Review of pediatric cochlear implantation records at the Tygerberg Hospital – Stellenbosch University Cochlear Implant Unit from 1990 – 2018

by

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Declaration

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Abstract

The decision of parents of deaf children to proceed with cochlear implantation is often based on the expectation that the improved access to sound provided by the cochlear implant system will result in spoken language development that will improve the long-term educational development and occupational prospects of the child. These positive outcomes associated with pediatric cochlear implantation may result in financial benefits to society. A variety of factors may however determine the outcomes achieved. Cochlear implantation is an elective procedure and information about the expected outcomes and prognostic variables associated with outcome should be available to parents of candidates and funders to enable them to make informed decisions about their options.

The main aim of the study was to describe the demographic and pre-operative clinical characteristics of pediatric cochlear implant recipients in the Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit, the long-term post-operative trends in outcome in the domains of implant use, mode of communication, speech perception development, educational placement, occupational status and the variables associated with mainstream educational placement.

The records of 216 recipients who received cochlear implants between 1990 and 2014 were reviewed. The recipients were grouped according to the age at onset of severe to profound sensorineural hearing loss: 0 to 2 years (n=166); > 2-5 years (n=17); >5 years (n=24) and Auditory Neuropathy Spectrum Disorder (n=9). Speech perception results recorded at annual visits were categorized in increasing order of difficulty in categories 0 to 7 (Dowell, Blamey & Clark, 1995). Implant use, mode of communication, educational placement during primary and secondary school and higher education were documented. The employment status of recipients was categorized according to the South African Standard Classification of Occupations (SASCO). Statistical analysis was performed to identify possible prognostic variables (age at onset and duration of deafness, age at implantation, co-morbidities, socio-economic status, maternal education, home language education and speech perception category) associated with mainstream educational placement.

All the recipients who were implanted during childhood continued to use their devices. Ninety-six percent of the recipients (n=207) developed open-set speech perception and 95% (n=206) used oral language as mode of communication. In primary school, 39.4% of the recipients were placed in mainstream schools, while 91.3% were able to follow a mainstream curriculum. Forty-three percent of the recipients were in mainstream education during secondary school and 77% of the recipients were able to follow a mainstream curriculum. Fifty-four percent of the recipients who completed basic education were enrolled for or completed higher education and training. Seventy-three percent of the recipients seeking employment were employed. The average SASCO occupational skill level for these recipients was 2.4 (SD 0.96).The absence of a co-morbidity, maternal education level and speech perception category after two years of implant use, were independent variables associated with mainstream placement at primary and secondary school level.

These findings are relevant for the parents of cochlear implant candidates, the funders of cochlear implant systems and clinicians.

Opsomming

Die besluit van ouers van dowe kinders om voort te gaan met ‘n kogleêre inplanting berus dikwels op die verwagting dat die toegang tot klank wat die sisteem bied, die kind in staat sal stel om gesproke taal aan te leer, wat opvoedkundige ontwikkeling en loopbaanvooruitsige op die langtermyn sal verbeter. Hierdie positiewe uitkomste wat geassosieer word met pediatriese kogleêre inplantering, mag ook ‘n finansiële voordeel vir die gemeenskap inhou. ‘n Verskeidenheid van faktore mag egter bepaal watter uitkomste behaal word. Kogleêre inplantering is ‘n elektiewe prosedure en inligting rakende die verwagte uitkomste en prognostiese veranderlikes wat geassosieer word met uitkomste, moet beskikbaar wees vir ouers van kandidate en finasiële hulpbronne, ten einde hulle in staat te stel om ingeligte besluite te neem.

Die doel van die studie was om die demografiese en pre-operatiewe kliniese eienskappe van ‘n groep pediatriese kogleêre inplanting ontvangers in die Tygerberg Hospitaal-Stellenbosch Universiteit Kogleêre Inplanting Eenheid, die langtermyn post-operatiewe uitkomste ten opsigte van inplanting gebruik, wyse van kommunikasie, spraakpersepsie ontwikkeling, opvoedkundige plasing, beroepe, asook die veranderlikes wat geassosieer is met hoofstroom opvoedkundige plasing, te beskryf.

‘n Rekordoorsig van 216 pediatriese ontvangers van kogleêre inplantings tussen 1990 en 2014, is uitgevoer. Die ontvangers is gegroepeer volgens ouderdom van aanvang van die erg tot uitermatige sensories-neurale gehoorverlies: 0 to 2 jaar (n=166); > 2-5 jaar (n=17); >5 jaar (n=24) en Ouditiewe Neuropatie Spektrum Afwyking (n=9). Spraakpersepsie resultate, soos bepaal tydens jaarlikse opvolge is gekategoriseer volgens die moeilikheidsgraad, in kategorieë 0 tot 7 (Dowell, Blamey & Clark, 1995). Die gebruik van die inplanting, wyse van kommunikasie en opvoedkundige plasing tydens basiese en hoër onderwys is gedokumenteer. Die beroepe van die ontvangers is gekategoriseer volgens die ‘Suid Afrikaanse Klassifikasie van Beroepe’ (SASCO). Statistiese berekeninge is uitgevoer, ten einde moontlike prognostiese veranderlikes te identifiseer (ouderdom van aanvang van doofheid en duur van doofheid, ouderdom van inplantering, verwante probleme, sosio-ekonomiese status, opvoedkundige kwalifikasie van moeder, moedertaalonderrig en spraakpersepsie kategorie) wat geassosieer kan word met hoofstroom opvoedkundige plasing.

Al die ontvangers van kogleêre inplantings as kinders, was volgehoue gebruikers. Ses-en-negentig present van die ontvangers (n=207) was in staat om oopstel spraakpersepsie toetse te doen en vyf-en- negentig present was in staat om orale taal te gebruik as wyse van kommunikasie. Nege-en dertig present van die gebruikers het hoofstroom skole bygewoon in die laerskool, terwyl 91.3% toegang gehad het tot 'n hoofstroom kurrikulum. Drie-en veertig present van die ontvangers het hoostroom skole in die hoërskool bygewoon en 77% het toegang gehad tot 'n hoofstroom kurrikulim. Vier-en-vyftig present van die gebruikers wat basiese onderwys voltooi het, was besig of het reeds hoër onderrig voltooi. Drie-en- sewentig present van die gebruikers wat wou werk, het 'n werk gehad. Die gemiddelde SASCO vaardigheids-vlak van hierdie gebruikers was 2.4 (SD 0.96).The afwesigheid van verwante probleme, opvoedkundige kwalifikasie van die moeder en die spraakpersepsie kategorie, twee jaar na inplantering, was onafhanklike veranderlikes wat geassosieer is met hoofstroom plasing.

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Chapter 1. Introduction

Deafness relentlessly dismantles the “machinery of human communication” and impairs quality of life (O’Donoghue, 2013, p. 1190). It is the most common sensory impairment in high income countries (Smith, Bale & White, 2005) and it is estimated that hearing loss may be even more prevalent in low and middle income countries (Olusanya & Newton, 2007). The sequelae of hearing loss in childhood extend beyond the sensory impairment (Wilson, Tucci, Merson & O’Donoghue, 2017) and have significant medical, social and cultural ramifications (Mohr et al., 2000; Smith et al., 2005).

A severe to profound degree of sensorineural hearing loss (SNHL), in the young child is of particular concern because it will impair the development of spoken language (Niparko et al., 2010). As a consequence, these children typically develop poorer literacy skills than their normal hearing peers (Lederberg, Shick, & Spencer, 2013) and academic achievement (Qi & Mitchell, 2012) as well as employment opportunities later in life may be compromised (Mohr et al., 2000). Congenital severe to profound SNHL will result in a significant financial burden to society, mainly due to the cost of special education and reduced earning potential (Mohr et al., 2000).

The primary goal of the management of the child with a severe to profound SNHL is to develop a communication system through either manual or spoken language (Liu, 2016). The advent of cochlear implant technology has dramatically changed the communication options available to these children (Geers, Tobey, Moog, & Brenner, 2008), as the development of spoken language was challenging for many children with a congenital severe to profound SNHL prior to cochlear implants (Niparko et al., 2010). Furthermore, the majority of children with hearing loss are born to parents with normal hearing. Understandably, these parents want their children to develop spoken language and experience the same long-term educational, social and occupational opportunities that were available to them (O ’Donoghue & Pisoni, 2014). Cochlear implantation has therefore become the treatment of choice for many families who prefer an intervention approach that emphasizes the development of spoken language (Tobey et al., 2013).

The outcomes from pediatric cochlear implantation have improved considerably over the years, mainly due to early cochlear implantation as a result of newborn hearing screening programs, early intervention therapy and improvement in the technology (Birman, Elliott, & Gibson, 2012). The successful use of cochlear implant systems has changed the long-term effect of deafness for many children (Niparko & Zwolan, 2013). It has enabled some children with a congenital onset of severe to profound SNHL to develop age appropriate spoken language skills (Dettman et al., 2016) and to attend mainstream schools (Geers & Brenner, 2003). Many variables related to the child, the environment or the audiological intervention may however be associated with a child's progress after cochlear implantation and therefore not all children achieve these optimal outcomes (Boons et al., 2012; Niparko et al., 2010; Tobey at al., 2013).

Cochlear implant systems are highly effective but very expensive devices (Barton, Stacey, Fortnum, & Summerfield, 2006b). While it has been demonstrated that unilateral pediatric cochlear implantation is a cost-effective option in high income countries (Barton, Stacey, Fortnum, & Summerfield, 2006c; Cheng et al., 2000) and state funding is available for suitable candidates in some of these countries (De Raeve & Wouters, 2013; Raine, 2013), it is not easily available to patients in low and medium income countries (Magro, Emmett, & Saunders, 2018) The concern regarding the cost-effectiveness of cochlear implants in low resource settings is a significant barrier to the extension of cochlear implant services in the Global South (Emmett et al., 2015; Magro et al., 2018). Positive cost-analysis reports of pediatric cochlear implantation in the Global North conclude that the expected financial benefits associated with spoken language development, such as mainstream educational placement and the increased earning potential of the individual (Cheng et al., 2000) will contribute to the cost effectiveness. An understanding of the prognostic factors that will contribute to the outcome of pediatric cochlear implantation is therefore vital in the selection of suitable candidates (Black, Hickson, Black, & Khan, 2014) especially in low resource settings (Magro et al., 2018).

Cochlear implantation is an elective procedure. Information about the expected outcomes and prognostic variables associated with outcomes should be available to parents of candidates and funders to enable them to make informed decisions about their options (Archbold, 2002). In an attempt to better understand the long-term outcomes and prognostic factors that may improve outcomes and therefore potentially the cost-effectiveness of pediatric cochlear implantation in South Africa, the study aimed to describe: the demographic and clinical characteristics of a group of pediatric cochlear implant recipients in the Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit, the long-term trends in outcome in the domains of implant use, mode of communication, speech perception development, educational placement and occupational status achieved by this group and explore the variables associated with mainstream educational placement.

Chapter 2. Literature Review

In reviewing the relevant literature the researcher will provide an overview of the literature pertaining to the prevalence and impact of severe to profound hearing loss in children, the research evidence regarding long-term outcomes of pediatric cochlear implantation in terms of speech perception development, education and employment and the prognostic factors that may contribute to these outcome domains.

2.1. The prevalence of profound hearing loss in childhood

Results from the 2015 Global Burden of Disease Study indicate that hearing loss is the 4th leading cause of disability (Wilson et al., 2017). The World Health Organization (WHO) criteria for a disabling hearing loss in adults and children generally refers to a sensorineural cause of hearing loss (Olusanya & Newton, 2007). The WHO estimates that over 5% of the global population or 466 million persons have a disabling hearing loss (World Health Organization, 2019). Ninety-three percent (432 million persons) of this group are considered to be adults with a permanent hearing loss greater than 40dBHL in the better ear (average of hearing thresholds at 0.5 kHz, 1 kHz, 2 kHz and 4 kHz). The remaining 7% (34 million persons) are children aged up to 15 years with a permanent hearing loss in the better ear greater than 30dBHL (Olusanya & Newton, 2007, World Health Organization, 2019). As frequently cited, the majority of individuals with disabling sensorineural hearing (SNHL) loss live in lower- and middle-income countries of Central or Eastern Europe and Central Asia, Sub-Saharan Africa, Middle East and North Africa, South Asia, Asia Pacific, Latin America and Caribbean and East Asia (World Health Organization, 2019).

Although the WHO estimates indicate that the prevalence of disabling hearing loss in children is less common than adult onset hearing loss (World Health Organization, 2019), a congenital or early onset hearing loss has serious implications for the child as it interferes with communication development (Stevens et al., 2011). Data from universal newborn hearing screening programs in high income countries indicate that 2 to 4 out of 1000 babies are born with a SNHL (Smith et al., 2005). Between 20 to 30% of this group will have a profound SNHL (Kral & O'Donoghue, 2010). It is furthermore expected that this number will rise during childhood as a result of meningitis, delayed onset of genetic hearing loss or late diagnosis (Fortnum, Summerfield, Marshall, Davis, & Bamford, 2001; Kral & O'Donoghue, 2010).

Due to poorer health and socio-economic conditions, Olusanya and Newton (2007) predicted that at least 6 out of 1000 babies in low to middle income countries will have a permanent congenital or early onset hearing loss. Although exact numbers are not known it is predicted that the prevalence of congenital or acquired profound SNHL in childhood will be even greater in lower- and middle-income countries than estimated for high income countries. Possible reasons for these disparities across geographical regions are the lack of immunization, consanguinity and a greater exposure to ototoxic agents in developing regions (Kral & O'Donoghue, 2010; Smith et al., 2005).

2.2. The impact of profound hearing loss in childhood

Hearing loss results from an interference in the transmission of sound energy at any point between the outer ear and the auditory cortex (Kral & O'Donoghue, 2010). In a SNHL, a deficit in the cochlea (inner ear) will restrict sound from stimulating the auditory nerve (Gordon et al., 2011). It is estimated that 10% to 15% of patients with congenital SNHL presents with Auditory Neuropathy Spectrum Disorder (Sharma, Cardon, Henion, & Roland, 2011). As first documented by Starr, Picton, Sininger, Hood & Berlin (1996), the disordered afferent neural transmission in Auditory Neuropathy Spectrum Disorder (ANSD) results in dys-synchronous firing of the auditory nerve and neural pathways of the auditory brainstem. This lack of transmission of sensory auditory information or dys-synchronous transmission of information prevents or disrupts normal auditory development and will cause structural and functional changes to the auditory nerve, brainstem and auditory cortex during childhood (Cardon & Sharma, 2013; Gordon et al., 2011).

The auditory cortex is immature at birth and needs synchronous auditory input as well as interaction with the environment to develop the neurobiologic and neurocognitive substrates essential for speech and language development (Gordon, Tanaka, Wong, & Papsin, 2008; Kral & O'Donoghue, 2010; O'Donoghue, 2013). In the absence of sound, the development of multimodal interactions between the auditory cortex and other brain regions will be limited, while non-auditory sensory modalities may overtake areas in the auditory cortex with severe consequences to cognitive development (Gordon et al., 2011; Kral & O'Donoghue, 2010).

The severity of central nervous system consequences of hearing loss will be more pronounced in a child with a congenital bilateral severe to profound SNHL and will most likely result in the inability to develop spoken language as the most prominent consequence (Fallon, Ryugo, & Shepherd, 2014; Kral & O'Donoghue, 2010; Niparko et al., 2010). The lack of language development alongside impaired cognitive functions in this population will restrict the development of literacy skills (Lederberg et al., 2013; Traxler, 2000) and limit educational achievement and employment opportunities later in life (Mohr et al., 2000). Consequently, severe to profound hearing loss in childhood will result in a considerable financial cost for both the individual and society (Chorozoglou, Mahon, Pimperton, Worsfold, & Kennedy, 2018; Mohr et al., 2000). It is therefore important to find intervention options that are accessible and cost-effective for this population.

2.3. Cochlear implantation as an intervention option

The primary goal of intervention for the child with a severe to profound SNHL, often referred to as deaf (Smith et al., 2005), is to develop a communication system that would enable access to education and social interaction (Liu, 2016). The intervention options would include the development of either a manual communication system such as sign language or spoken language through the amplification of residual hearing with hearing aids or cochlear implants (Iseli & Buchman, 2015; Liu, 2016).

To select a mode of communication for the young deaf child, has been described as a cultural choice (Niparko, 2000). Deaf culture (with an uppercase “D”) refers to a cultural group, “united by strengths and traditions that arise from the use of sign language” (Smith et al., 2005, p. 879). From this perspective, hearing loss defines the Deaf community (Smith et al., 2005) and should not be viewed as a medical condition (Niparko, 2000) or a disability (Lane, Hoffmeister & Bahan, 1996, as cited in Balkany, Hodges & Goodman, 1998). However, as sign language is used by a minority culture it will restrict engaging with mainstream society (Niparko, 2000) and will have significant educational and social implications (Mellon, 2000).

As the majority of deaf children are born to parents with normal hearing (O’Donoghue & Pisoni, 2014), these families typically choose intervention programs that emphasize the development of spoken language (Tobey et. al., 2013). The advent of cochlear implant technology has dramatically changed the communication options for deaf children, as the development of spoken language has been challenging for many children with a congenital severe to profound SNHL, while using hearing aids (Niparko et al., 2010).

2.4. How a cochlear implant system works

A cochlear implant system is an electronic device that bypasses the deficit in the cochlea and provides direct electrical stimulation to the auditory nerve (Kral, 2013). These systems consist of a surgically placed internal receiver-stimulator connected to an electrode array that is placed in the cochlea and an externally worn unit that consist of a microphone, sound processor and transmitter coil. Sound captured by the microphone will be converted into electrical signals and coded by the sound processor. These coded signals will be sent via the external transmitter coil to an internal receiver-stimulator from where specific, tonotopically placed electrodes in the cochlea will be stimulated (Kral, Kronenberger, Pisoni, & O'Donoghue, 2016; Müller & Wagenfeld, 2003). Although the electrical hearing provided by a cochlear implant system will differ perceptually from acoustical hearing, it can provide the deaf child with sufficient stimulation necessary for central auditory development and facilitate the development of speech and language (Sharma & Campbell, 2011). Cochlear implants have therefore been described as the most successful neuroprosthetic device to date (Kral et al., 2016) and have dramatically changed the long-term impact of deafness, for children who do not benefit from hearing aids (Niparko & Zwolan, 2013).

2.5. Cochlear implant candidacy

The first multichannel cochlear implant system was approved by the USA's Food and Drug Administration (FDA) for adults in 1985 (Clark, 1997) and children older than 24 months in 1990 (Dettman et al., 2016). The positive outcome of early pediatric clinical trials led to a reduction in the minimum FDA approved age at implantation to 18 months in 1998 and to 12 months in 2000 (Dettman et al., 2016). In countries not bound by FDA regulations, like South Africa, children as young as 4 months have been implanted (Dettman et al., 2016). Initially, FDA criteria specified a bilateral profound SNHL but in 2000 candidacy criteria was expanded to include children with severe to profound SNHL if they are older than 24 months (Zwolan & Sorkin, 2016).

The decision to implant a child should always be supported by evidence of the lack of development of age appropriate speech, language and listening skills after being fitted with the appropriate amplification (Kral & O'Donoghue, 2010). The indications for pediatric cochlear implantation continued to expand over the years with more children receiving bilateral cochlear implants (Ramsden et al., 2012) and children with multiple medical conditions (Birman et al., 2012), ANSD (Cardon & Sharma, 2013; Harrison, Gordon, Papsin, Negandhi, & James, 2015) and unilateral deafness (Zeitler et al., 2019) being considered as candidates.

