processor consistently (>8 hours/day), and the results indicated binaural hearing benefits for speech recognition in noise and localisation. Likewise, Plontke et al. (2013) reported a case of an 8-year-old boy who received a CI for UD 5 months after a lateral skull-base fracture. Six months following initial activation, he showed significant improvement for speech understanding in noise and localisation.

In the present study, S3 had fairly good speech perception in noise prior to cochlear implantation, whereas S4 required a more positive SNR. S4 achieved SNRs improvement that are higher than those reported by Hassepass et al. (2013). However, his performance post-CI was still not as good as the pre-implantation performance of S3. We suspect this was related purely to coping strategies that we see clinically in subjects with congenital deafness. In contrast S4 had a recently acquired HL and did not have the time to develop such strategies. In adult populations with post-lingual UD, bilateral integration of acoustic hearing (normal hearing-ear) and the electrical signals of the CI in the deaf ear seems to be feasible, even in patients with long-term deafness duration (Tâvora-Vieira et al., 2013b). Therefore, we assume that children with post-lingual UD of short duration would demonstrate rapid binaural integration and benefits from their CI because the bilateral auditory pathways are established and intact.

In the present study localisation results were available for two children (S3 and S4). S4 achieved a remarkable improvement in sound localisation performance post-implantation. In contrast, S3 was not able to localise the sound source with or without CI.
S3’s results did not indicate any evidence of binaural integration, or clear benefit of binaural hearing over monaural hearing. Although S3 describes the electrical stimulation as “vibration”, she has been wearing the implant for over 12 months at the time of this study and was satisfied with it. She has all electrodes activated and there is no discomfort with stimulation. Reports from family suggested that it is the feeling of “fullness” and “being complete” that explained her consistent CI use. S1 is the only one of the three congenitally deaf children that appears to benefit from the CI, although he is too young to be assessed on formal measures at this time. It will be interesting to track S1’s auditory development with the CI as he continues to mature.

Bilateral CIs aim to restore the advantages of binaural hearing such as sound localisation skills, binaural summation, and enhancement of speech discrimination, particularly in noise (Ching et al., 2006; Litovsky et al., 2009; Basura et al., 2009). However, there is a critical period for the establishment of binaural integration (Gordon et al., 2011; 2013a; Kral et al., 2013). Even though long-term auditory stimulation through a CI enables auditory development at brainstem and cortex level (Gordon et al., 2007; 2008; 2010), children receiving “CIs simultaneously have the best chance of developing bilateral auditory pathways capable of processing binaural cues” (Gordon et al., 2011). In the case of unilateral CI stimulation, the contra-lateral auditory deprivation may cause reorganisation of the auditory cortex, potentially disrupting the bilateral auditory pathways (Gordon et al., 2013b).

It is proposed that this sensitive period may last 1.5 years for binaural auditory development (Gordon et al., 2013b), and therefore, the time between sequential implantations should not exceed that. Grothe et al. (Grothe et al., 2010) explained that in the case of unilateral cochlear implantation there is an
abnormal strengthening of the ipsi-lateral pathways as a consequence of the absence of inhibition that would come from the opposite ear. In this case, children with congenital UD might permanently lose their opportunity to attain bilateral integration of hearing benefits after the critical period. To avoid this, we suggest that early intervention for children with congenital UD might be the best approach to regain binaural hearing functions.

The authors recognise the limitations of this study, which includes small number of subjects and lack of binaural hearing assessment for S1. However, based on the previously listed studies showing the influence of long-term monaural stimulation in bilaterally deafened subjects, this report offers an insight regarding the application of CI in congenital UD. It is possible that S2 and S3 have missed the sensitive period for binaural hearing development due to the unilateral stimulation from the normal hearing ear, which translates into S2 not being able to use the implant and S3 not achieving any signs of CI benefits to date.

The influence of long-term electrical stimulation remains to be verified overtime for S3. It is important to consider that current test protocols may not be sufficiently sensitive to show the full benefit of cochlear implantation for S3 given the excellent speech perception scores in the unaided condition.