2.6. Cochlear implantation in South Africa

In Sub-Saharan Africa, cochlear implant units have been established in South Africa, Kenya, Namibia, Nigeria, Senegal, Tanzania and Uganda (http://www.cochlear.com). The first cochlear implant unit in South Africa was established at the Department of Otorhinolaryngology of Stellenbosch University at Tygerberg Hospital, Cape Town and the first multichannel cochlear implant surgery was performed here in 1986. At the time, the aim of the unit was to provide cochlear implantation to suitable candidates in South Africa, to develop evaluation material, train professionals and set the standard for future units in the country (Müller & Wagenfeld, 2003). After experience was gained in the implantation of postlingually deafened adults, the first child was implanted in 1988 (A.M.U. Müller, personal communication, September 20, 2016). During 1991, two more cochlear implant units were established in Pretoria and Johannesburg (Müller & Wagenfeld, 2003) and there are currently 12 units, throughout South Africa (https://www.sacig.org.za).

2.7. Cochlear implantation funding in South Africa

Cochlear implant systems are expensive electronic devices (Barton et al., 2006b) that requires long-term financial commitment for a lifetime of use (Kerr, Tuomi, & Muller, 2012). The cost of cochlear implantation therefore extends beyond the initial cost of the system, the surgery and post-operative rehabilitation and includes the long term maintenance of the internal and external components (Kerr et al., 2012; Magro et al., 2018).

The South African health care system is divided into a government-operated public health care sector and a private sector that provides care for persons covered by private medical insurance or those who can afford to pay for healthcare themselves (Scheepers, Swanepoel, & Le Roux, 2014). It is estimated that only 17% of the South African population has access to private medical insurance (Blecher, Kollipara, De Jager, & Zulu, 2011). The burden on state resources to provide health care for the majority of the population is therefore high, especially in light of the significant burden of disease from child and maternal mortality, HIV and AIDS, Tuberculosis and non-communicable diseases (Western Cape Government, Health, 2014). As in many other low and middle income countries, suitable candidates in South Africa do not have equal access to cochlear implantation as it is not funded by the National Department of Health (Kerr et al., 2012; Khan, Mukhtar, Saeed, & Ramsden, 2007). A limited number of government funded cochlear implant systems per annum are available for patients in the public sector at Tygerberg Hospital in Cape Town and the Chris Hani Baragwanath Hospital in Johannesburg. Very limited cochlear implantation services are also available at Steve Biko Academic Hospital in Pretoria, Pelonomi Academic Hospital in Bloemfontein, the Nelson Mandela Academic Hospital in Mthatha, Frere Hospital in East-London, Port-Elizabeth Provincial Hospital and Groote-Schuur Hospital in Cape Town. These hospitals have very limited access to funding and some of these centres rely on donated cochlear implant systems and pro bono surgeries performed by surgeons in the private sector.

The Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit had access to state funded cochlear implant systems since 1987. Although Tygerberg Hospital has generously increased the allocation of funds over the years, the majority of implants are still paid for by medical aid schemes or private funds, resulting in the inequity in access to cochlear implantation in South Africa.

2.8. The long-term outcomes of pediatric cochlear implantation

Summerfield and Marshall (1999, p. 141) hypothesized that cochlear implantation in early childhood will result in a "cascade of benefits" ranging from improved auditory skills to the development of spoken language, educational progress, greater employment opportunities and social independence in adulthood. The ultimate goal of pediatric cochlear implantation has been described as the development of oral language through listening, mainstream education and the prospect to have a career (Liu, 2016). Assessing the multi-faceted impact of pediatric cochlear implantation imposes significant challenges, as it will have to include multiple outcome domains (Liu, 2016) and the timeframe over which some of these outcomes are achieved may extend over many years of a person's life (Beadle et al., 2005). The outcome domains of speech perception development, educational placement and employment will be discussed.

2.8.1. Speech perception as a fundamental prerequisite for spoken language development

Since the inception of pediatric cochlear implantation, speech perception has been considered as an important consideration in the selection of candidates (Dowell, 1997) and investigated as a means to evaluate outcome (Barnard et al., 2015). Boothroyd (1997) described auditory perception as the interpretation of sensory information, within the context of the object or event that caused the sound. This process involves knowledge, memory, attention and processing skills. Auditory speech perception involves the process of interpreting auditory language patterns in order to derive meaning from words, phrases, sentences and narratives.

Children with normal hearing have the ability to discriminate speech sounds from birth (Streeter, 1976, as cited in Niparko et al., 2010) and refinement of their perceptual abilities continues during childhood (Boothroyd, 1997). The development of spoken language in the child with a severe to profound SNHL is however inhibited due to the difficulty experienced in the detection of acoustic cues essential for the identification of speech (Niparko et al., 2010; O'Donoghue, Nikolopoulos, & Archbold, 2000). As discussed in section 2.4, cochlear implant systems are designed to improve the peripheral auditory ability (Sharma & Campbell, 2011). The child with an early onset hearing loss has to use the acoustic information provided by the cochlear implant system and incorporate this improved auditory ability to develop an oral language system (Sharma & Campbell, 2011).

Documenting speech perception development during childhood is a challenge as children are constantly acquiring new auditory milestones and may therefore demonstrate a range of perceptual skills (Wang et al., 2008). In order to monitor this developmental trajectory, speech perception performance in children is measured by a variety of developmentally appropriate speech perception tests (Gordon, Daya, Harrison, & Papsin, 2000). As discussed by Barnard et al. (2015), the tests involve a continuum of tasks, ranging from the detection of speech sounds, the recognition of objects or pictures from a limited set of words (closed set speech perception) to the identification of words or sentences from an infinite number of possibilities (open set speech perception).

The ability of the congenitally deaf child to perform open set speech perception tasks, is of particular importance, as the integration of sensory, linguistic and cognitive processes involved in performing open set speech perception tasks is considered to be a fundamental pre-requisite skill necessary for spoken language development (Barnard et al., 2015; Kirk, Diefendorf, Pisoni, & Robbins, 1997). The development of open set speech perception skills of deaf children with multichannel cochlear implants has been investigated by several studies over the years.

In 1991, Staller, Beiter, Brimacombe, Mecklenburg and Arndt, reported on the collaborative effort of 30 investigational centres that participated in the early FDA trials. The inclusion criteria for these trials were conservative and candidacy was defined as a bilateral profound hearing loss and the inability to perform open set speech tasks. Participants were tested according to 4 perceptual categories viz: detection of sound, discrimination or recognition of the suprasegmental aspects of speech, closed set speech recognition and open set speech identification. The data indicated significant perceptual benefit over a broad range of abilities and 27 of the 80 (34%) children were able to perform open set speech perception tasks one year after implant use. After an analysis of the speech perception results of the first 100 children who received the Nucleus 22 multichannel cochlear implant, Dowel Blamey and Clark (1995) concluded that 60% of the children demonstrated some open set speech perception. These children were however tested at different durations of implant use, including test intervals after 2 years of use. Shortly afterwards, Waltzman et al. (1997) reported on the outcomes of 38 children with congenital bilateral profound deafness, who were consecutively implanted before the age of 5 years. These children were all able to perform open set speech perception tasks tested at intervals from 1 to 4 years.

Although outcomes varied, the data obtained from these 3 early studies demonstrated that it was possible for children with a profound deafness to achieve open-set speech perception following implantation with multi-channel cochlear implant systems. It was highlighted that a postlingual onset of deafness was associated with better outcomes (Staller et al., 1991) while longer durations of deafness were negatively associated with speech perception outcomes (Dowell et al., 1995; Staller et al., 1991).

2.8.1.1. Duration of deafness and central auditory development, speech perception development and language development

The initiation of universal newborn hearing screening or screening programs for high risk infants in some countries facilitated earlier diagnosis of infants with hearing loss and earlier implantation of children than was possible in the early cochlear implant trials (Dettman, Pinder, Briggs, Dowell, & Leigh, 2007). The advantages of earlier implantation for the child with a congenital hearing loss and the existence of sensitive periods for central auditory development have been described by several studies (Cardon & Sharma, 2013; Sharma, Dorman, Spahr, & Todd 2002a; Sharma, Dorman, & Spahr 2002b; Sharma, Gilley, Dorman, & Baldwin, 2007).

As discussed in section 2.2, the ability of the central auditory system to process auditory information does not develop in the absence of sound. It is further stated in the literature that sensitive periods of development exist in the auditory cortex, due to heightened levels of neuroplasticity during the first few years of life (Sharma & Campbell, 2011). The latency of the P1 component of cortical auditory evoked potentials (CAEPs) has been used as a measurement of the time limits for these sensitive periods (Sharma & Campbell, 2011). Due to maturation of the central auditory pathways, the latency of the P1 component, systematically decreases as children get older (Sharma & Campbell, 2011). Studies done by Sharma et al. (2002a) and Sharma et al. (2002b) examined the P1 latencies of congenitally deaf children with SNHL implanted at different ages and concluded that cochlear implantation before the age of 3 years and 6 months may result in normal P1 latencies after 6 months of implant use, while children implanted after the age of 7 showed abnormal latencies even after years of implant use. The response latencies for children implanted between 3 years 6 months and 7 years, varied. From these findings the authors concluded that the sensitive period for central auditory development for children with SNHL ends around 3.5 years (Sharma et al., 2007). In a more recent study, Cardon and Sharma (2013) documented that this sensitive period for central auditory development may even end around 2 years of age for children with ANSD.

In support of the sensitive period for central auditory development a positive association between a shorter duration of deafness and speech perception development (Gordon et al., 2000; Sarant, Blamey, Dowell, Clark, & Gibson, 2001; Barton, Stacey, Fortnum, & Summerfield 2006a; Waltzman & Roland, 2005) as well as spoken language development (Arterières, Vieu, Mondain, Uziel, & Venail, 2009; Boons et al., 2012, Ching, Dillon, Leigh, & Cupples, 2018; Niparko et al., 2010; Tobey et al., 2013) has consistently been reported in the literature. The characteristics of the participants, the stratification of the duration of deafness and test material used in these studies often varied and direct comparison of the results are therefore not possible. In a large multi-centre study, Dettmann et al. (2016) compared the open set speech perception and spoken language development of children with congenital severe to profound SNHL implanted before 12 months, between 13 and 18 months, between 19 and 24 months, between 25 and 42 months and between 43 and 72 months. The authors demonstrated that congenitally deaf children who received cochlear implants before the age of 24 months performed significantly better on open set speech perception tests than children implanted later, while children implanted before 12 months were more likely to develop age appropriate expressive and receptive language skills. These results suggest that auditory deprivation may affect more than the peripheral and central auditory system and that different sensitive periods exist for the development of speech perception and spoken language ability (Dettman et al., 2016; Houston & Miyamoto, 2010). It is therefore possible that the child with a congenital severe to profound SNHL will receive auditory benefit from a cochlear implant system when implanted after a period of deafness of more than 3 years. However, to enable optimal spoken language development, intervention should take place before the age of 12 months (Dettman et al., 2007; Dettman et al., 2016; Leigh, Dettman, Dowell, & Briggs, 2013).

In summary, it is possible that the improved peripheral hearing provided by multi-channel cochlear implant systems will enable the child with a severe to profound SNHL to develop the open-set speech perception ability fundamental for spoken language development (Niparko et al., 2010). It was discussed that different sensitive periods exist for central auditory development, speech perception and spoken language development (Dettman et al., 2016; Houston & Miyamoto, 2010). In order to facilitate optimal spoken language development in the child with congenital deafness, cochlear implantation should happen before 12 months (Dettman et al., 2007; Dettman et al., 2016; Leigh et al., 2013).

2.8.2. Educational placement

The education of deaf children has remained a controversial topic over the years (Kral & O'Donoghue, 2010). The child with a severe to profound SNHL needs to develop a communication system in order to access education (Liu, 2016). Traditionally, the two extreme views as to which communication approach would be the most effective, were those who believed that deaf children should communicate via oral language, aided by the amplification of residual hearing and those who argued that deaf children should communicate in a manual language such as sign language (Archbold, 2001).

The deaf child will only benefit from oral education if residual hearing can be optimally utilized from an early age (Yoshinago-Itano, Sedey, Coulter, & Mehl, 1998). In light of the severity of the primary sensory impairment, proficiency in spoken language, with the use of conventional amplification, has not been a realistic goal for many deaf children (O'Donoghue & Pisoni, 2014) and subsequently they developed poor linguistic and academic skills in an oral educational environment (Conrad, 1979, as cited in Archbold, 2001). These outcomes, along with increased pressure from the Deaf community, emphasized the need for the use of signed methods of communication in the education system (Archbold, 2001). Following international trends, signed methods of education were introduced in South African Schools and took the form of total communication, utilizing spoken language with signed support or a bilingual approach that incorporates both sign language and spoken language (Archbold, 2001; Parkin, 2010).

The acquisition of sign language and the subsequent development of literacy skills and educational progress may however be restricted by the following: The majority of deaf children have hearing parents (O'Donoghue & Pisoni, 2014) who are not able to provide the young deaf child with a fluent and sophisticated sign language model for optimal language acquisition in the home environment

(Mellon, 2000). As a result, the development of sign language may be compromised during the early years (Mellon, 2000; Niparko, 2000). Within the South African context it has been acknowledged that an insufficient number of teachers are fluent in South African Sign Language (SASL). The deaf child may therefore not have access to appropriate language models once they have entered the education system. This may result in substandard language development and will have a negative effect on the quality of education (Magongwa, 2010). Sign language also differs grammatically and syntactically from spoken language. The linguistic skills acquired in sign language will therefore not transfer easily to spoken language and the development of reading and writing skills. As a result, these children may develop poor literacy skills which will undermine educational progress (Mellon, 2000).

Challenges experienced by deaf learners in an oral or a sign language education system, stem from the difficulties experienced in the acquisition of language. The acquisition of both language modalities requires early language exposure. The acquisition of oral language may be limited by insufficient access to auditory information, while the acquisition of sign language may be impaired by the lack of adequate language models (Mellon, 2000; Niparko, 2000).

As discussed in section 2.4, multi-channel cochlear implant systems provide critical acoustic information that was previously unavailable to children with a severe to profound SNHL (Sharma & Campbell, 2011). This improved peripheral hearing has dramatically changed the communication ability and therefore also the education options available to deaf children (Geers et al., 2008) and made mainstream education possible (Geers & Brenner, 2003). Success in a mainstream school environment will depend on the spoken language skills and cognitive ability of the child (Kral & O'Donoghue, 2010). As documented by Niparko et al. (2010) and Boons et al. (2012) pediatric cochlear implant users may demonstrate variation in language skills post implantation and not all users will achieve optimal outcomes. Mainstream education will therefore not be the optimal placement for all learners and should not be the aim at all cost (Archbold, 2001).

The possibility of a child to attend mainstream education, may however have long term financial implications for the individual and society. Learners who do not attend mainstream education will be less likely to enroll for tertiary education, which may lead to under or unemployment (Illg, Haack, Lesinski-Schiedat, Buchner, & Lenarz, 2017). Further, it has been estimated by Mohr et al. (2000) that, on average, the cost of special education will contribute 21% of the total lifetime societal cost of severe to profound deafness in childhood in the United States. It has been documented that pediatric cochlear implantation could reduce this cost of education if mainstream education is possible (Barton et al., 2006b).

In summary, pediatric cochlear implantation has placed mainstream education within the reach of some learners with a severe to profound hearing loss (Geers & Brenner, 2003). When appropriate, mainstream education may improve the deaf child's options for tertiary education and future career and earning potential (Ilgg et al., 2017). It could also be argued that the high cost of cochlear implantation may be "partly offset" by savings in the cost of special education (Barton et al., 2006b, p. 187).

2.8.3. Employment

Mohr et al. (2002) estimated that 65% of the societal cost of severe to profound SNHL could be attributed to the limited earning potential of these individuals. Only a few studies reported on the occupational outcomes of recipients who were implanted during childhood. In a prospective longitudinal study, Beadle et al. (2005) documented that in a group of 30 consecutively implanted children at an average age of 5.2 years, only 5 of the children had already completed secondary school and higher education at the time of the investigation. Four of these children were employed. Huber, Wolfgang and Klaus (2008) reported on 15 children with an onset of deafness before 2 years and a mean age of implantation of 8.5 years. In this group of 15 children only 13 were seeking employment as 2 were enrolled for full time studies. Twelve of the 13 children were gainfully employed. The employment rates in this group compared favourably with the Austrian national average. Illg et al. (2017) investigated occupational outcomes of children implanted in Hannover, Germany between 1986 and 2000 via a questionnaire. In their cohort of 933 children, 174 questionnaires were returned. Their study group included children with a pre-, peri and postlingual onset of deafness and the mean age of implantation was 5.4 years. At the time of survey 57% of the respondents completed secondary education, vocational training or studying and 91% of them were employed. However, 49% of the participants had indicated that they have been unemployed at some point during their working life. According to the German General Social Survey for 2012, this was significantly higher than the 29% unemployment figure for the hearing population. Seventy-four participants indicated that they were able to obtain the occupation they wanted. The occupations of the participants in this study was classified according to the International Standard Classification of Occupations -88 skill level (ISCO) and an average skill level was calculated for the group. The average skill level of the cochlear implant group was significantly lower than the national average. The authors contributed this discrepancy in occupational skill level to the fact that cochlear implant recipients who had not completed secondary school education in mainstream environments were excluded from tertiary education and were therefore more limited in their career options.

In the more recent publications, the authors tend to compare the occupational outcomes of recipients with the national average (Huber et al., 2008; Illg et al., 2017). As acknowledged by Illg et al. (2017), the unemployment rate of people with hearing impairment will vary in different countries. This will be particularly relevant in a developing context like South Africa, where the general unemployment rate in 2019 was estimated to be 29 % (Statistics South Africa, 2019).

2.9. Prognostic indicators of pediatric cochlear implantation outcomes

The literature reviewed in the previous sections of this chapter indicated that the outcomes of pediatric cochlear implantation include multiple outcome domains and continue to develop over many years. Since the inception of pediatric cochlear implantation, improvement in the development of spoken language was observed as a result of improved perception of speech sounds (Boons et al., 2012). Mainstream education subsequently became an educational option for some of these children (Geers & Brenner, 2003). Great variability however exists in the outcomes achieved by pediatric cochlear implant recipients (Boons et al., 2012; Niparko et al., 2010).

It is recognized that the age of implantation in the child with congenital deafness is a critically important parameter with high prognostic value for outcome (Semenov et al., 2013). The evidence from the literature is undisputable that early cochlear implantation is a positive predictor for the development of speech perception (Sarant et al., 2001; Barton et al., 2006a; Waltzman & Roland, 2005) spoken language competency (Arterières et al., 2009; Boons et al., 2012, Ching et al., 2018 Niparko et al., 2010; Tobey et al., 2013) and mainstream educational placement (Le Roux et al., 2016; Semenov et al., 2013). In order to facilitate the possible development of age appropriate spoken language skills, cochlear implantation should happen before the age of 12 months (Dettman et al., 2007; Dettman et al., 2016).

In light of the sensitive periods for central auditory development, speech perception and language development, it is anticipated that children with an acquired SNHL who experienced a period of normal auditory development may demonstrate better outcomes with a cochlear implant than recipients with a congenital severe to profound hearing loss (Osberger, Todd, Berry, Robbins, & Miyamoto, 1991; Staller et al., 1991).

Individual differences in performance of children are however not explained by age at implantation alone. Some children implanted at young ages may not acquire age appropriate language skills, while some older-implanted children do manage to overcome language delays (Tobey et al., 2013). Additional prognostic factors that have emerged from the literature will be discussed below:

Cochlear implantation candidacy criteria have expanded to include children with severe to profound SNHL. Better pre-operative hearing levels were associated with improved oral language (Niparko et al., 2010) and speech perception development after implantation (Chiossi & Hyppolito, 2017).

A positive association between bilateral cochlear implantation and spoken language development has been established by Boons et al. (2012) and Sarant, Harris, Bennet and Bant (2014). The latter study demonstrated that earlier ages for the sequential implant was associated with the most significant improvement in oral language development.

In a study investigating the language outcomes in a group of children with different degrees of hearing loss, Moeller (2000) established that intervention after the age of two years had a less detrimental effect on outcome if high levels of parental involvement were observed. This positive relationship between parental involvement and language development after pediatric cochlear implantation was also documented by Niparko et al. (2010) and Boons et al. (2012). Maternal sensitivity to the communication needs of the child was associated with improved language (Niparko et al., 2010) and speech perception development (Barnard et al., 2015). A higher maternal education level (Ching et al., 2018) and a higher family income was associated with improved language outcomes after cochlear implantation (Niparko, 2010).

Approximately a third of pediatric cochlear implant recipients will present with a co-morbidity (Birman et al., 2012). The presence of co-morbidities has shown to negatively influence spoken language development (Birman et al., 2012; Boons et al., 2012; Ching et al., 2018) and may therefore be a barrier to mainstream educational placement (Le Roux et al., 2016).

2.10. Rationale for the study

In this chapter, it was stated that pediatric cochlear implantation will result in a 'cascade of benefits', from improved speech perception ability that will facilitate spoken language development, educational progress and improved employment prospects (Summerfield & Marshall, 1999, p.141). The outcomes from pediatric cochlear implantation have improved considerably over the years (Birman et al., 2012) and some children with a congenital onset of severe to profound SNHL will develop age appropriate spoken language skills (Dettman et al., 2016) and attend mainstream schools (Geers & Brenner, 2003).

The reduction in the disabling consequences of deafness and societal benefits of cochlear implantation in high income countries has established this technological approach as one of the most cost-effective treatments in modern medicine (Magro et al., 2018; Niparko & Zwolan, 2013). Positive cost-analysis reports of pediatric cochlear implantation in the Global North conclude that the expected financial benefits associated with spoken language development, such as mainstream educational placement and the increased earning potential of the individual, will contribute to the cost effectiveness of this expensive intervention option (Barton et al., 2006b; Cheng et al., 2000).

Although cochlear implant programs have been established in some middle-income countries, such as China, Saudi Arabia, Iran, Brazil and Egypt, the majority of cochlear implant recipients still reside in the Global North (Khan et al., 2007; Magro et al., 2018). Concerns regarding the cost effectiveness of cochlear implantation in low resource settings are one of the most significant barriers to the implementation of this service in the Global South (Magro et al., 2018).

Many prognostic variables related to a child, the environment or the audiological intervention may be associated with a child's progress after cochlear implantation and outcomes may vary quite significantly (Boons et al., 2012; Niparko et al., 2010; Tobey at al., 2013). Cochlear implantation is an elective procedure and the expected outcomes should be available to parents of the prospective recipients (Archbold, 2002; Birman et al., 2012) and funders of the systems (Archbold, 2002) to enable them make informed decisions. Further, accurate selection of cochlear implant candidates depends on a clear understanding of the prognostic factors that may contribute to the outcomes of pediatric cochlear implantation (Black et al., 2014) The selection of appropriate candidates will improve the cost-effectiveness of the intervention, which is of particular concern in low resource settings (Magro et al., 2018).

Although several international studies have reported on communication development after pediatric cochlear implantation (Liu, 2016) only a few studies have reported on long term educational placement and occupational outcomes (Beadle et al., 2005; Illg et al., 2017; Uziel et al., 2007). In a recent South African multi-centre study, Le Roux et al. (2016) identified prognostic indicators for speech perception development, speech intelligibility, mode of communication and educational placement in a group of pediatric cochlear implant recipients from 5 South African cochlear implant units in the Gauteng and Free State Provinces. Although the children in the latter study were implanted over a period of 22 years, the authors reported on cross-sectional data in the 4 outcome domains and did not provide longitudinal information. To the best of our knowledge, vocational outcomes after pediatric cochlear implantation has not previously been investigated within the South African context.

The Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit (TH-SU-CIU) pioneered cochlear implantation in South Africa and are still managing the largest adult and pediatric caseloads in the country. The unit is in a unique position to manage cochlear implant recipients from both the private and public health care sectors in South Africa. Until 2010, the TH-SU-CIU was the only unit in the Western Cape Province that provided cochlear implantation services. To date, a paucity in published data regarding the characteristics of pediatric cochlear implant recipients, the long-term outcomes attained after implantation and the prognostic indicators for outcome for recipients from the TH-SU-CIU and within the South African context at large exists.

In summary, the outcome of pediatric cochlear implantation is multi-faceted, will develop over many years and may be influenced by a multitude of factors. In a low resource environment, it is essential to document these outcomes and investigate the prognostic factors that will improve outcomes and therefore potentially the cost-effectiveness of the intervention.

With the above in mind, the following research questions guided the study:

1. What are the demographic, socio-economic and the pre-operative audiological characteristics of a cohort of children implanted at the Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit, between 1990 and 2014.

2. What are the long-term post-operative outcomes in this group of children, in terms of device use, mode of communication speech perception development, educational placement, higher education and occupational status.
3. What are the variables associated with mainstream educational placement in this cohort of children.

Chapter 3. Aims and Objectives

3.1. Main aim of the study

The main aim of this retrospective record review was threefold:

3.1.1. To describe the characteristics of the study cohort in terms of demographic, socio-economic and pre-operative audiological characteristics.

3.1.2. To describe the long-term outcomes of the study cohort after cochlear implantation in terms of implant use, mode of communication, speech perception development, educational placement, higher education and occupational status.

3.1.3. To describe the variables associated with mainstream educational placement in the study cohort.

3.2 The objectives of the study

The first aim of the study was addressed in objectives 1 to 4, the second aim in objectives 5 to 11 and the last aim in objective 12.

3.2.1. Describe the demographic characteristics of the study cohort in terms of gender, ethnicity, home language and language for education.

3.2.2. Describe the socio-economic status of the study cohort based on family income and maternal education.

3.2.3. Describe the aetiologies of hearing loss and co-morbidities in the study cohort.

3.2.4. Describe the pre-operative auditory characteristics of the study cohort in terms of the age at onset of hearing loss, degree of hearing loss, speech perception ability and duration of hearing loss prior to intervention..

3.2.5. Describe the types of implant systems used.

3.2.6. Describe implant use of the study cohort after cochlear implantation.

3.2.7. Describe the mode of communication of the study cohort after cochlear implantation.

3.2.8. Describe the longitudinal speech perception development of the study cohort after cochlear implantation.

3.2.9. Describe the long term educational placement of the study cohort after cochlear implantation.

3.2.10. Describe the higher education and training qualifications obtained by the study cohort or enrolled for after cochlear implantation.

3.2.11. Describe the occupational status of the study cohort after cochlear implantation.

3.2.12. Describe the variables associated with mainstream educational placement in the study cohort.

Chapter 4. Method

4.1. Research design

In order to address the aims and objectives of the present study, a descriptive retrospective record review of a cohort of children who received cochlear implants at a tertiary cochlear implant unit over a period of 27 years and 11 months was performed. A retrospective record review uses recorded patient information, to answer specific research questions (Kaji, Schriger, & Green, 2014; Sarkar & Seshadri, 2014; Worster & Haines, 2004) and may direct subsequent prospective studies (Wilkinson, 2016). This method is widely used by healthcare disciplines (Vassar & Holzmann, 2013) and is an emerging tool in the communication sciences (Wilkinson, 2016).The record review study design was deemed appropriate as it allowed the analysis of a large sample of observational data (Sarkar & Seshadri, 2014; Wilkinson, 2016) and the analysis of the development of patterns over a prolonged period of time (Worster & Haines, 2004).

The most significant advantage of the retrospective record review is that the data is already collected (Worster & Haines, 2004). It is therefore considered to be less resource intensive in terms of time and cost and less intrusive for patients, compared to prospective studies (Sarkar & Seshadri, 2014). However, the data in patient records was not originally recorded for research purposes. Inaccuracies in the information reported by patients or recorded by clinicians, variation in the manner clinical information was recorded by different professionals, incomplete records or misinterpretation and miscoding of data during the extraction process may therefore introduce systemic error and reduce the quality of the research (Kaji et al., 2014; Sarkar & Seshadri, 2014; Worster & Haines, 2004).

Implementing procedures to ensure the validity and reliability of data are essential in the design of the retrospective record review (Wilkinson, 2016). Internal validity is the extent to which a study measured what it aimed to do, while the external validity is the extent to which the results of a study can be generalized to other populations (Grimes & Schultz, 2002). Reliability or rigour in quantitative research refers to the consistency of the results obtained and the ability to deliver the same results, should the study be repeated (Bless, Higson-Smith, & Sithole, 2013).

Based on earlier publications (e.g. Gearing, Mian, Barber and Ickowics (2006), Gilbert, Lowenstein, Koziol-McLain, Barta and Steiner (1996), and Worster & Haines (2004)), Vassar and Holzmann (2013) proposed 10 considerations to ensure methodological rigor in the retrospective record review. These considerations, as applied to this study, are summarized in the list below:

1. Create well defined research questions
2. Consider the sample size and sampling method
3. Develop explicit inclusion and exclusion criteria
4. Operationalize the variables included in the record review
5. Develop and use standardized data abstraction forms
6. Conduct a pilot study
7. Create a data abstraction procedure manual
8. Train and monitor data capturers
9. Address inter-rater and intra-rater reliability
10. Address confidentiality and ethical considerations

These considerations will be addressed in more detail in the description of the method of the study. The methodological strengths and weaknesses of the study will be summarized in the final section of this chapter.

4.2. Research site

The study took place at a single tertiary cochlear implant unit, the Tygerberg Hospital-Stellenbosch University Cochlear Implant Unit (TH-SU-CIU) in Cape Town, South Africa.

4.3. Selection of the study records

Gearing et al. (2006), Vassar and Holzmann (2013) and Worster and Haines (2004) highlight the importance of a sample size and a sampling method that would include a study sample that is representative of the population from which the sample of records was drawn. A study sample that is not representative of the broader population will limit the generalizability of the results which will be a threat to the external validity of the study. Incomplete records and records that include patients with confounding co-morbidities would however degrade the internal validity of the study (Vassar & Holzmann, 2013). The sampling of records and inclusion and exclusion criteria of the records selected for review, were therefore defined as follows.

4.3.1. Inclusion criteria

The study sample included all children who received cochlear implants from the TH-SU-CIU before they were 18 years old. Data recorded from 1990 to 2018 were reviewed for recipients who underwent implantation from 1990 to 2014. All recipients therefore had a minimum duration of implant use of 3 years at the time of data collection.

4.3.2. Exclusion criteria

To ensure that comprehensive longitudinal records were available for all the recipients in the study sample, children implanted at the TH-SU-CIU, but transferred to other cochlear implant units were excluded from the review. Severe intellectual impairment and severe autism were identified as confounding co-morbidities as children diagnosed with these conditions could not reliably perform post-operative speech perception tests and were therefore excluded from this review. Deceased users were excluded as well.

4.3.3. Description of the recipients

From September 1988 to December 2014, 320 children under 18 years of age received cochlear implants from the TH-SU-CIU. In December 2014, 224 of the 320 children (70%) were still managed by the above unit, 93 children (29%) were referred to other cochlear implant units and 3 children (1%) were deceased. Eight of the remaining 224 children were excluded from the review due to a diagnosis of severe autism or confirmed severe or profound intellectual impairments as a result of microcephaly, Towes Brock Syndrome or a Cytomegalovirus infection. A total of 216 children met the criteria for the present study. This form of convenience sampling was considered to be appropriate, as it included all implanted recipients over a significant period of time who met the inclusion and exclusion criteria.

The first child who met the selection criteria, was implanted in February 1990. The 216 suitable recipients who received cochlear implants between February 1990 and December 2014 (24 years and 10 months) were included in the study. Data recorded in patient records from February 1990 to January 2018 (27 years and 11 months) was reviewed.

4.4. Data abstraction instrument

Gearing et al. (2006), Gilbert et al. (1996), Vassar & Holzmann (2013) and Worster & Haines, (2004) emphasize the importance of the use of a standardized data abstraction instrument and conducting a pilot study. The internal validity of the study will be compromised if data is not captured in an accurate or consistent manner. In order to maintain consistency in the data abstraction for the 216 recipients and reduce error, a secure Excel database was developed in consultation with a statistician. The order of the data fields in this database was simplified after the first 10 records were reviewed.

4.5. Data abstraction

Data were collected retrospectively for the 216 recipients included in the study, from their case files, stored in the TH-SU-CIU and an existing electronic patient database. Data recorded in patient records from February 1990 to January 2018 (27 years and 11 months) were reviewed.

4.6. Data fields

Gilbert et al. (1996) and Vassar and Holzmann (2013) highlight the importance of clearly defined research questions and operational variables. The research questions formulated in chapter 2 were central to the formulation of the 12 objectives of the study. In order to address the objectives of the study, specific variables were captured from the records of the recipients. These variables were organized in data fields in the data abstraction instrument and will be discussed according to the 12 objectives of the study:

4.6.1. Demographical information

Objective 1	**Describe the demographic characteristics of the study cohort in terms of gender, ethnicity, home language and school language.**

4.6.1.1. Gender

The gender of the recipients was captured as categorical variables, male or female.

4.6.1.2. Ethnicity

Ethnicity was captured according to the 4 categories used in South Africa for official purposes (Störbeck & Young, 2016) as Black, Coloured, Indian or White.

4.6.1.3. Home language

The home language of families was captured as Afrikaans, English, isiXhosa, Sign Language or Other according to a numerical code. If more than one language was used by family members living with the recipient, all languages used were documented.

4.6.1.4. Language for education

The language in which a recipient received education was captured according to the same options for home language.

4.6.2. Socio-economic status

Objective 2	**Describe the socio-economic status of the study cohort based on family income and maternal education.**

4.6.2.1. Family income

The income of the recipients was captured according to the Tygerberg-Hospital Income Classification System (Appendix A). According to this system, the annual family income was classified into 1 of 5 categories, H0, H1, H2, H3 or P, ranging from those with no income (H0) to those classified as private patients (P). Parents of recipients may become unemployed or change jobs or recipients themselves may complete education and find their own employment. The captured income level therefore only reflected cross sectional data at the time of data capturing and served as a proxy for the income level of the recipients.

4.6.2.2. Maternal education level

The highest educational level achieved by the mothers of recipients was recorded from the case files, as it was documented on their first visit to the TH-SU-CIU. The education level captured was categorized according to a numerical value from 2 to 7. The mean of these values resembled the mean maternal education level. The categories used to classify maternal education level and the associated values are summarized in Table 1.

Table 1: Summary of the maternal education level categories

Associated value	**Description of category**
0	Data not in file
2	No formal education
3	Primary school education
4	Secondary school education: grade 8 to grade 11
5	Secondary school grade 12
6	Qualification after grade 12 (non-degree)
7	Graduate or post graduate qualification

4.6.3. Aetiologies and co-morbidities of hearing loss

Objective 3	**Describe the aetiologies of hearing loss and co-morbidities in the study cohort**

4.6.3.1. Aetiologies of hearing loss

The aetiologies of hearing loss were categorized as unknown, genetic (non-syndromic), syndromic, viral or bacterial, trauma and hypoxia, premature birth, ototoxic drugs, other and Auditory Neuropathy Spectrum Disorder (ANSD). Although ANSD is associated with numerous causes (Harrison et al., 2015), for the purpose of this study, this group was categorized separately. These categories are summarized in Table 2.

Table 2: The classification of the aetiologies of hearing loss

Aetiology category	Aetiology
Unknown	Congenital unknown
	Progressive unknown
	Unknown
Genetic (non-syndromic)	Consanguinity
	Connexin 26
	Dilated vestibular aqueducts
	Genetic progressive
	Genetic unknown
Syndromes	Usher syndrome
	Waardenburg Syndrome
	De Toni Fanconi Syndrome
	Noonan Syndrome
	Pendred Syndrome
Viral or bacterial	Congenital rubella
	Cytomegalo Virus (CMV)
	Mumps
	Meningitis
	Pyrexial illness
	Viral unknown
Trauma or hypoxia	Birth injury
	Head injury
	Hypoxia
Premature birth	Premature birth
Other	Hypoplastic nerve
	Rhesus incompatibility
Auditory Neuropathy Spectrum Disorder (ANSD)	ANSD

4.6.3.2. Co-morbidities

Based on available reports in the case files from neurologists, ophthalmologists, pediatricians, psychologists, occupational therapists and speech and language therapists, co-morbidities were identified and classified as below:

- Cognitive Impairment
- Learning problem or language learning problem
- Attention deficit disorder (ADD)
- Cerebral Palsy (CP)
- Blind or visual impairment
- Dyspraxia
- Autism
- Developmental delay
- Emotional and behavioural problems
- Epilepsy

4.6.4. Pre-operative audiological characteristics of the recipients

Objective 4	**Describe the pre-operative auditory profile of the study cohort in terms of age at onset of hearing loss, degree of hearing loss, duration of hearing loss prior to intervention and speech perception ability.**

4.6.4.1. Study groups

a. Sensorineural hearing loss:

Recipients with SNHL were grouped according to the age at onset of the severe to profound hearing loss. It is anticipated that children who have experienced a period of normal development of the central auditory system may have better outcomes with a cochlear implant than recipients who were born with a severe to profound hearing loss (Osberger et al., 1991).

In light of the sensitive periods for language acquisition, the onset of hearing loss is often described as prelingual, perilingual and postlingual (Osberger et al., 1991; Staller et al., 1991). A prelingual hearing loss implies that the loss occured before the acquisition of spoken language, while a perilingual hearing loss, occurs after some spoken language has been developed, but before the language system was fully established. A postlingual hearing loss occurs after the acquisition of spoken language (Tye-Murray, 2009).

There are no universally accepted cut-off times to define the prelingual, periligual and postlingual periods. In the present study recipients with SNHL were grouped according to a prelingual, perilingual or postlingual onset of severe to profound SNHL, as defined by Tye-Murray (2009):

- Group 1: Prelingual onset of severe to profound SNHL: 0 to 23 months
- Group 2: Periligual onset of severe to profound SNHL: 24 to 59 months
- Group 3: Postlingual onset of severe to profound SNHL: >= 60 months

b. Auditory Neuropathy Spectrum Disorder (ANSD)

Recipients diagnosed with ANSD were placed in the 4th study group.