8.6. Conclusion
Children with post-lingual UD may be able to rapidly integrate acoustic normal hearing and the electrical signal from a CI.
Cochlear implantation in children with congenital UD may provide some of the benefits of binaural hearing if implantation occurs within the critical period for bilateral auditory development.

Based on these case reports, the period of auditory deprivation for the ear with congenital deafness should be reduced to enable binaural development of auditory pathways and prevent permanent reorganisation of the auditory functions.
9. **General discussion**

9.1. **Background**

Historically, the handicap associated with UD has been underestimated (Tharpe *et al*., 2008; Wiley *et al*., 2011). The prevalence of UHL varies from 3% to 6.3% depending on how it is defined (Tharpe *et al*., 2008; Oyler *et al*., 1988). However, the prevalence rates of UD are unknown. Treatment options are limited to: (1) development of compensation strategies; (2) use of FM systems; and (3) transfer of sounds from the deaf side to the hearing ear via conventional CROS hearing aids or bone conduction implant. Recently, the research focus on CI for the treatment of UD has increased due to the rapid expansion of CI candidacy criteria and increased awareness of the consequences of UD.

9.1.1. **CIs as a treatment for tinnitus associated with UD**

CI is considered to be the most effective treatment option for severe to profound bilateral HL. CI was first used as a treatment for tinnitus in patients with UD in 2008 (Van de Heyning *et al*., 2008).

Prior to undertaking the experimental work for this thesis, only three published studies had investigated the use of CI for UD with associated tinnitus. Vermiere *et al.* (2009) reported on the hearing outcomes of 20 patients. These comprised 11 subjects with normal hearing in the opposite ear and nine with some degree of HL for which they wore hearing aids. The CI led to significant improvement in speech understanding when it was presented from the CI side and noise from the front. There was also significant improvement of self-perceived benefits of binaural hearing on speech understanding, quality of hearing and spatial hearing.
In the second study, Arndt et al. (2010) compared the hearing performance of 11 patients in the unaided condition, using BAHA, using conventional CROS and 6 months after receiving a CI to treat their UD. Localisation, tinnitus disturbance and subjective improvement of hearing were also assessed. Speech comprehension was significantly improved with CI when speech was presented to the implanted side and noise to the normal hearing ear. There were significant differences between the treatment options when speech and noise were presented from the front. CI was significantly better than CROS and BAHA when noise was presented from the aided side, but there was no improvement when compared to the unaided condition. Localisation error was significantly reduced with CI. Subjective self-rated hearing performance and quality of life were significantly better with CI compared to unaided, CROS and BAHA for speech hearing and spatial hearing subsections. Furthermore, tinnitus intensity was significantly reduced 6 months after implantation when the CI was on.

In the third study, Buechner et al. (2010) reported the outcomes for five patients with UD and tinnitus. For three of these patients, CI provided significantly better speech perception scores when speech was presented from the front and noise from the normal hearing ear. In addition to these studies, Kleinjung et al. (2009) reported the successful use of CI for tinnitus treatment caused by sudden UD.

During the course of this thesis, further studies supporting the benefits of CI on tinnitus suppression for UD patients were published. Punte et al. (2011) reported positive long-term outcomes of CI on tinnitus suppression. Ramos et al. (2012) studied 10 adult patients with UD of duration ranging from 1-5
years. Two patients experienced total tinnitus suppression and seven patients reported that CI helped to decrease tinnitus perception.

In a review of CI for tinnitus suppression, Arts et al. (2012) reported that CI may represent a treatment option for people with tinnitus caused by unilateral cochlear deafferentation. They found no reports of an increase in tinnitus following CI (Arts et al., 2012). Table 9.1 presents a summary of the publications on UD to date.