4.6.4.2. Pre-operative residual hearing

The pre-operative unaided pure tone air conduction thresholds at 500Hz, 1000Hz, 2000Hz and 4000Hz measured by means of behavioral audiometry via headphones or insert earphones were obtained from case files. Ear specific results were entered into the Excel database. If a recipient had no response at a tested frequency, the level was recorded as the maximum output of the audiometer in dBHL +1. The pre-operative hearing levels were only accepted for recipients with comprehensive 4-frequency thresholds available for both ears. The mean of the 4-frequency pure tone average (500, 1000, 2000 and 4000Hz) for the best ear and worst ear were calculated for the 4 study groups. The 4-frequency average for the best ear served as a proxy for residual hearing prior to cochlear implantation (Niparko et al., 2010).

4.6.4.3. Pre-operative speech perception ability

a. Speech Perception Categories

As discussed in section 2.8.1, the variety of speech perception test materials that may be used in a group of pediatric cochlear implant recipients over a period of time impose a challenge in reporting longitudinal outcome in speech perception development. In the literature, this challenge is often addressed by implementing a scale that organizes speech perception abilities into a limited number of categories (Wang et al., 2008). The speech perception ability of a child was therefore described as a category number as opposed to a score achieved on a specific test. This approach allows comparison of different tests used.

The CID Early Speech Perception test, described by Moog and Geers (1990), organizes speech perception development according to a hierarchical scale, ranging from categories 0 to 4. A category score of 0 indicated that the child was not able to detect speech at a level of 65dBHL, while children who were able to detect speech at 65dBHL, were assigned a category score of 1 and could proceed with further speech perception testing (Geers, 1994). The criteria for the remaining categories 2 to 4 is based on the performance of the 3 subtests of the standard and low verbal version of The CID Early Speech Perception Test (Moog & Geers, 1990). Categories 2 to 4 are hierarchically organized and monitored the child's ability to discriminate words in a closed set based on temporal and stress cues as well as phonemic identification. Dowell et al. (1995) have added open set categories 5 to 7 to the scale suggested by Moog and Geers (1990) and graded the child's ability to perform open set word identification according to the score achieved on the Phonetically Balanced Kindergarten test (Haskins, 1949).

The Categories of Auditory Performance (CAP), described by Archbold, Lutman and Marshall (1995) is another example of a categorical classification scale that is frequently used in the literature. The CAP consists of 8 categories that classifies functional listening from no awareness of sound to the ability to have a telephone conversation with a familiar speaker. These categories are assigned by the professional responsible for the rehabilitation of the child (Archbold et al., 1995).

Due to the retrospective nature of the present study, the categorical scales suggested by Moog and Geers (1990) and Dowell et al. (1995) were considered to be more suitable as it provided clear guidelines to categorize retrospective results obtained by speech perception tests. A hierarchical scale version of their scales, already used by the TH-SU-CIU, was used to categorize the speech perception performance of the recipients and is summarized in Table 3.

Table 3: Speech perception categories	
Speech perception category	**Description of category**
0	Not able to detect speech at 65dBHL
1	Able to detect consonants and vowels at 65dBHL
2	Able to discriminate between words in a closed set based on suprasegmental information of duration and stress
3	Start to discriminate between words in a closed set based on spectral information
4	Able to recognize words in a closed set based on vowel recognition (80 to 100% correct)
5	Identify words or sentences in an open set: 10 to 20% correct
6	Identify words or sentences in an open set: 21 to 50% correct
7	Identify words or sentences in an open set: 51 to 100% correct

b. Pre-operative speech perception categories

As per departmental protocol, the pre-operative speech perception evaluation was performed by a battery of age and developmentally appropriate speech perception tests in English, Afrikaans and isiXhosa. All tests were performed in the auditory only condition (without visual cues). Depending on the test, speech material was presented via live voice or recorded speech at 40dBHL. At the time of data collection, the results of the pre-operative speech perception test were categorized according to the 8 categories outlined in Table 3.

4.6.4.4. The age at intervention and duration of deafness prior to intervention

The ear specific age at onset of the severe to profound SNHL, age at hearing aid fitting and age at cochlear implantation were recorded from the case files.

a. The age at hearing aid fitting and duration of deafness prior to hearing aid fitting

The age at onset of the severe to profound SNHL and the age at hearing aid fitting were recorded as continuous variables. The duration between the onset of the hearing loss and hearing aid fitting was calculated for the ear that was implanted first.

b. Age at cochlear implantation and duration of deafness prior to implantation

The age at implantation was recorded as a continuous variable. The duration of severe to profound SNHL prior to cochlear implantation was calculated for the ear that was implanted first or if a recipient received bilateral implants simultaneously, the right ear.

4.6.5. Cochlear implant systems

Objective 5	**Describe the types of implant systems that were used.**

All the recipients received cochlear implant systems manufactured by Cochlear Limited® from 1990 to 2014 (Cochlear ®, 2017). The implant systems that were commercially available during this period, are summarized in Appendix B. The operation date, type of implant system used and ear (left or right) were recorded for the first and in the case of bilateral users for the sequential ear as well.

4.6.6. Device use after cochlear implantation

Objective 6	**Describe device use and non-use after cochlear implantation.**

Device use was categorized as use or non- use, at the time of data capturing.

4.6.7. Mode of communication after cochlear implantation

Objective 7	**Describe the mode of communication of the study cohort after cochlear implantation.**

Data about the mode of communication used by the recipients after cochlear implantation was obtained from case files or in consultation with a recipient's managing audiologist. The mode of communication was categorized as oral, when recipients only used spoken language, as total communication, when recipients used spoken language with signed support (Archbold, 2001) or thirdly as sign language.

4.6.8. Speech perception development after cochlear implantation

Objective 8	**Describe the longitudinal speech perception development of the study cohort after cochlear implantation.**

The speech perception results recorded at annual visits were reviewed retrospectively from case files. As discussed in paragraph 4.6.4.3, the results of a range of speech perception tests were categorized according to a scale of 8 speech perception categories by the researcher (Dowell et al., 1995; Moog & Geers 1990).

4.6.9. Educational placement after cochlear implantation

Objective 9	**Describe the long-term educational placement of the study cohort after cochlear implantation**

Education in South Africa is governed by the Department of Basic Education (DBE) and the Department of Higher Education and Training (DHET). The DBE is responsible for the management of primary schools (grade R to 7) and secondary schools (grade 8 to 12). Universities and other training facilities are managed by the DHET (Department of Education, 2019).

For the purpose of the present study, the longitudinal progress of recipients during the basic education phase was recorded for the following 4 stages:

- Preschool (0 to 4 years)
- Grade R
- Primary School (grade 1 to 7)
- Secondary School (grade 8 to 12)

Retrospective information was documented for every recipient. It was indicated if a recipient was enrolled for or had completed a particular stage. In this section, the type of school attended was also recorded. It was documented if a recipient had attended a mainstream school or a school for specialized education. The recipients in the present study had attended a variety of special school settings. The special schools for hearing impaired learners followed a mainstream curriculum during the primary school phase, but at secondary school level some schools offered recipients the option to follow a special skills or trade curriculum. In the Western Cape Province there is only one English medium oral school, Dominican Grimley School for the Deaf, for hearing impaired learners that offers a mainstream academic curriculum until grade 12. Non-English speaking recipients or recipients who could not access this school were therefore placed in total communication or sign schools, for example De La Bat School or in schools for learners with learning problems, for example Jan Kriel School. The types of special education schools that recipients attended were therefore documented as:

- Oral school for hearing impaired learners: mainstream curriculum
- Total communication or sign language school for hearing impaired learners: mainstream curriculum
- Total communication or sign language school for hearing impaired learners: special skills or trade curriculum
- Special school for learners with learning problems: mainstream curriculum
- Special school for learners with learning problems: special curriculum

4.6.10. Higher education and training

Objective 10	**Describe the higher education and training qualifications obtained by the study cohort or enrolled for after cochlear implantation.**

The National Qualifications Framework (NQF) is a set of principals by which skills and knowledge obtained from grade 9 (secondary school) and onwards are recognized according to a hierarchical scale of 10 levels. These 10 levels are divided into 3 bands; levels 1 to 4 equate to high school grades 9 to 12 or vocational training, levels 5 to 7 are college diplomas and technical qualifications, while university degrees fall in categories 7 to 10 (South African Qualifications Authority, 2019). For the purpose of the present study, the qualifications obtained or enrolled for by recipients who had left secondary school at the time of data capturing were categorized according to the NQF framework. These categories are summarized in Table 4.

Table 4. Summary of the National Qualifications Framework (NQF) levels

Qualification	**NQF level**
Apprenticeship, Learnership or Skills Programs Include National and Occupational Certificates	1 to 4
Higher Certificate, Advanced Certificate, Occupational Certificates, Diploma	5 to 6
Bachelors Degree	7
Honours or Masters Degree	8 to 9
Doctoral Degree	10

4.6.11. Occupational status

Objective 11	**Describe the occupational status of the study cohort after cochlear implantation**

The occupational status and occupations of the recipients who had completed basic education and higher education and training were collected from case files or in consultation with the managing audiologist.

4.6.11.1. Employment status

Recipients were categorized as employed or not employed.

4.6.11.2. Employment categories

The occupations of the recipients were categorized according to the South African Standard Classification of Occupations (SASCO). SASCO was developed according to the same conceptual basis as the United Nation's International Standard Classification of Occupations (ISCO-08) and the purpose is to provide a national framework for occupations that would enable comparability on an international level. SASCO classifies occupations in 9 major groups and assigns a skill level to each of these major groups. The skill level is defined based on the characteristics of the tasks to be performed in an occupation and/or the level of formal or informal training necessary to complete the tasks (Statistics South Africa, 2012). The 9 major groups and the 4 associated skill levels are summarized in Appendix C.

4.6.12. Describe the variables associated with mainstream education

Objective 12	**Describe the variables associated with mainstream educational placement in the study cohort.**

The investigation of the variables associated with mainstream educational placement at primary and secondary school level was conducted in 2 phases. This will be described in section 4.9.

4.7. Data abstraction process

Data was captured over a period of 8 months from March to December 2014. During 2018, the post-operative speech perception results recorded between December 2014 and January 2018 were captured for recipients implanted during 2012, 2013 and 2014, to ensure that that the longitudinal speech perception category scores reflected a minimum period of 3 years for all the recipients. The educational placement, higher education qualifications and occupational categories were recorded for all the recipients during 2018.

All the data capturing was done by the investigator, who had been employed as a clinical audiologist in the unit for more than 7 years. The considerations regarding the training of data capturers and creating a procedure manual for data capturers to improve inter-rater reliability were therefore not relevant for the present study (Gilbert, 1996; Vassar & Holzmann 2013).

4.8. Ethical considerations

Ethical approval was obtained from the Human Research Ethics Committee of the University of Stellenbosch to conduct the study. (Appendices D & E). The data was collected and entered into the Excel database by the researcher. The privacy of the recipients was protected and confidentiality maintained as all data were de-identified by using a specific study number instead of names and surnames (Vassar & Holzmann, 2013). The digital data containing information about the study will be securely stored for two years after publication of the results and then be destroyed. A waiver of informed consent was granted by the Ethics review board as the study only involved a retrospective review of clinical records, see Appendix D.

4.9. Statistical analysis

Data collected during the retrospective record review was organized according to the objectives of the study and analyzed in a quantitative manner. Statistica 13, a statistical software package, was used by a statistician to analyze the quantitative data to establish the significance of relationships between variables.

Descriptive statistics were used to describe the demographic and clinical profile of the recipients. Means and standard deviations were reported for continuous variables and numbers and percentages for categorical variables. Where appropriate, these values were displayed in tables or figures.

As discussed in section 4.6.4.1 the recipients were grouped into 4 study groups. The mean pre-operative degree of hearing loss, duration of deafness prior to intervention and pre-operative speech perception category for the 4 groups were calculated. In situations where the F-ratio suggested rejection of the null hypothesis, the Least Significant Difference test (LSD) was used to identify the study groups whose means where statistically different.

As discussed in section 4.6.4.3 the post-operative speech perception ability of the recipients was categorized in one of 8 categories (0-7). The means of these categories, as documented per annual test interval, were calculated for a 10-year period after implantation and for each of the 4 study groups over a 3 year period. The LSD test was used to identify statistically significant mean scores per test interval over the 10-year period and to identify inter-group and intra-group differences over the 3 year period.

The investigation of the variables associated with mainstream placement at primary and secondary school placement, was conducted in 2 phases. First, in a univariate analysis of variables, the differences between the mainstream and special education groups were examined for selected variables at baseline. The chi-square test was used to examine group differences by categorical variables. For continuous variables, one-way analysis of variance (ANOVA) was used to examine for group difference. In the second phase, logistic regressions were conducted in an attempt to identify the variables that reliably differed for the mainstream or special education groups, independent of other variables. Significance for all statistical tests was set as $p < 0.05$.

4.10. Validity and reliability

The strengths and weakness of the present study will be analyzed in the following section in terms of the data source, the study sample and the data abstraction process.

4.10.1. Data source

As discussed in section 4.1, the data in the records of recipients was not originally recorded for research purposes. Error in the information reported by the recipients or recorded by the clinicians and variation in the manner clinical information was recorded by different professionals, may therefore exist. These inaccuracies or variations in the data could result in systemic bias in the study (Kaji et al., 2014). This is an inherent limitation of the retrospective record review and it is not possible to report on the reliability of the original data sources (Worster & Haines, 2004). However, the content of the records selected for review in the present study, were administered by only 4 audiologists employed in the TH-SU-CIU between 1988 and 2018. The limited number of clinicians involved in recordkeeping could have reduced the variation and potential error in the original data sources and may therefore have protected the reliability and validity of the study.

4.10.2. Study sample

As discussed in section 4.3.3, 320 children received cochlear implants at the TH-SU-CIU from 1988 to 2014. In order to protect the external validity of the study, the records of the 216 (68%) children who met the inclusion criteria were included in the study sample. The 8 children identified with confounding co-morbidities were considered to be a threat to the internal validity of the study and were therefore excluded from the study sample. Children transferred to other units or deceased during 1988 to 2014, potentially had incomplete longitudinal records and were therefore excluded from the study sample as well. However, if the characteristics of the 30% of the children with incomplete records were significantly different than those of the study sample, excluding them could have introduced a form of selection bias in itself, which is a limitation of the study that was not addressed.

4.10.3. Data abstraction process

Misinterpretation and miscoding of data during the abstraction process can result in error and misclassification bias (Kaji et al., 2014). However, as discussed in section 4.6, all variables were clearly defined and limited to categorical and continuous variables. These variables were entered into an electronic database. The selection of categorical variables in the database was simplified by the use of a coded response or a limited option response. The data abstracted from patient records was typically found in case history forms, assessment reports and pre- and post-operative audiograms. This limited misinterpretation of data in the free text format typically used in medical records (Worster & Haines, 2004). The reliability and validity of the results was therefore protected by the use of clearly defined variables that were easy to find in the records and a standardized data abstraction instrument.

In the present study, the data abstraction was done by the investigator. The fact that the data abstraction was done by a single data abstractor who was not blind to the aims of the study, was a potential source of bias and intra-rater reliability needs to be considered. However, as discussed previously, all variables were clearly defined and limited to categorical and continuous variables. Due to the nature of the data that was captured, potential abstractor bias and intra-rater variability was therefore not considered to be a serious threat to the validity and reliability of the study (Worster & Haines, 2004). To confirm accuracy, all data entries were double-checked by the investigator.

Chapter 5. Results and Discussion

The results of the retrospective record review will be discussed according to the 12 objectives of the study. In sections 5.1 to 5.4, the study cohort will be described in terms of demographic, socio-economic and pre-operative audiological characteristics. The long-term outcomes after cochlear implantation, in terms of implant use, mode of communication, speech perception development, educational placement, higher education and occupational status will be discussed in sections 5.5 to 5.11. The variables associated with mainstream educational placement will be discussed in section 5.12.

5.1. Demographical characteristics of the recipients

The gender, ethnicity, home language and language of education of the recipients are summarized in Table 5.

Table 5: Demographical characteristics of the study cohort (N=216)

Characteristic	*n*	%
Gender		
Female	114	53
Male	102	47
Ethnic Category		
White	100	46.3
Coloured	94	43.5
Black	21	9.7
Indian	1	0.5
Home Language		
Afrikaans	99	46
English	86	40
isiXhosa	17	8
Other	5	2
Bilingual	9	4
Language of Education		
Received Education in Home Language	196	91
Did not receive Education in Home Language	20	9

According to the National Census 2011, the ethnic distribution for the Western Cape Province was 15.7% White, 48.8% Coloured, 32.8% Black and Indian 1% (Statistics South Africa, 2011). It is therefore clear that the Black population in the Western Cape is currently underserved compared to the other ethnic groups. A similar trend was reported in a recent South African multi-centre study by Le Roux, Swanepoel, Louw, Vinck, and Tshifularo (2015). In a retrospective record review of 4 cochlear implant units situated in the Gauteng Province and 1 in the Free State Province, they reported the ethnic distribution of pediatric cochlear implant recipients implanted between 1996 and 2013 as 66.2% White, 20.5% Coloured, Black, 7.6% and Indian 5.7%.

As could have been predicted from the unrepresentative ethnic distribution of the study participants, the 8% isiXhosa speaking families in the study was proportionally lower than the 24.7% documented for the Western Cape Province (Statistics SA, 2011). Eighteen of the 20 participants who did not receive home language education were from isiXhosa speaking families. These 18 children were educated in English, as there is currently no isiXhosa-medium, oral school for hearing impaired learners in the Western Cape Province.

5.2. Socio-economic characteristics of the recipients

5.2.1. Family income

As discussed in section 4.6.2.1, the income of the recipients was categorized according to Tygerberg Hospital's Income Classification system (Appendix A) and is summarized in Figure 1.

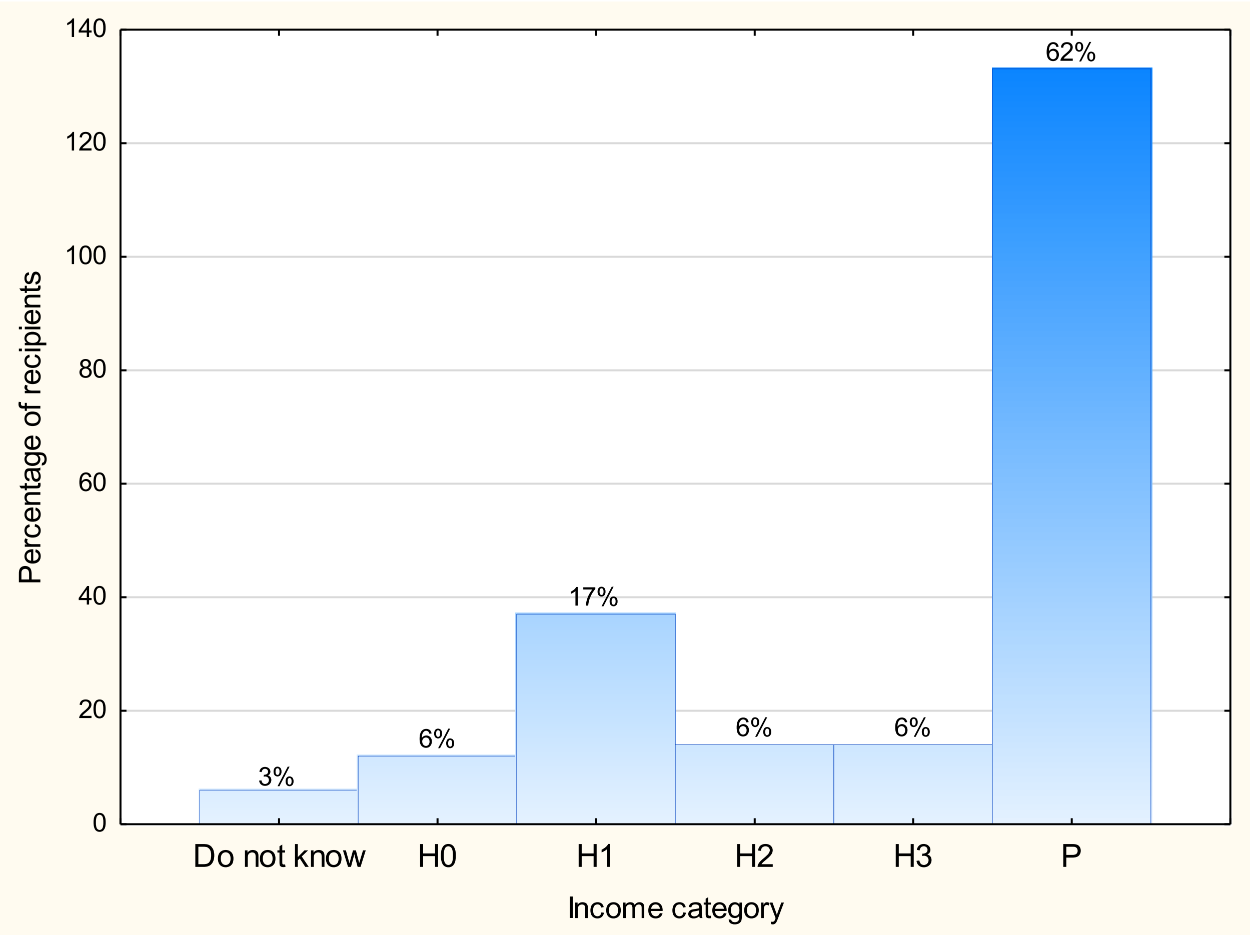

Figure 1. Income categories of recipients (N=216)

As seen in Figure 1, the majority of the recipients belonged to a medical aid scheme. This is significantly higher than the estimated 17% of South African families who had access to private health insurance in 2011 (Blecher et al., 2011).

5.2.2. Maternal education level

The highest education level achieved by the mothers of the recipients is summarized in Figure 2.

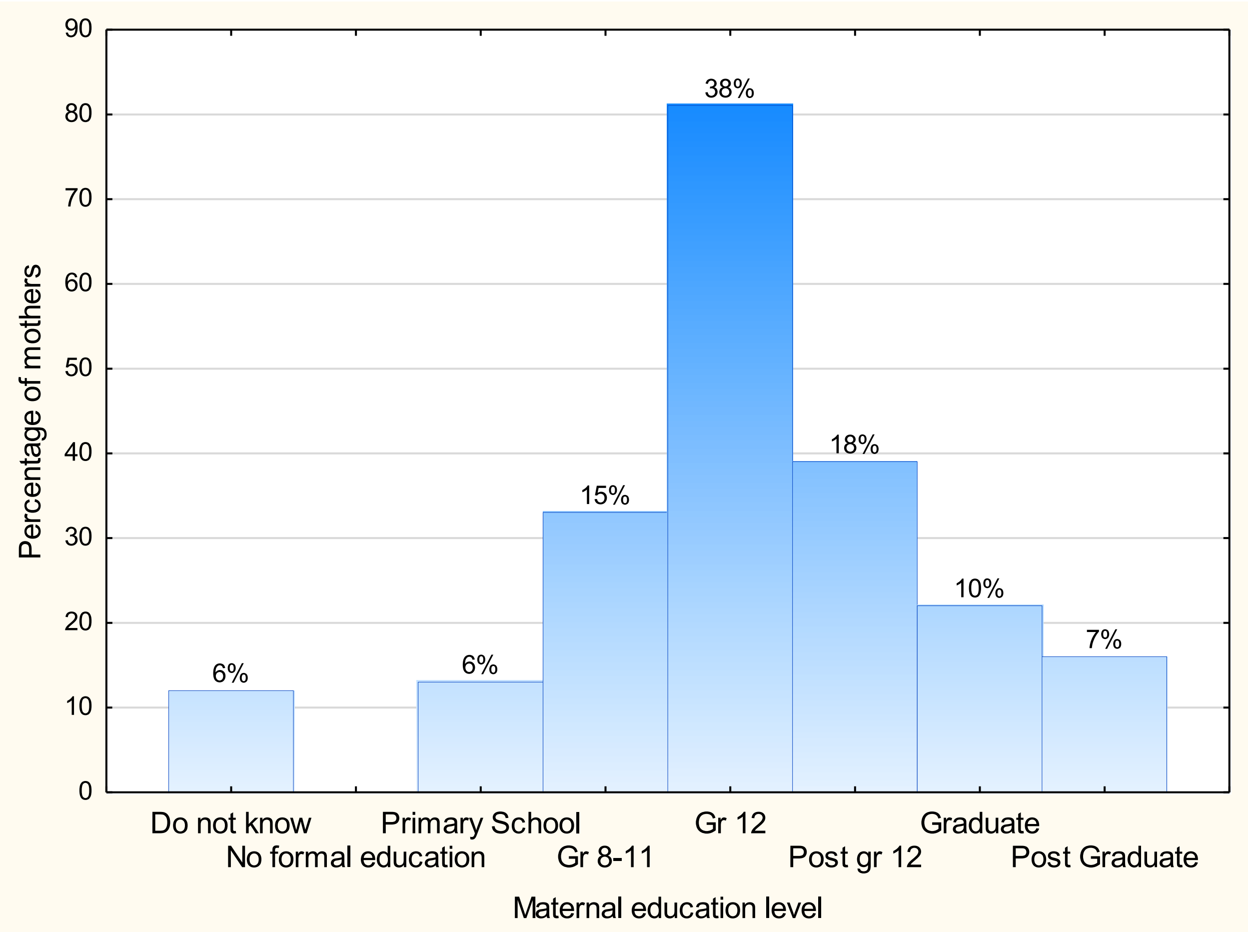

Figure 2. Highest maternal education level (N=216)

The majority of mothers achieved a minimum education level of grade 12. According to the national Census 2011, 42.2% of persons older than 20 years in the Western Cape Province, obtained a minimum qualification level of grade 12 (Statistics South Africa, 2011). Although this percentage does not specifically refer to females, the average maternal education level of the mothers in the study population was noticeably higher than that of a similar age group in the province.

Summary and discussion

The demographic and socio-economic characteristics of the study cohort was reported in sections 5.1 and 5.2. In summary, the ethnic distribution of the recipients in this study cohort was not representative of the ethnic distribution in the Western Cape Province. The black population was the most under represented ethnic group. In terms of family income and maternal education level, the socio-economic status of the majority of the recipients was higher than what is documented to be the provincial or national average (Blecher et al., 2011; Statistics South Africa, 2011).

As discussed in section 2.7, the South African health care system is divided into a government-operated public health care sector and a private sector that provides care for persons covered by private medical insurance or those who pay for healthcare themselves (Scheepers et al., 2014). As mentioned, it is estimated that more than 80% of the South African population depends on health care from the public sector (Blecher et al., 2011). The TH-SU-CIU is based at a tertiary hospital and serve patients from both the private and public health care sectors, but government funding is available for only a limited number of public sector patients per year. Although an increase in the number of recipients who received cochlear implants annually through public funding was observed since 1986, the majority of implants are still funded via medical aid schemes or private funding and are therefore more accessible to families with access to these resources.

The disparities in cochlear implant provision amongst ethnic and socio-economic groups documented in the study cohort, was observed in the Gauteng and Free State provinces as well (Le Roux et al., 2015) and can possibly be attributed to a lack of funding for cochlear implants by the National Department of Health (Kerr et al.,2012). As the majority of the South African population depends on the public sector for health care (Blecher et al., 2011) the inequity in cochlear implant provision observed in the study cohort, is likely to exist in the larger South African context.

5.3. Aetiologies and co-morbidities of hearing loss

5.3.1. Aetiologies of hearing loss

The aetiologies of hearing loss for the recipients are summarized in Table 6.

Table 6: Aetiologies of hearing loss (N=216)

Aetiology	***n***	**%**
Unknown	**84**	**38**
Congenital unknown	73	
Progressive unknown	10	
Unknown	1	
Genetic (non-syndromic)	**43**	**20**
Consanguinity	1	
Connexin 26	19	
Dilated vestibular aqueducts	2	
Genetic progressive	2	
Genetic unknown (congenital hearing loss)	19	
Syndromes	**10**	**5**
Usher syndrome	4	
Waardenburg syndrome	2	
De Toni Fanconi syndrome	2	
Noonan syndrome	1	
Pendred syndrome	1	
Viral or bacterial	**51**	**24**
Congenital rubella	12	
Cytomegalo virus (CMV)	5	
Meningitis	24	

Mumps	6	
Pyrexial illness	2	
Viral unknown	2	
Trauma or hypoxia	**6**	**3**
Birth injury	1	
Head injury	1	
Hypoxia	4	
Ototoxic drugs	**2**	**1**
Premature birth	**9**	**4**
Other	**2**	
Hypoplastic auditory nerve	1	
Rhesus incompatibility	1	
Auditory Neuropathy Spectrum Disorder (ANSD)	**9**	**4**

In 38% of the study cohort the aetiology of the hearing loss was unknown. An additional 21 recipients (10%) had a family history of congenital or progressive hearing loss, but the exact genetic cause was unknown. These numbers are in keeping with the 40.3% (95% CI 32.8 to 48.0) unknown cases reported in a systematic literature review of 16 studies that investigated the aetiology of deafness in 5069 paediatric cochlear implant recipients implanted from 2001 to 2011 (Petersen, Jørgensen,& Ovesen, 2015).

5.3.2. Co-morbidities

In the present study, 47 recipients (22%) presented with at least one co-morbidity. Some recipients presented with more than one co-morbidity. The prevalence of the various co-morbidities is summarized in Table 7.

Table 7: Prevalence of co-morbidities

Co-morbidity	%
Cognitive impairment	30,2
Learning problem or language learning problem	34
Attention deficit disorder (ADD)	5,7
Cerebral palsy	5.7
Blind or visual impairment	7.5
Dyspraxia	3.7
Autism	1,9
Developmental delay	7,5
Emotional and behavioral problems	1,9
Epilepsy	1,9

The most prevalent co-morbidities in the study population were cognitive impairment and learning or language learning problems.

5.4. Pre-operative audiological characteristics of the recipients

The majority of recipients, 207 (96%) presented with bilateral sensorineural hearing loss and 9 (4%) recipients were diagnosed with bilateral Auditory Neuropathy Spectrum Disorder (ANSD). The first report on auditory neuropathy was published in 1996 (Starr et al., 1996). It is therefore possible that ANSD was not differentiated from SNHL prior to 1996 in the study cohort.

5.4.1. Study groups

As discussed in section 4.6.4.1, recipients with SNHL were grouped according to a prelingual, perilingual and postlingual onset of severe to profound SNHL. Recipients diagnosed with ANSD were the fourth group. The number of recipients in the 4 study groups are summarized in Table 8. It is indicated how many recipients in the prelingual, perilingual and postlingual onset of deafness groups, suffered from a progressive hearing loss.

Table 8: Summary of the recipients in the 4 study groups (N=216)

Study Groups	n	%	Progressive loss
Group 1 Prelingual onset of deafness (age of onset: 0 to 23 months)	166	77	3
Group 2 Perilingual onset of deafness (age of onset: 24 to 59 months)	17	8	7
Group 3 Postlingual onset of deafness (age of onset: 60 months and older)	24	11	11
Group 4 Auditory Neuropathy Spectrum Disorder	9	4	0

5.4.1.1. Group 1: Prelingual onset of severe to profound SNHL (0 to 23 months)

The majority of the recipients in the study cohort had a prelingual onset of severe to profound SNHL. For 149 (90%) of these recipients the onset was congenital, while 14 recipients (8%) acquired a sudden hearing loss, after birth, due to meningitis (12), a pyrexial illness (1) and a temporal bone fracture (1). Three recipients in this group suffered from a progressive hearing loss that had reached a severe to profound level by 23 months. For 2 of these recipients with progressive hearing loss the aetiology was unknown and 1 recipient had a CMV infection.

5.4.1.2. Group 2: Perilingual onset of severe to profound SNHL (24 to 59 months)

Ten recipients (59%) in this group acquired a sudden hearing loss due to meningitis (6), mumps (1), an unknown viral cause (1) and ototoxic drugs (1). The remaining 7 recipients (41%) suffered from a progressive hearing loss. For 4 of these recipients with progressive hearing loss the aetiology was unknown, 1 participant was born prematurely, 1 participant had De Toni Fanconi Sydrome and 1 participant had a family history of progressive SNHL.

5.4.1.3. Group 3: Postlingual onset of severe to profound SNHL (60 months and older)

As was observed for the perilingual group, the majority (54%) of the recipients with a postlingual onset of hearing loss, lost their hearing suddenly. This was due to meningitis (6), mumps (4), ototoxic treatment (1), a viral infection (1) and for 1 recipient the cause was unknown. The aetiologies for the recipients with progressive hearing loss in this group were mostly unknown, but 1 recipient had a family history of progressive SNHL, 1 recipient developed a progressive hearing loss after mumps, 1 recipient had De Toni Fanconi Syndrome and the last recipient had Pendred Syndrome.

5.4.1.4. Group 4: Auditory Neuropathy Spectrum Disorder (ANSD)

The onset of the hearing loss for the 9 recipients with ANSD was congenital.

5.4.2. Pre-operative residual hearing

As discussed in section 4.6.4.2, the 4-frequency pure tone average (PTA) of unaided air-conduction thresholds at 500, 1000, 2000 and 4000Hz were calculated for the right and left ears separately. Only recipients with bilateral comprehensive threshold information at the required 4 frequencies were included. The 4-frequency average for the ear with the most residual hearing served as a proxy for residual hearing prior to cochlear implantation (Niparko et al., 2010).

The mean 4-frequency PTA and standard deviation of the better ear, are summarized in Table 9. A pairwise comparison of the means was performed, using the LSD test.

Table 9: Pre-operative residual hearing			
Study groups	**n**	**Mean 4-fr PTA** [1]	**SD**
Group 1: Prelingual onset	114	101.75 [a]	17.45
Group 2: Perilingual onset	16	104.18 [a]	18.52
Group 3: Postlingual onset	24	99.99 [a]	18.29
Group 4: ANSD group	8	70 [b]	17.55
Note. Mean 4-fr PTA=Mean 4-frequency pure tone average; SD=standard deviation			

Bilateral comprehensive threshold data were available for 69% of the recipients in the prelingual onset group. Pure-tone audiometry requires behavioural responses and cannot reliably be performed by very young children (Chiossi & Hyppolito, 2017). In agreement with the quality standards guidelines for pediatric cochlear implantation of the South African Cochlear Implant group (SAGIC, 2019), the pre-operative degree of hearing loss for young children in the prelingual onset group was confirmed by means of objective electrophysiological tests. It was therefore not possible to calculate the 4-frequency PTA for some of these recipients.

As seen in Table 9, the mean 4–frequency PTA for the recipients with ANSD was significantly better when compared with the three groups with SNHL. On average the mean 4-frequency PTA for the recipients in the three SNHL groups was greater than 90dBHL in the better ear, indicating that on average these recipients suffered from a bilateral profound SNHL prior to cochlear implantation (Smith et al., 2005). However, the standard deviation from the mean PTA was greater than 17dBHL for all three groups. This spread in data around the mean is consistent with the expansion in criteria for pediatric cochlear implantation to include children with more pre-operative residual hearing (Zwolan & Sorkin, 2016).

[1] The letters in Table 9 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be > 0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be < 0.05.

5.4.3. Pre-operative speech perception

The speech perception ability of the hearing impaired child, as measured with appropriately fitted hearing aids, is one of the main considerations in the selection of suitable candidates for cochlear implantation (Dowell, 1997). As discussed in section 4.6.4.3 speech perception measurements involve a hierarchy of tasks ranging from the detection of speech sounds, the recognition of words or sentences from a limited set (closed set speech perception) to the identification of words or sentences from an infinite number of possibilities (open set speech perception). Early candidacy criteria for cochlear implantation were conservative and defined by the inability to recognize speech in a closed set (Staller et al., 1991). Although candidacy criteria were expanded to include children with minimal aided open-set speech perception abilities, the rationale behind implantation must be that the child will derive more benefit from the cochlear implant than from the hearing aid (Zwolan et al., 1997).

As discussed in section 4.6.4.3, the pre-operative speech perception tests results were placed into one of 8 categories (categories 0 to 7) and are summarized in Table 10.

Table 10: Pre-operative speech perception categories (N=216)

Study groups	**Percentage of the recipients in closed-set categories**					**Percentage of the recipients in open-set categories**		
	Cat 0	Cat 1	Cat 2	Cat 3	Cat 4	Cat 5	Cat 6	Cat 7
Group 1: Prelingual onset	71	13	3	0	1	6	6	0
Group 2: Perilingual onset	59	17	0	12	0	6	6	0
Group 3: Postlingual onset	50	8	0	0	0	21	21	0
Group 4: ANSD	11.2	33.3	0	0	0	22.2	33.3	0

Note. CI=Cochlear Implant; Cat=Category; all values

In both the prelingual and perilingual onset groups 88% of the recipients could not perform open set speech perception tasks pre-operatively, while the remaining 12% were able to. In both these groups none of the recipients who demonstrated open-set speech perception prior to cochlear implantation had reached category 7 while using hearing aids. Forty-two percent of the recipients in the postlingual onset group and 55.5 % of the recipients with ANSD were able to perform open-set speech perception tasks pre-operatively, but had also not reached category 7.

5.4.4. Age at intervention and duration of deafness prior to intervention

5.4.4.1. Age at hearing aid fitting and duration of deafness prior to hearing aid fitting

As discussed in section 4.6.4.4, the age at hearing aid fitting and duration of deafness prior to hearing aid fitting were recorded for the ear that was implanted first. The mean age at hearing aid fitting and standard deviation for the recipients who used amplification prior to cochlear implantation are summarized in Table 11.

Table 11: Mean age at hearing aid fitting

Study groups	**Recipients who used ha’s**	**Mean age in months at ha fitting**	**SD**
Group 1: Prelingual onset (n=166)	163	16.68	10.68
Group 2: Perilingual onset (n=17)	17	38.88	10.8
Group 3: Postlingual onset (n=24)	21	101.16	41.28
Group 4: ANSD (n=9)	9	21.72	16.68

Note ha=hearing aid, SD=Standard deviation

The mean duration of deafness prior to hearing aid fitting and the standard deviation, for the study groups, are summarized in Table 12.

Table 12: Mean duration of deafness prior to hearing aid fitting			
Study groups	**Recipients who used ha's**	**Mean duration prior to ha** [2]	**SD**
Group 1: Prelingual onset (n=166)	163	15.6 [a]	10.92
Group 2: Perilingual onset (n=17)	17	3.72 [b]	6.36
Group 3: Postlingual onset (n=24)	21	5.28 [b]	11.28
Group 4: ANSD	9	16.44 [a]	8.76
Note ha=hearing aid, SD=standard deviation, duration=months			

The majority of the recipients in all four study groups used bilateral amplification before cochlear implantation. Three recipients (1.8%) in the prelingual onset group and 3 recipients (12.5%) in the postlingual group had no measurable hearing in the ear that was implanted and therefore did not use hearing aids prior to implantation. The duration of deafness prior to amplification was significantly longer for recipients with a prelingual onset of deafness and recipients with ANSD.