Table 9.1. Published studies that investigated the benefits of CI in UD with or without associated tinnitus

<table>
<thead>
<tr>
<th>Authors</th>
<th>No. recipients</th>
<th>Study type</th>
<th>Implant manufacturer</th>
<th>Research focus</th>
</tr>
</thead>
<tbody>
<tr>
<td>Van de Heyning et al. (2008)</td>
<td>21</td>
<td>Prospective</td>
<td>Med-El</td>
<td>Tinnitus</td>
</tr>
<tr>
<td>Vermeire &amp; van de Heyning (2009)</td>
<td>20</td>
<td>Prospective</td>
<td>Med-El</td>
<td>Hearing</td>
</tr>
<tr>
<td>Buechner et al. (2010)</td>
<td>5</td>
<td>Prospective</td>
<td>Advanced Bionics</td>
<td>Tinnitus / Hearing</td>
</tr>
<tr>
<td>Kleinjung et al. (2009)</td>
<td>1</td>
<td>Case report</td>
<td>Med-El</td>
<td>Tinnitus</td>
</tr>
<tr>
<td>Arndt et al. (2010)</td>
<td>11</td>
<td>Prospective</td>
<td>Cochlear Corporation</td>
<td>Tinnitus / Hearing</td>
</tr>
<tr>
<td>Ramos et al. (2011)</td>
<td>10</td>
<td>Prospective</td>
<td>Cochlear Corporation</td>
<td>Tinnitus</td>
</tr>
<tr>
<td>Stelzig et al. (2011)</td>
<td>5</td>
<td>Case series</td>
<td>Med-El</td>
<td>Hearing</td>
</tr>
<tr>
<td>Firszt et al. (2012)</td>
<td>3</td>
<td>Case series</td>
<td>Cochlear Corporation</td>
<td>Hearing</td>
</tr>
<tr>
<td>Arts et al. (2012)</td>
<td></td>
<td>Literature review</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hansen et al. (2013)</td>
<td>19</td>
<td>Prospective</td>
<td>Cochlear Corporation</td>
<td>Tinnitus / Hearing</td>
</tr>
<tr>
<td>Plontke et al. (2013)</td>
<td>1</td>
<td>Case study</td>
<td>Advanced Bionics</td>
<td>Hearing Paediatric</td>
</tr>
<tr>
<td>Hassepass et al. (2013)</td>
<td>3</td>
<td>Prospective</td>
<td>Cochlear Corporation</td>
<td>Hearing Paediatric</td>
</tr>
<tr>
<td>Vlastarakos et al. (2014)</td>
<td></td>
<td>Literature review</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
9.1.2. Hearing improvement provided by CI in UD

Stelzig et al. (2011) was the first to investigate the outcomes of cochlear implantation in patients without tinnitus. They reported on a series of five adult patients who received CI for the treatment of UD. A battery of speech tests in varied conditions was used to investigate the benefits of binaural hearing (acoustic hearing + CI) over monaural hearing (acoustic hearing alone). Binaural hearing was found to be better than monaural hearing for these patients and the subjective rating of benefits was more prominent than the objective assessment.

Firszt et al. (2012) reported the outcomes for three adult patients with less than 5 years of UD. CI improved the localisation and spectral difference for all participants and improved temporal difference discrimination for two patients. Word recognition in noise was significantly better for two patients, but sentence recognition in noise scores did not show significant improvement. Patients also reported subjective hearing improvement.

Hansen et al. (2013) reported the outcomes for patients receiving CI as treatment for UD, including subjects with Meniere’s disease. Speech in quiet was administered to the implanted ear and showed that CI could restore auditory function to the deaf ear. Sound localisation was also improved in most patients in an experience-dependent manner and tinnitus perception decreased after implantation.

A comprehensive review of the studies on CI in UD was recently published by Vlastarakos et al. (2014). At the end of 2013, a total of 108 patients with UD had received a CI, comprising 66 with hearing difficulties and 42 with tinnitus. This review concluded that CI appears to improve sound
localisation, speech perception in noise when presented to the implanted ear, subjective perception of speech and spatial hearing, as well as decreasing tinnitus perception.

9.1.3. First paediatric experience
Very limited knowledge exists with regards to CI for UD in the paediatric population. Hassepass et al. (2013) presented results for three children who underwent CI as a treatment for non-congenital UD. They were aged 4, 10, and 11 years, and two had a short duration of deafness of 5 and 18 months. All three children were reported to wear the audio processor consistently (>8 hours/day) and the results indicated binaural hearing benefits for speech discrimination in noise and localisation. Limited objective information was obtained for the 4-year-old child enrolled in the study. Plontke et al. (2013) also reported the case of an 8-year-old boy who received a CI for UD at 5 months after a lateral skull-base fracture. The child showed significant improvement in speech discrimination in noise scores at 6 months post-CI, with localisation also showing improvement.

Both of the above studies showed that CI could improve speech understanding in noise, localisation and subjective hearing performance. The children in both reports had non-congenital deafness.

9.1.4. Aims of this work
Although several studies had demonstrated that CI may be a feasible option for tinnitus suppression in UD, there was still only scant knowledge regarding hearing outcomes from CI treatment for UD. These studies were limited by small numbers of subjects and the use of a variety of methods for outcome evaluation. They offered little insight into factors that may influence
outcomes from CI and did not address the candidacy criteria for implantation. Furthermore, the literature lacks long-term data and information relating specifically to the paediatric population. Table 9.1 summarises the findings from studies that preceded this thesis, as well as those that were published during the course of this study.

In view of the preceding work, the aims of this thesis were:

1. To determine the impact of CI on UD with and without tinnitus in a large number of patients.

2. To investigate some of the factors that could influence the outcomes.

3. To verify the long-term benefits of CI in UD.

4. To further explore the use of CI in the rehabilitation of paediatric populations with UD.

A comprehensive evaluation was carried out in terms of speech discrimination in noise, spatial acuity, tinnitus suppression and subjective hearing improvement. The implications of duration of deafness were analysed and the speech coding strategy that provided the best sound quality was investigated. In addition, rehabilitation strategies were described and the use of CI in congenital UD in the paediatric population was evaluated for the first time.

9.2. Major findings

Twenty-eight adults and four children with unilateral profound sensorineural HL, including 13 adult patients with tinnitus, received a CI in this prospective
study. Similar to previous reports, all patients used their CI on a full-time basis, thus demonstrating their overall satisfaction with the benefits.

Speech discrimination in noise improved significantly over time in all spatial setups ($S_0/N_0$, $S_0/N_{HE}$ and $S_{CI}/N_{HE}$). Significant improvement in speech perception in noise scores has been reported in most previous publications. However, as there is no established protocol for assessment of this group of patients, direct comparison with previous work is difficult since the spatial configuration used varied amongst the studies. Vermeire et al. (2009) demonstrated that speech discrimination was significantly improved when speech was presented to the CI side and noise from the front. This setup was not used in the current study. Arndt et al. (2010) reported greater improvement in speech understanding when speech was presented to the CI side and noise to the normal hearing ear ($S_{CI}/N_{HE}$) which was similar to this study. Furthermore, Buechner et al. (2010) also showed that scores were better when speech was presented to the front and noise to the hearing ear ($S_0/N_{HE}$). The results of the present study concur with those of previous publications and indicate that patients with UD may gain some of the benefits associated with binaural hearing.

This study is the first to investigate whether duration of deafness and age at implantation can affect outcomes. These factors did not appear to influence the results obtained in all three spatial configurations.

The SSQ and APHAB measures of subjective outcomes both showed significant improvement of hearing with CI use. Analysis of SSQ scores confirmed significant improvement over time for all three subscales – speech, spatial hearing and quality of hearing. Similarly, Arndt et al. (2010) reported
significant improvement in speech and spatial hearing subscales, while Vermeire and Van de Heyning (2009) also reported improvement in all three subscales. Together, these results demonstrate that subjective perception of hearing improves with CI use in UD subjects.

The APHAB results showed improvement in the subscales for ease of hearing, reverberation and background noise. Scores for the aversiveness subscale did not change significantly before and after surgery. This demonstrates that CI in UD enhances the patients’ self-perception of hearing performance and does not incite any tolerance issues.

Our results showed that age at implantation and duration of deafness did not significantly affect speech perception in noise scores, subjective hearing performance as measured by SSQ and APHAB, or improvement of tinnitus (Tables 4.1-4.8) This confirmed our initial hypothesis that such factors do not play a major role in the rehabilitation of post-lingual UD using CI. The SSQ questionnaire was repeated at 24 months post-CI and the scores showed that subjective benefit remained stable in the long-term.