5.4.4.2. Age at cochlear implantation and duration of deafness prior to cochlear implantation

The mean age at cochlear implantation and standard deviation are summarized in Table 13.

Table 13: Mean age at cochlear implantation		
Sub group	**Mean age at CI**	**SD**
Group 1: Prelingual onset (n=166)	54.6	47.88
Group 2: Perilingual onset (n=17)	65.16	29.4
Group 3: Postlingual onset (n=24)	137.52	39.96
Group 4: ANSD (n=9)	55.92	30
Note. CI=cochlear implant; SD=standard deviation; age in months		

[2] The letters in Table 13 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be > 0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be < 0.05.

The mean duration of deafness prior to cochlear implantation and standard deviation, for the study groups, are summarized in Table 14. The LSD test was used to perform a pairwise comparison of the mean values. Means that were significantly different based on the LSD analysis were labelled with different letters in the table.

Table 14: Mean duration of deafness to cochlear implantation

Study groups	**Mean duration of deafness to CI** [3]	**SD**
Group 1: Prelingual onset (n=166)	53.64 [a]	48.36
Group 2: Perilingual onset (n=17)	28.92 [b]	28.44
Group 3: Postlingual onset (n=24)	38.28 [ab]	35.76
Group 4: ANSD	50.52 [ab]	31.2

Note. CI=cochlear implant, SD=standard deviation.

The duration of deafness prior to implantation for the recipients in the perilingual onset group were significantly shorter than the duration of deafness for the recipients with a prelingual onset of deafness. No further inter-group differences were observed.

Summary and discussion

The audiological characteristics of the recipients pertaining to their type of hearing loss, the age at onset of deafness, the pre-operative degree of hearing loss, the pre-operative speech perception ability and the duration of deafness prior to intervention were reported in section 5.4.

In summary, the majority of recipients in the study cohort suffered from a sensorineural hearing (SNHL) loss of congenital onset. On average, the degree of hearing loss for the recipients with a SNHL was greater than 90dBHL, consistent with a bilateral profound SNHL (Smith et al., 2005). However, the variation in the pre-operative pure tone thresholds documented for recipients with

[3] The letters in Table 14 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be > 0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be < 0.05.

SNHL are consistent with the expansion in the pediatric cochlear implantation criteria and confirmed that some of the recipients in the study cohort had better pre-operative residual hearing than the average (Zwolan & Sorkin, 2016). The average pre-operative hearing level of recipients with ANSD were significantly better than that of recipients with SNHL. In ANSD there will be a discrepancy between the behavioral thresholds and speech perception ability. The pre-operative hearing levels of children with ANSD are therefore not a true reflection of their listening ability (Harrison et al., 2015).

All the recipients in the study cohort used bilateral hearing aids pre-operatively, except for those who had no measureable hearing. As discussed in section 2.8.1 the ability of the child to perform open set speech perception tasks, is of particular importance, as it is considered to be a fundamental pre-requisite skill necessary for spoken language development (Barnard et al., 2015; Kirk et al., 1997). The speech perception ability of the deaf child, as measured with appropriately fitted hearing aids, is therefore one of the main considerations in the selection of suitable cochlear implant recipients (Dowell, 1997). Although selection criteria were expanded to include children with minimal aided open-set speech perception, the decision to proceed with cochlear implantation is still based on unsatisfactory speech perception development with conventional amplification (Zwolan et al., 1997). Although some recipients in the dataset were able to perform open set speech perception tasks, they had reached a plateau with conventional hearing aids and were not able to progress to category 7. The majority of recipients with SNHL were not able to perform open set speech perception prior to implantation.

The average duration of deafness before hearing aid fitting was significantly longer for recipients with a prelingual hearing loss or ANSD. Although considerable variation in the data was observed, the average age at hearing aid fitting for both these groups were later than the recommended age of 6 months (Joint Committee on Infant Hearing, 2007) and the average age at cochlear implantation was later than the recommended age of 12 months (Dettman et al., 2007; Dettman et al., 2016).

It was not documented how many recipients in the study cohort had access to newborn hearing screening (NHS), however as reported by Meyer, Swanepoel, Le Roux and Van der Linde (2012) and Theunisen and Swanepoel (2008), only a limited number of hospitals in South Africa in both the public and private health care sectors offer NHS. As documented by Le Roux et al., (2015), it is likely that the lack of access to NHS contributed to the delay in intervention in the study cohort.

5.5. Cochlear implant systems

All recipients received multichannel cochlear implant systems manufactured by Cochlear Limited® (Appendix B) and were implanted between February 1990 and December 2014. In this cohort of 216 recipients, 139 recipients (64.4%) received a cochlear implant in one ear, 64 recipients (29.6%) received bilateral implants sequentially and 13 recipients (6%) received bilateral simultaneous implants. A total of 293 implants were implanted over the period of 24 years and 10 months. The types and numbers of implant systems that were used are summarized in Table 15.

Table 15: Implant systems used from 1990 to 2014

Implant name	Number of implants
Cochlear Nucleus Profile with Contour Advance Electrode	37
Cochlear Nucleus CI422 Cochlear Implant	3
Nucleus Freedom with Contour Advance Electrode	139
Nucleus 24 with Contour Advance Electrode	22
Nucleus 24 Contour Electrode	18
Nucleus 24k with Straight Electrode	3
Nucleus 24 with Double array	1
Nucleus 24 with Straight Electrode	36
Nucleus 22	34
Total	293

Recipients were implanted with a variety of implant systems that were commercially available from 1990 to 2014. The Nucleus Freedom with Contour Advance electrode was used in 139 of the 293 procedures, thus more than any other device. This electrode was available from 2005 and was only replaced when the Cochlear Nucleus Profile with Contour Advance Electrode became available in 2014.

5.6. Device use after cochlear implantation

Although an investigation regarding the outcomes of pediatric cochlear implantation will involve many different variables that will be important to parents and funders, the most important group to consider will be the children themselves. The child's experience of benefit will be indicated by their choice to use the device or not (Archbold, 2002). Further, device non-use will have a detrimental effect on the cost-effectiveness of cochlear implantation (Magro et al., 2018). None of the recipients in the study population were identified as non-users. This result compares favorably with the 4% of children reported by Uziel et al. (2007) who became non users over a 10 year period and the 7% who stopped using their cochlear implants during a 15 year review, as documented by Raine, Summerfield, Strachan, Martin and Totten (2008).

5.7. Mode of communication after cochlear implantation

The majority of the recipients (206/216, 95.4%) were oral communicators, while 5 recipients (2.3%) used total communication and another 5 recipients (2.3%) communicated via South African Sign Language.

5.8. Speech Perception Development after Cochlear Implantation

As discussed in section 4.6.8, the postoperative speech perception results for the recipients were reviewed retrospectively and available data from annual visits was recorded. The duration of implant use for the recipients at the time of data capturing was between 3 years and 27 years 11months. As a result, the number of annual tests intervals varied among the recipients. Ideally, every recipient should have had speech perception results available at every annual interval of implant use. This has not been achieved for a number of practical constraints, for instance incomplete test information in records and missed appointments.

As discussed in section 4.6.4.3, the speech perception test score at annual visits, as measured by a range of speech perception tests, without visual cues, in different languages and test conditions (live voice or recorded speech) were categorized according to a scale of 8 speech perception categories. These categories from 0 to 7 are summarized in section 4.6.4.3. Categories 1 to 4 represent closed set speech perception indicating that the recipient was able to recognize objects or pictures from a limited set of words and categories 5 to 7 indicate that the recipient was able to perform open set tasks by identifying words or sentences from an unlimited number of possibilities.

As discussed in section 2.8.1, the integration of sensory, linguistic and cognitive processes involved in performing open set speech perception tasks are considered to be a fundamental pre-requisite skill necessary for spoken language development (Kirk et al., 1997; Barnard et al., 2015). As documented in section 5.4.3, the majority of recipients were not able to perform open set speech perception tasks prior to cochlear implantation.

5.8.1. Longitudinal speech perception development over a 10-year period

In an attempt to obtain an overview of speech perception development for the recipients in the dataset, the mean score in speech perception category per test interval were calculated for recipients with available results. The use of a mean speech perception category score was described by Illg et al. (2017) in their retrospective review of pediatric cochlear implant recipients.

The speech perception development trajectory for the recipients over a 10-year period after cochlear implantation is displayed in Figure 3.The number of recipients with results per test interval, the mean score in speech perception category and standard deviation are summarized in Table 16.

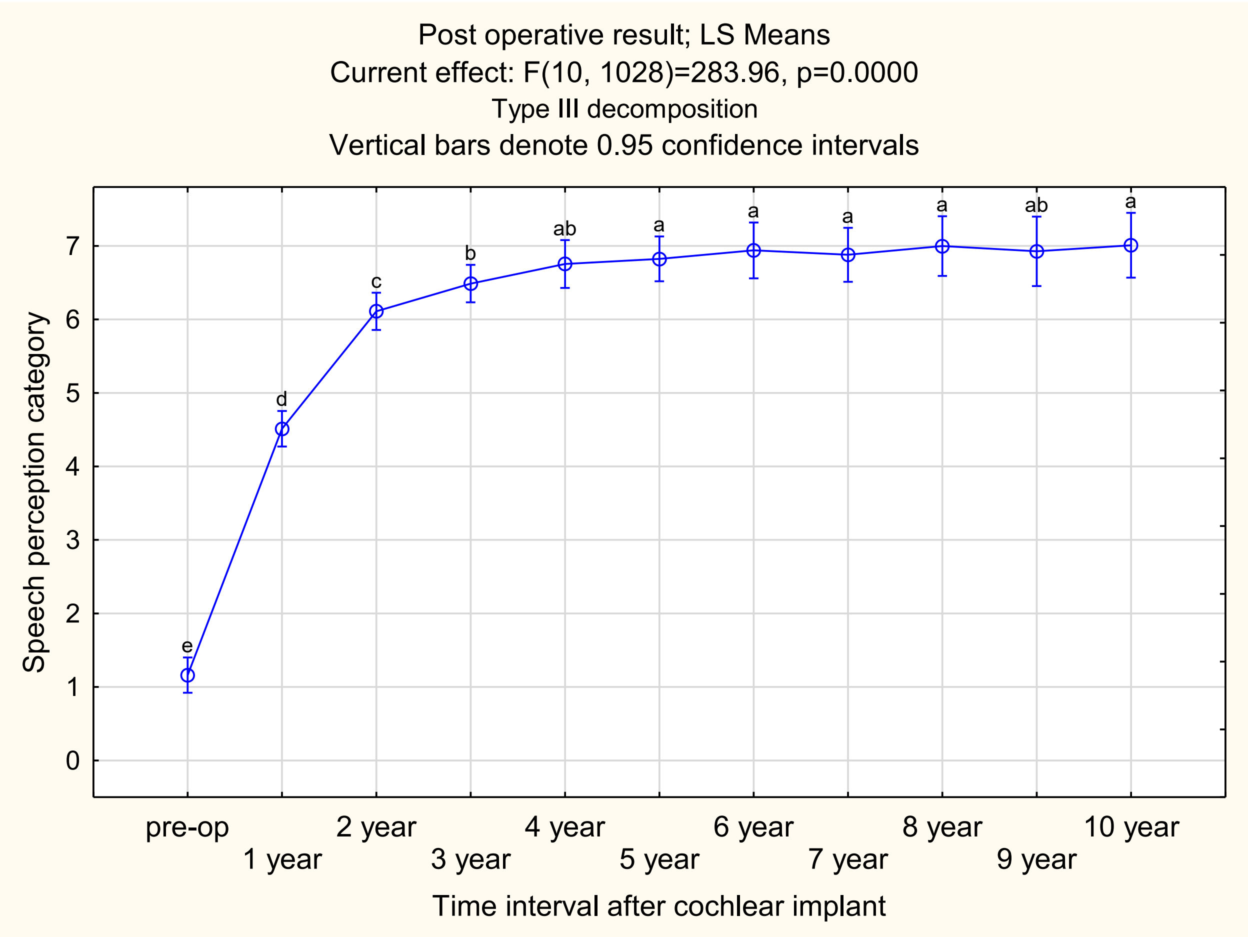

Figure 3. Speech perception development trajectory 10 years after cochlear implantation

Note: The letters in Figure 3 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. In this way, any mean on the graph can be compared to any other mean. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be >0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be <0.05)

Table 16: Mean score in speech perception category			
Time interval	**Recipients with results**	**Mean score in speech perception category**	**SD**
Pre-op	216	1.15	2.04
1 year	206	4.52	2.60
2 year	183	6.10	1.75
3 year	179	6.50	1.23
4 year	93	6.45	1.49
5 year	110	6.61	1.36
6 year	63	6.60	1.33
7 year	69	6.64	1.27
8 year	54	6.74	1.08
9 year	38	6.61	1.42
10 year	45	6.73	1.16
Note. pre-op =pre-operative; SD=Standard deviation			

As seen in Figure 3 and Table 16, the most significant change in the mean score in speech perception category was observed between the pre-operative and one year intervals, between the one and two year intervals and between the two and three year intervals. On average, recipients could not perform open-set speech perception tasks pre-operatively and one year after implant use, but were able to do so after a second year of use. On average, the performance of the recipients remained stable after three years of implant use.

As documented in the study by Staller et al. (1991), the sample size was different at each annual interval and therefore does not allow a direct comparison of the same recipients. However, a clear trend of improvement for the recipients in this dataset was observed during the first 3 years of cochlear implant use.

In the next section, the speech perception development trajectory will be separated for the recipients in the different SNHL onset groups and recipients with ANSD.

5.8.2. Speech perception development for the study groups over a 3-year period

The speech perception development trajectory for the 4 study groups, 3 years after cochlear implantation are displayed in Figure 4. The number of recipients with results, the mean score in speech perception category and the standard deviation are summarized in Table 17.

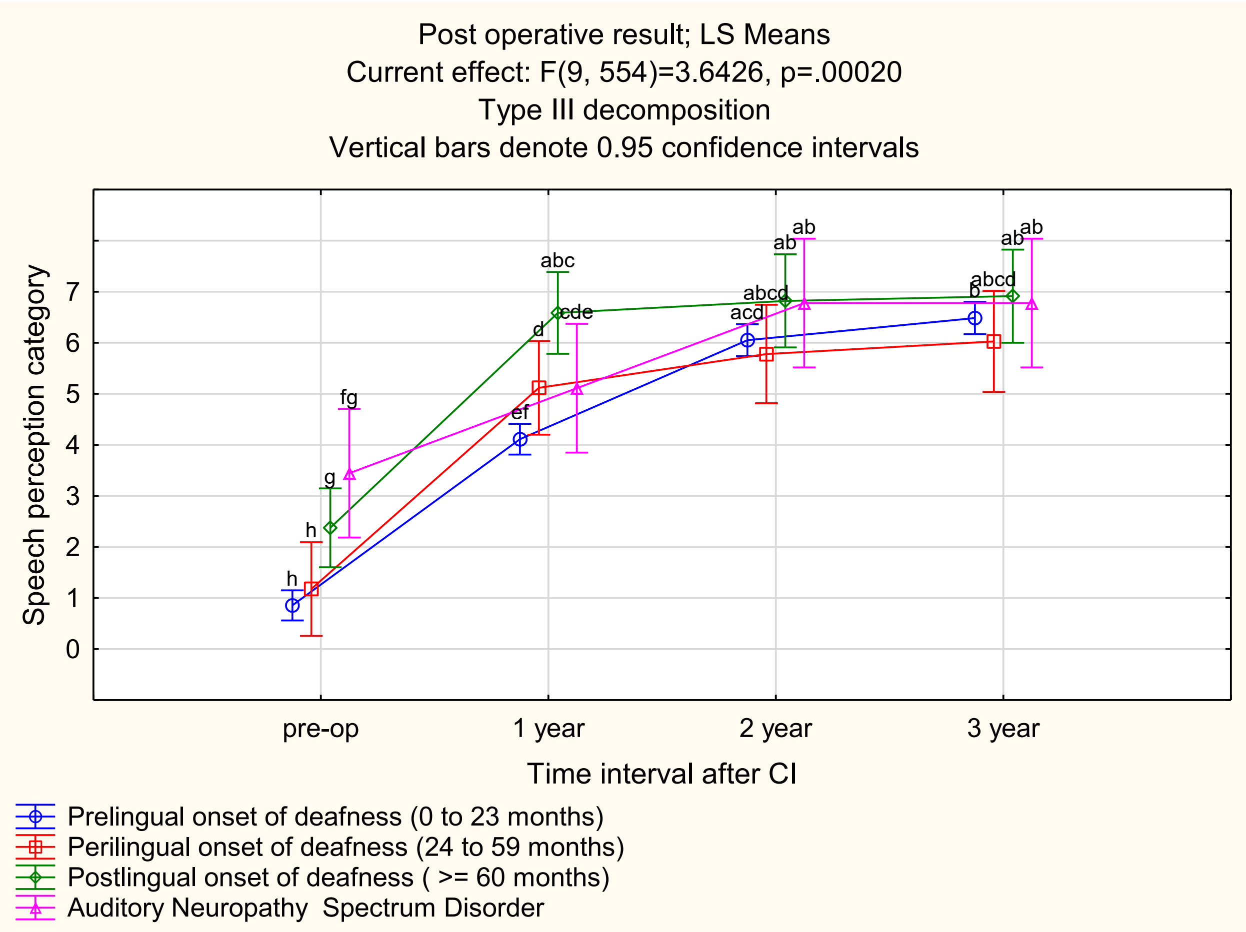

Figure 4. Speech perception development trajectory for the 4 study groups 3 years after cochlear implantation

Note: The letters in Figure 4 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. In this way, any mean on the graph can be compared to any other mean. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be >0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be <0.05.

Table 17: Mean score in speech perception category for the 4 study groups

Study group	Year 1			Year 2			Year 3		
	n	**MSSPC**	**SD**	**n**	**MSSPC**	**SD**	**n**	**MSSPC**	**SD**
Group 1 Prelingual onset	158	4.13	2.6	143	6.01	1.79	140	6.48	1.24
Group 2 Perilingual onset	17	5.12	2.71	15	5.87	2.36	14	6.07	1.9
Group 3 Postlingual onset	22	6.64	1.14	16	6.81	0.54	16	6.88	0.34
Group 4 ANSD	9	5.11	2.47	9	6.78	0.67	9	6.78	0.67

Note. MSSPC=mean score in speech perception category, SD=standard deviation

As seen in Figure 4 and Tabel 17, a significant improvement in the mean speech perception category score was observed for all 4 groups between the pre-operative and one year test intervals. As documented by Staller et al. (1991) the performance of the recipients with a prelingual hearing loss in the present study was significantly lower than that of recipients with a peri- or postlingual hearing loss, one year after implantation. On average the recipients with a prelingual hearing loss were only able to perform open set speech perception tasks after a second year of implant use. Although no significant differences between the 4 groups were observed from the second annual interval, the prelingual onset group continued to make gradual progress and a further significant improvement was observed between year two and year three.