Tinnitus was present in 13 adult patients. The disturbance caused by tinnitus also decreased significantly in all subjects who had tinnitus prior to surgery. Further follow-up at 24 months showed the improvement was stable in the long-term. Similar to recent literature (Arts et al., 2014), no evidence was seen for an increase in tinnitus following implantation. Thus, CI can be an effective means of reducing tinnitus disturbance, as described in Chapter 4.

The present study was the first to investigate localisation acuity in a large number of CI users. Very few studies have investigated the localisation ability of unilaterally deafened CI recipients with a duration of deafness less
than 10 years. Arndt et al. (2010) investigated the localisation ability of 11 adults with UD and CI using a setup comprising of seven loudspeakers positioned at intervals of 30 degrees between -90 and 90 degrees. Hassepass et al. (2013) used a similar setup in a study of three children who received a CI to treat UD. Firszt et al. (2012) also reported the localisation error in a small group of three adults. Localisation testing was performed using 15 loudspeakers located 10 degrees apart and the results indicated that CI significantly improved localisation. In the present study, patients also performed significantly better with CI on than with CI off (Figure 5.1). Nevertheless, direct comparisons with published literature are not possible because of substantial differences in methodology, including the stimuli used and the number of speakers. The present study is unique in that it used two loudspeakers simulating an ILD. Therefore, the mechanism used by patients to localise the sound source is likely to differ from other studies.

With regard to age of implantation on the localisation results, the difference between CI off and CI on was significant for both age groups. However, the sample size for both age groups was relatively small and a larger number of cases would more accurately reflect the population mean. Gender and duration of deafness did not influence the results. However, it is important to note that of the six patients with >10 years of deafness duration, four lost their hearing after the age of 12 and this may have influenced the results. These findings were presented in Chapter 5.

Further investigation of the influence of deafness duration was performed in a cohort of five patients with >25 years of deafness. The speech perception in noise scores obtained in these patients were compared with results obtained in traditional CI users with bilateral HL, as well as in adults with normal
hearing. The results showed that 1 year after implantation, the scores obtained by individuals with UD improved towards those of individuals with normal hearing. The SSQ scores were compared to scores obtained by individuals with bilateral CI and to individuals with normal hearing. This comparison suggested that before implantation, individuals with long-term UD had poorer hearing abilities than those with bilateral CI. Following implantation, their abilities in the SSQ sections improved towards the scores obtained by individuals with bilateral CI and those with normal hearing. These results were described in Chapter 6 and suggest that duration of deafness should not be considered the sole determining factor for the success of CI in patients with post-lingual UD.

UD subjects are potentially the most demanding CI users, with a common expectation to match hearing from the CI with their normal hearing in the contra-lateral ear. Therefore, it was thought this group could provide additional information regarding the speech coding strategy to be used in order to achieve the most acceptable sound quality, as described in Chapter 8. With the Med-El CI system, patients preferred sound delivered by the FS4-p coding strategy, which is the newest development in fine structure processing (FSP). FS4-p can simultaneously stimulate two of the four fine structure channels, thus providing the temporal code specific for each of the two channels with higher accuracy. Temporal information is thought to enhance the users’ appreciation of music and of speech understanding in noise (Vermeire et al., 2010). This enhanced temporal information may have contributed to the improved sound quality provided by the CI.

In view of the positive outcomes achieved by adult recipients, four children including three with congenital UD (S29, S30, S31) and one with
non-congenital UD (S32) received a CI. They were aged 17 months, 4.5 years, 6.8 years and 9 years respectively at the time of implantation. The child with non-congenital UD had a similar rehabilitation pattern to that seen in the adult population. S29 had 17 months of unilateral auditory stimulation, while S30 had 4.5 years and S31 had 6.8 years of monaural hearing at the time of implantation. They all received the same implant and the same rehabilitation program from our CI team. However, the outcomes differed greatly. S30 did not persist in wearing the implant and the results obtained so far by S31 show no evidence of binaural integration or clear benefit of binaural hearing over monaural hearing. The youngest child in the group (S29) presented with the highlighted improvement among the congenitally deafened children and is able to understand speech in his CI alone condition. A sensitive period lasting 1.5 years has been proposed for binaural auditory development (Gordon et al., 2013b). It is possible that S30 and S31 missed this sensitive period for binaural hearing development due to unilateral stimulation from the normal hearing ear for a period longer than 1.5 years. Other factors such as child’s environmental condition, family support and commitment to auditory rehabilitation and also child’s general development should be always considered. This work was described in Chapter 7.