5.8.3. Incomplete records

Incomplete records can lead to nonresponse bias, if the results of the recipients with incomplete records should differ from the results of recipients with complete records (Worster et al., 2004). The results of recipients with incomplete records at the 3 year test interval were therefore reviewed. If a recipient had already reached an open set speech perception category at the first or second year test interval, it was accepted that they could perform the task, as it is not likely that performance would deteriorate (Archbold et al., 1995).

In the prelingual onset group 12 of the 16 recipients with incomplete records at year 3 were able to perform open set speech perception tasks at an earlier test interval and 2 of the 3 recipients in the perilingual onset group with incomplete records at year 3, also reached an open set speech perception category earlier. The 8 recipients in the postlingual group with incomplete records were all able to perform open set speech perception prior to year three and the 9 recipients with ANSD had complete records at all 3 test intervals.

5.8.4. Poor performers

In the prelingual onset group, 13 of the 166 recipients could not perform open set speech perception tasks after 3 years of implant use and 4 recipients with incomplete records at the 3 year test interval had not reached an open set category at an earlier test interval. It was documented that 12 of these 17 recipients developed open set speech perception at a later test interval (after 4 years of implant use) but the remaining 5 recipients were never able to perform open set speech perception tasks. Even though they made slower progress, 9 of the recipients in the prelingual onset group who developed open set speech perception after the 4 year test interval communicated via oral language and 3 participants used total communication. The 5 participants who were not able to perform open set speech tasks, even after years of implant use, used sign language or total communication as mode of communication. Three of the 17 recipients in the perilingual onset group had not reached an open set speech perception category after 3 years of implant use and 1 recipient with incomplete records at year 3 had not reached this skill at an earlier test interval. Two of these recipients were able to perform open set speech tasks at a later interval and were able to use spoken language as mode of communication, while the remaining 2 recipients were never able to perform open set tasks and communicated via sign language. All the participants with a postingual onset of hearing loss or ANSD were able to perform open set speech perception tasks after 3 years of implant use.

Summary and Discussion:

The long term speech perception development of the study cohort was reported in section 5.8. On average, the most significant improvement in speech perception for the recipients in this dataset was observed during the first three years after implantation and the majority had reached an open set speech perception category at this test interval. It was documented that 16 recipients (7.4%) did not develop open set speech perception at the 3 year test interval and a further 5 recipients (2.3%) with incomplete test records at the 3 year test interval, had not developed open set speech perception at an earlier test interval. These participants had either a prelingual or perilingual onset of deafness. Although they made slower progress, it was documented that 14 recipients (6.5%) developed open set speech perception at a test interval after 4 years of implant use. The remaining 7 recipients (3.2%) with a pre- or perilingual onset of hearing loss, however never developed open set speech perception.

In summary, it was documented that 195 recipients (90.27%) demonstrated open set speech perception after three years of implant use. A further 14 recipients developed this skill at a later test interval. A total of 209 recipients (96%) were able to perform open set speech perception tasks at the time of data collection. The recipients who could not perform open set speech perception or made slower progress had a prelingual or perilingual onset of deafness.

5.9. Educational placement during basic education

As discussed in section 4.6.9 the progress of the 216 recipients during the 4 stages of basic education (preschool, grade R, Primary and Secondary School) is summarized in Table 18. The historic placement of children in preschools and grade R had not been recorded consistently in case files. The numbers of recipients who had completed these 2 stages were therefore not analyzed.

Table 18: Summary of the educational status of the recipients

Basic education stage	Status of the recipient	Number of recipients
Preschool	Enrolled	2
Grade R	Enrolled	6
Primary School	Enrolled	77
	Completed	131
Secondary School	Enrolled	32
	Completed	87
	Did not complete	12

5.9.1. Educational placement in primary school

At the time of data capturing, 77 recipients were enrolled in primary school and 131 recipients had already completed primary school. The type of schools, as discussed in section 4.6.9, that these 208 recipients attended are displayed in Figure 5.

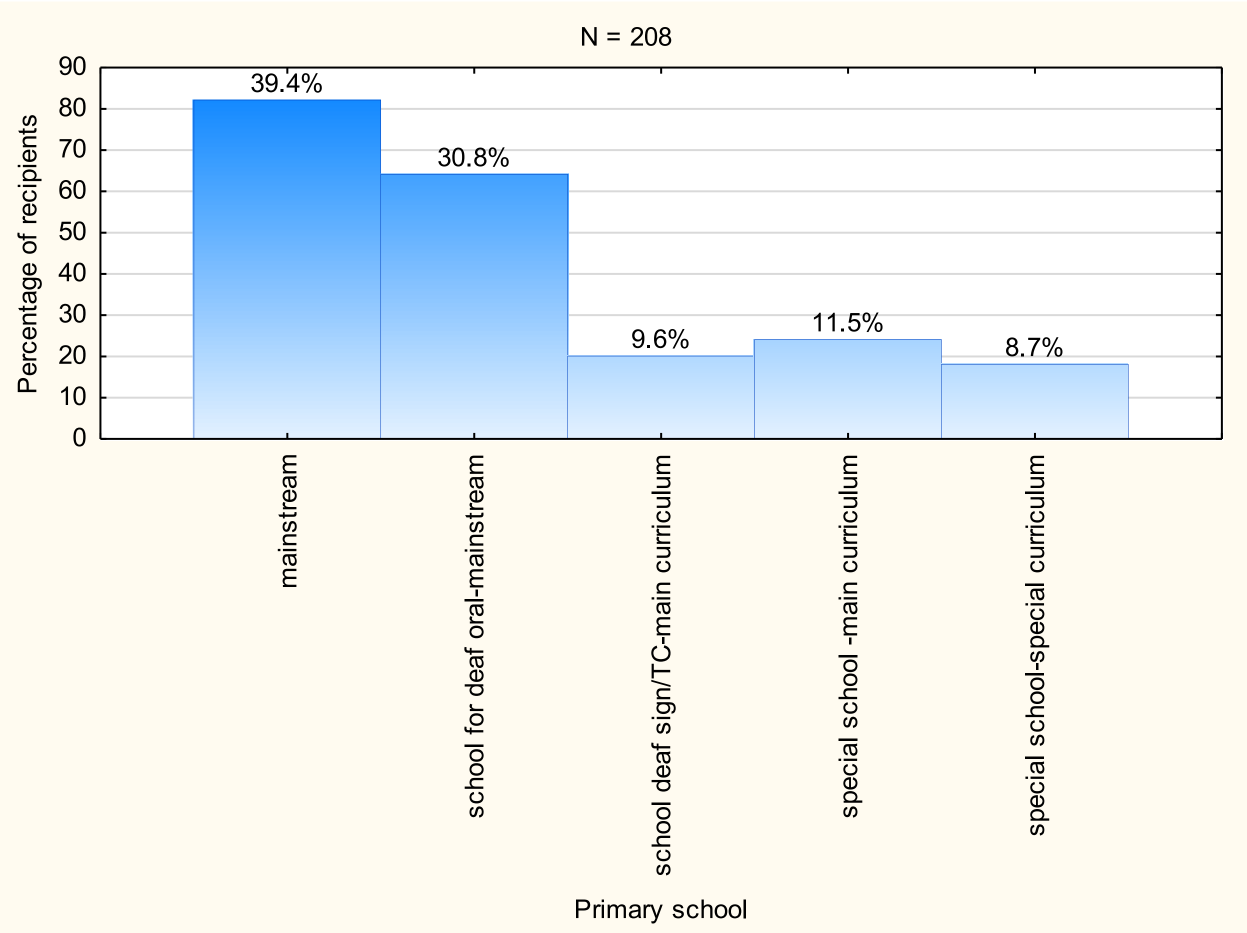

Figure 5. Educational placement of the recipients in primary school (n=208)

As seen in Figure 5, 39.4% of the recipients were in mainstream schools, while 60.6% of the recipients needed some form of special education at primary school level. The type of special school most typically attended was a school for the deaf that used spoken language as the medium for education. Although the majority of recipients were placed in special education, 91.3% of the recipients were able to follow a mainstream curriculum.

5.9.2. Educational placement in secondary school

At the time of data capturing, 32 recipients were enrolled in secondary school education, 87 recipients had completed secondary school and 12 recipients did not complete secondary school. The educational placements for the 131 recipients are displayed in Figure 6.

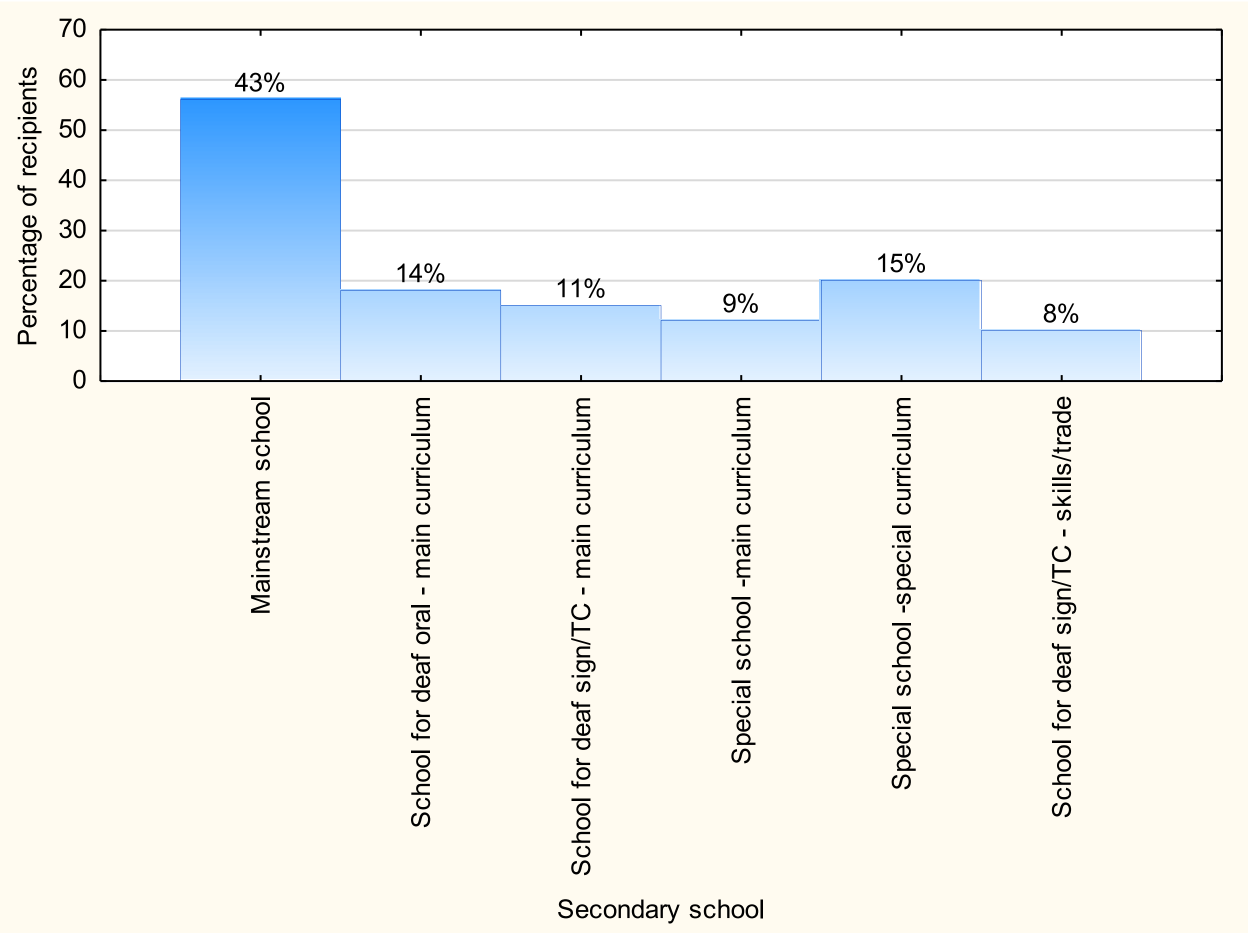

Figure 6. Educational placement in secondary school (n=131)

As seen in Figure 6, 43% of the recipients were placed in mainstream education at secondary school level. As was observed for the recipients in primary school, the majority of the children in secondary school (57%) were placed in special schools. Seventy-seven percent of the recipients were however able to follow a mainstream curriculum at secondary school level. It was documented that 18% of the recipients in secondary school received education via total communication or sign language, compared to the reported 9.6% of the recipients at primary school level. This may be due to the fact that the only English medium oral school for learners with hearing loss in the Western Cape Province, does not offer a trade or special skills curriculum. Some of the recipients who could not complete the mainstream syllabus at secondary school level, therefore transferred to a total communication environment, in order to access the trade or special skills curriculum.

5.10. Higher education and training

As documented in Table 19, 99 recipients had left basic education at the time of data capturing. Fifty-four of these recipients were either enrolled for or had completed higher education and training. As discussed in section 4.6.10, the qualifications these participants had obtained or were studying towards, were categorized according to the NQF and are summarized in Table 19.

Table 19: Summary of the NQF level qualifications obtained or enrolled for (n=54)

NQF level	**Description of qualification**	***n***
1 to 4	Apprenticeship, Learnership or Skills Programs (Include National and Occupational Certificates)	11
5 to 6	Higher Certificate, Advanced Certificate, Occupational Certificate, Diploma	18
7	Bachelors Degree	18
8 to 9	Honours or Masters Degree	7

Note. NQF= National Qualifications Framework

More than half of the recipients who completed basic education were enrolled for or had completed higher education.

5.11. Occupational status

5.11.1. Employment status

Of the 99 recipients who left basic education, 29 recipients were enrolled for full time higher education and training, 1 recipient repeated grade 12 to obtain better grades and 5 recipients took a gap year after secondary school. Forty-seven (73%) of the 64 recipients seeking employment were employed at the time of data capturing and 17 recipients (27%) were unemployed.

5.11.2. Occupational categories

As discussed in section 4.6.11 the occupations of the recipients were categorized according the guidelines of the South African Standard of Occupations (SASCO) and an average skill level was calculated for the recipients who were employed at the time of data capturing. A summary of these occupational categories and associated skill levels is provided in Appendix C.

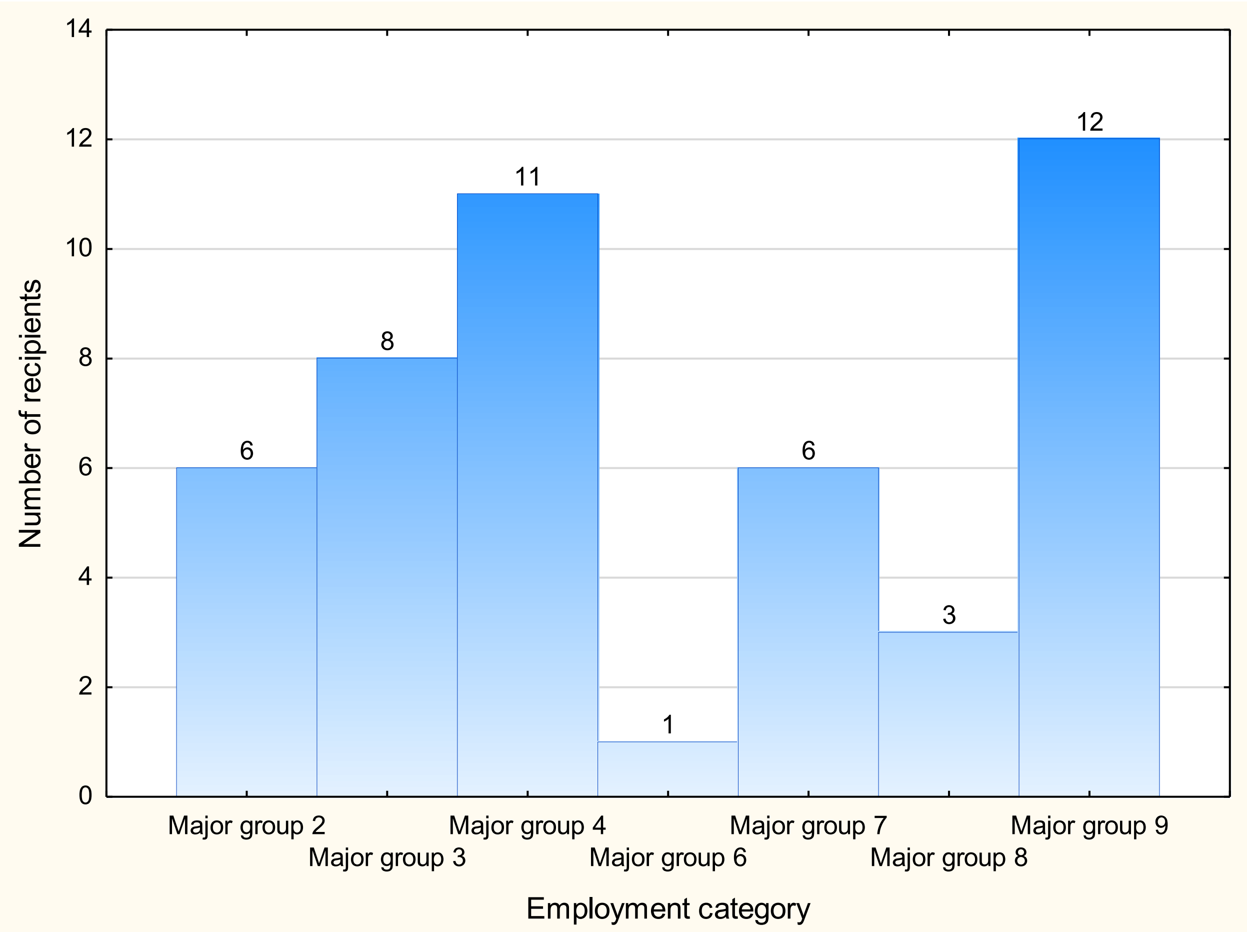

Figure 7. SASCO categories of employment (n=47)

As seen in Figure 7, the recipients were employed in a range of occupations. The average SASCO skill level (as summarized in Appendix C) of the 47 recipients was 2.4 (range: 1 to 4; SD: 0.96). This average skill level, was in keeping with the average occupational skill level of 2.24 (range 1 to 4; SD 0.57) reported by Illg et al., (2017) for a group of German pediatric cochlear implant recipients. To our knowledge, this was the only other study that reported on the long term occupational skill level reached by a group of children who received cochlear implants in childhood.

A skill level of 2 is required for employment as clerical support workers, service and sales workers, skilled agricultural, forestry and fishery workers, craft and related trade workers and plant and machine operators. In the South African context a skill level of 2, typically requires an NQF qualification of level 4 or less (Statistics South Africa, 2012). However, the standard deviation of nearly one point from the mean, indicate variation in the skill level of the recipients in the study cohort. Illg et al. (2017) reported that the average skill level of the cochlear implant recipients in their study was significantly lower than what was reported by the German General Social Survey for 2012. The authors contribute this discrepancy in occupational skill level to the fact that cochlear implant recipients who had not completed secondary school education in mainstream environments will be excluded from tertiary education, which will limit their career prospects.

The variables associated with mainstream educational placement, was investigated in the final objective of this study and will be discussed in the next section.