9.3. Limitations of this study and future research directions
CI is well established as a treatment for severe to profound bilateral HL. The use of CI for UD has only emerged in the past 5 years and the literature in this context is rapidly increasing. However, more prospective studies including a large number of patients as well as long-term data are necessary to establish CI as an efficient treatment of UD. The main limitation of this study is that the
trial was not randomised. A prospective randomised trial of CI versus other technologies would establish high-level evidence for the benefits of CI over less invasive treatments for UD.

This thesis demonstrated that age at implantation and duration of deafness do not influence the outcomes. The study cohort comprised patients with 6 months to 40 years of post-lingual deafness duration. Important future research topics include investigation of the role played by the onset of deafness and identification of the cutoff for onset and duration of deafness.

Unilaterally deafened CI users can give unique information regarding the sound quality provided by electrical stimulation. This can be used to develop speech coding strategies that mimic natural sound and could also be used for further investigations into electrode design and cochlear coverage benefits/limitations.

The patients included in this study received intensive auditory training and this may have contributed to improved outcomes. Further work is needed to evaluate the importance of auditory rehabilitation exercises. Another vast field of research is the use of CI in UD children, as very little literature has been published so far in this area. Therefore, future studies suggested by the results of this work include:

- Investigate CI use in congenital UD.
- Investigate how unilaterally deafened CI users perceive music.
- Investigate if the type and frequency of auditory training predict CI outcomes.
- Investigate if age of onset of deafness is a determining factor for CI outcomes.
- Investigate if the depth of electrode insertion is a determining factor for integration of electrical and acoustic stimulation.
- Investigate the cost/benefits associated with the use of a CI in UD patients.

9.4. Conclusions
CI has proven to be an effective treatment for UD with and without tinnitus.
The major results of the present study in subjects with unilateral severe-profound UD treated by CI can be summarised as:

1. Improved speech perception in noise in three spatial configurations including those reported as the most challenging for patients
2. Improved subjective perception of hearing performance as measured by two separate questionnaires
3. Decreased disturbance from tinnitus as assessed by the TRQ, which may in turn have contributed to the overall positive subjective impression of the benefits of CI
4. Significantly improved localisation acuity

This research showed that age of implantation and duration of deafness did not affect outcomes and that CI can significantly improve the quality of life in subjects with UD. Intensive auditory training is likely to contribute to the integration of acoustic and electric signals and to promote the appreciation of hearing benefits of a CI in subjects with UD. Finally, children with congenital UD may achieve binaural hearing development if the period of monaural hearing is short.
The major finding of this study can be summarised as:

- Unilaterally deafened CI users can integrate electrical and acoustic stimulation
- CI significantly improves speech perception in noise
- CI significantly improves subjective perception of hearing speech and spatial hearing, and the benefit remains significant up to 2 years after CI activation
- CI decreases tinnitus perception and the benefit remains stable up to 2 years post-implantation
- Localisation acuity is significantly improved with CI activated
- Age at implantation and duration of deafness do not appear to determine the outcomes
- Patients subjectively prefer to use a speech coding strategy that is believed to enhance temporal information
- Children may benefit from the “earlier the better” approach in order to establish a binaural hearing pathway.

The clinical implications of this study are:

- CI received TGA approval for treatment of UD in Australia in October 2013.
- Proposed clinical protocol for auditory training of UD CI users as described in Chapter 4.
- Creation of a UD national working group involving Australian-wide institutions to establish clinical protocols for assessment, rehabilitation and evaluation in the clinical setting.
Bibliography