5.12. Variables associated with mainstream educational placement

As discussed in sections 5.9.1 and 5.9.2, 39.4 % of the recipients received mainstream education at primary school level and 43% of the recipients received mainstream education at secondary school level. As discussed in section 2.8.2, mainstream education will not be the appropriate placement for all cochlear implant recipients and should not be the aim at all cost (Archbold, 2001) However, when appropriate, mainstream education may improve the deaf child's options for tertiary education and future career and earning potential (Ilgg et al., 2017). It could also be argued that the high cost of cochlear implantation may be "partly offset" by savings in the cost of special education (Barton et al., 2006b, p. 187). Mainstream education may therefore contribute to the cost effectiveness of cochlear implantation as an intervention option (Magro et al., 2018).The variables associated with mainstream educational placement in the study cohort were therefore explored.

The investigation of the variables associated with mainstream placement at primary and secondary school level, was conducted in 2 phases: The differences between the mainstream and special education groups were examined for selected variables, namely home language education, access to private or state health services, maternal education level, co-morbidities, age at onset of deafness, duration of deafness prior to hearing aid fitting, duration of deafness prior to cochlear implantation, unilateral or bilateral cochlear implant use and speech perception category, at baseline . The chi-square test was used to examine group differences by categorical variables. For continuous variables, one-way analysis of variance (ANOVA) was used to examine for group difference. In the second phase, logistic regressions were conducted in an attempt to identify the variables that reliably differed for the mainstream or special education groups, independent of other variables. Only the variables with significant differences on the chi-square test or ANOVA were included in the logistic regressions. Significance for all statistical tests were set as $p < 0.05$.

5.12.1. Differences between the mainstream and special education groups

5.12.1.1. Home language education

Primary School

As illustrated by Figure 8, 78 of the recipients (41%) who received home language education were in mainstream schools while 4 of the recipients (22%) who did not receive home language education were in the mainstream. The data for one of the recipients was unknown at the time of data capturing. Although the recipients who received home language education represented a higher percentage of cases in mainstream education, there was not a significant difference between the groups. (Chi-square (df =1) = 2.68, p=0.10). Home language education was therefore not significantly associated with mainstream placement in the primary school.

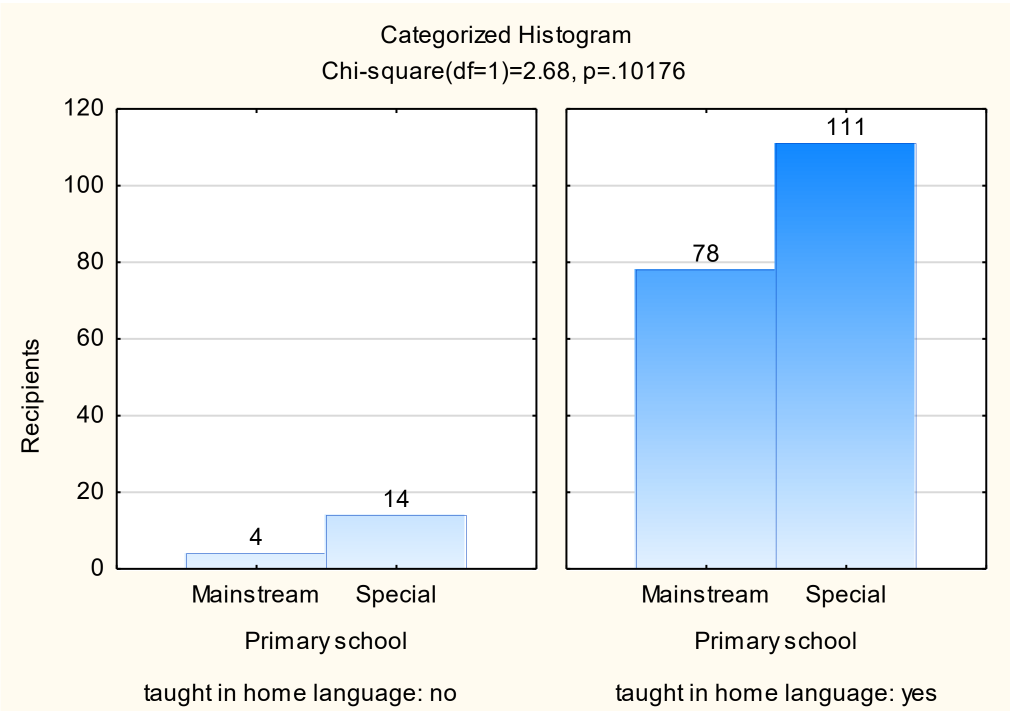

Figure 8. Group differences between recipients in mainstream and special education in terms of language of education in primary school

Secondary School

Home language education was not associated with educational placement in secondary school either. Figure 9 illustrates that the 51 recipients who received home language education represented 43% of the recipients in mainstream schools relative to the 5 recipients (45%) who did not receive home language education (Chi-square (df =1)=0.04, p=85).

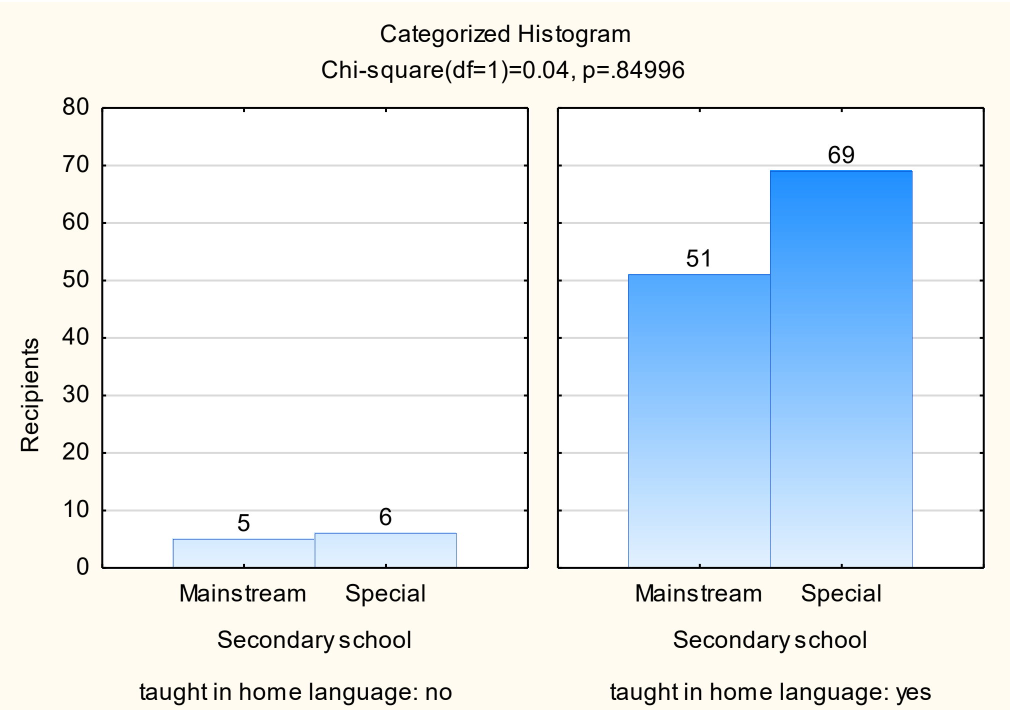

Figure 9. Group differences between recipients in mainstream and special education in terms of language of education in secondary school

5.12.1.2. Private or state sector health services

As discussed in section 4.6.2.1, the income of the recipients was categorized according to the Tygerberg Hospital Income Classification system (Appendix A). As documented in section 5.2.1, 62% of the recipients belonged to a medical aid scheme. These families were classified as private recipients and the 35% of recipients who did not belong to a medical aid were classified as state recipients. For 3% of the recipients, the income category was unknown at the time of data collection.

Primary School

As seen in figure 10, 65 of the private recipients (51%) were in mainstream education in primary school, relative to the 17 state recipients (23%) who were in the mainstream. Access to private medical insurance was significantly associated with mainstream placement at primary school level. (Chi-square (df=1) =15.66, $p<0.01$).

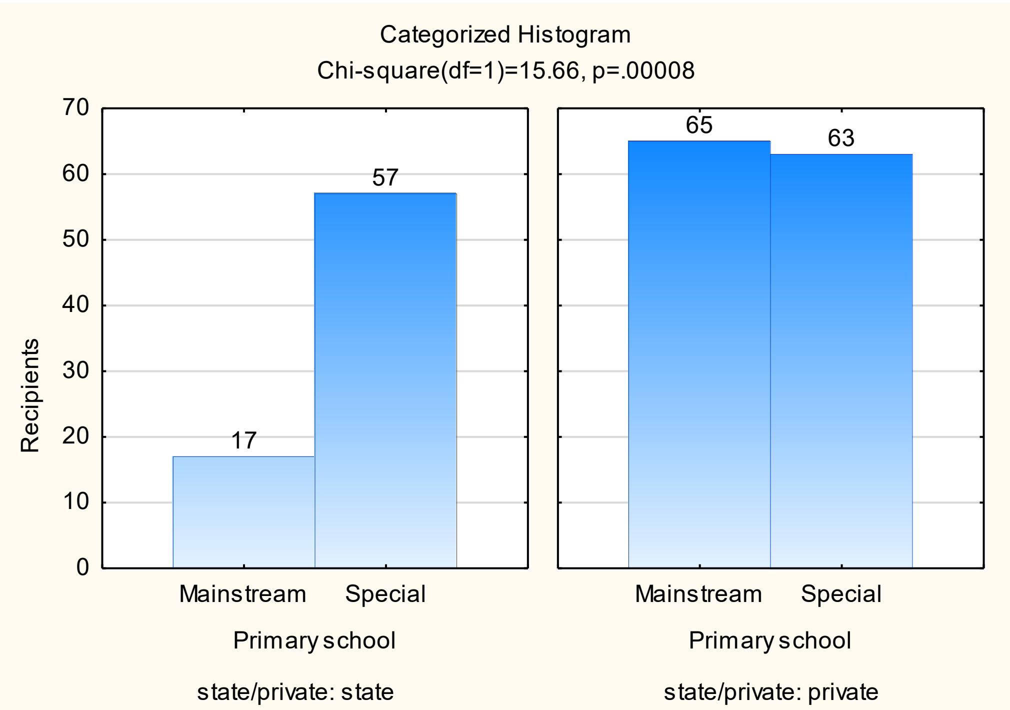

Figure 10. Group differences between recipients in mainstream and special education in terms of access to private or state health services in primary school

Secondary School

As seen in Figure 11, 37 private recipients (47%) were in the mainstream compared to the 18 state sector recipients (38%) in mainstream education at secondary school level. Although private sector recipients once again represented a higher percentage of cases in mainstream education relative to state sector recipients, the difference between the groups was not significant square

(Chi-square(df=1)=1.2,p=.27). The association between private health insurance and educational placement was therefore not significant in secondary school.

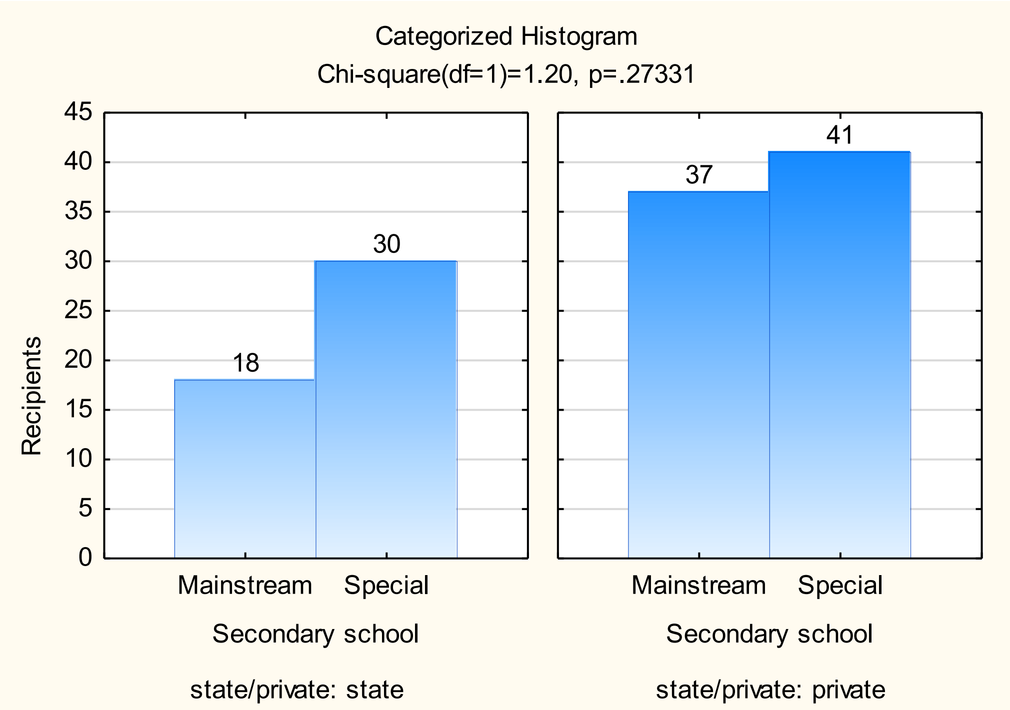

Figure 11. Group differences between recipients in mainstream and special education in terms of access to private or state health services in secondary school

5.12.1.3. Maternal education level

Primary School

As seen in Figure 12, the average maternal education level for the recipients in mainstream education in the primary school was 5.91 (SD: 1.27). As discussed in section 4.6.2.2, this value represents a mean maternal education level between grade 12 and a post grade 12 qualification. The mean maternal education level for the recipients in special education was 4.94 (SD: 1.14) which implies that on average the mothers of recipients in special education did not complete grade 12. The mainstream and special education groups differed significantly in terms of the mean level of maternal education (F1, 194) =31.16, p=<0.01). A higher maternal education level was significantly associated with mainstream placement at primary school level.

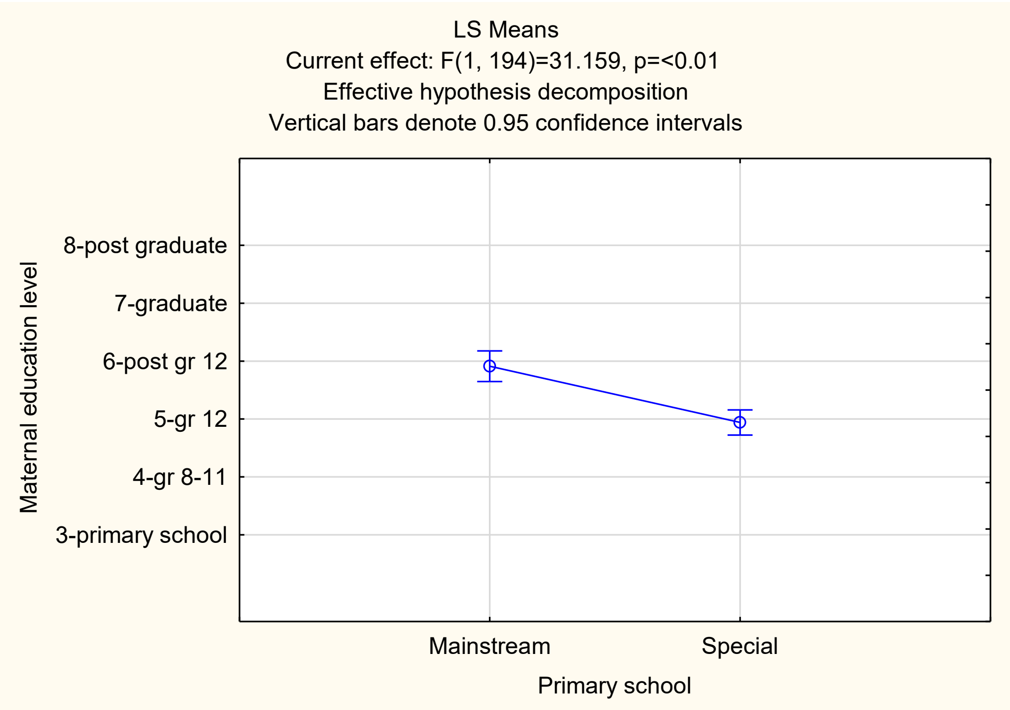

Figure 12. Group differences between recipients in mainstream and special education in terms of maternal education level in primary school.

Secondary School

As seen in Figure 13, a similiar trend was observed at secondary school level. The mean maternal education level for the recipients in mainstream education was 5.7(SD 1.4), which implies that on average the minimum maternal qualification obtained was grade 12. The mean value for mothers of the recipients in special education was 4.97 (SD: 1.13) which indicates that on average the mothers of children in special education did not complete grade 12. The mean maternal education level for mainstream recipients in the secondary school was significantly higher than that of recipients in special education (F (1,118) =10.340, p=<0.01). A higher maternal education level was therefore significantly associated with mainstream educational placement in secondary school as well.

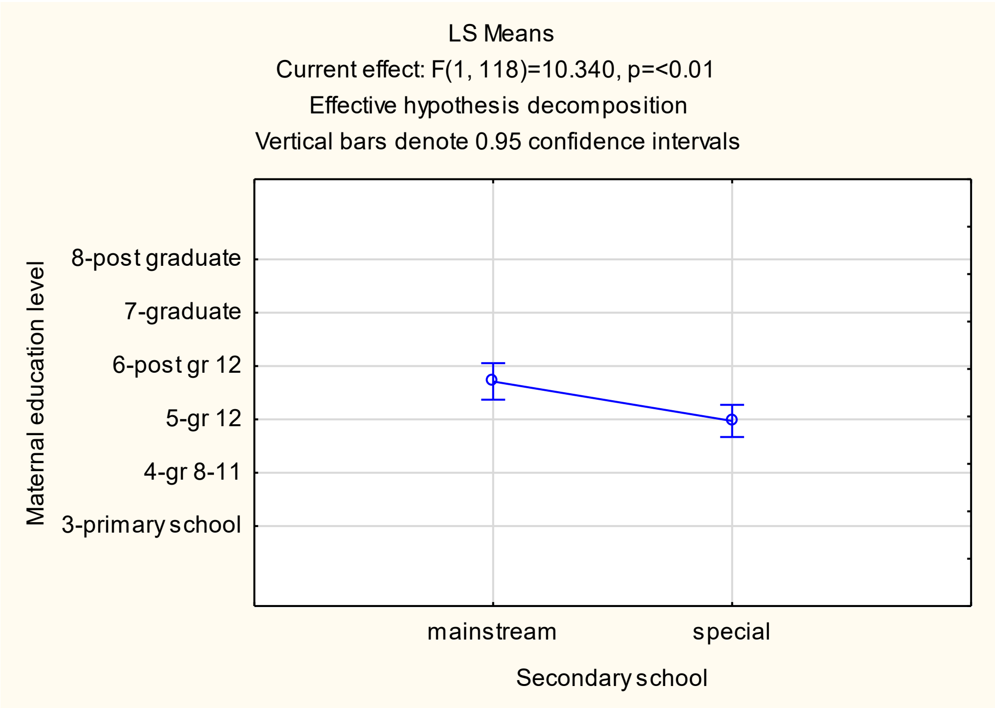

Figure 13. Group differences between recipients in mainstream and special education in terms of maternal education level in secondary school

5.12.1.4 Co-morbidities

Primary School

As seen in Figure 14, 74 of the recipients (46%) without a co-morbidity were in mainstream education, relative to the 8 recipients (17%) with a co-morbidity. The mainstream and special education groups differed significantly in terms of the presence or absence of a co-morbidity (Chi-square (df=1)=13.10, p=0.00). The presence of a co-morbidity was negatively associated with mainstream placement at primary school level.

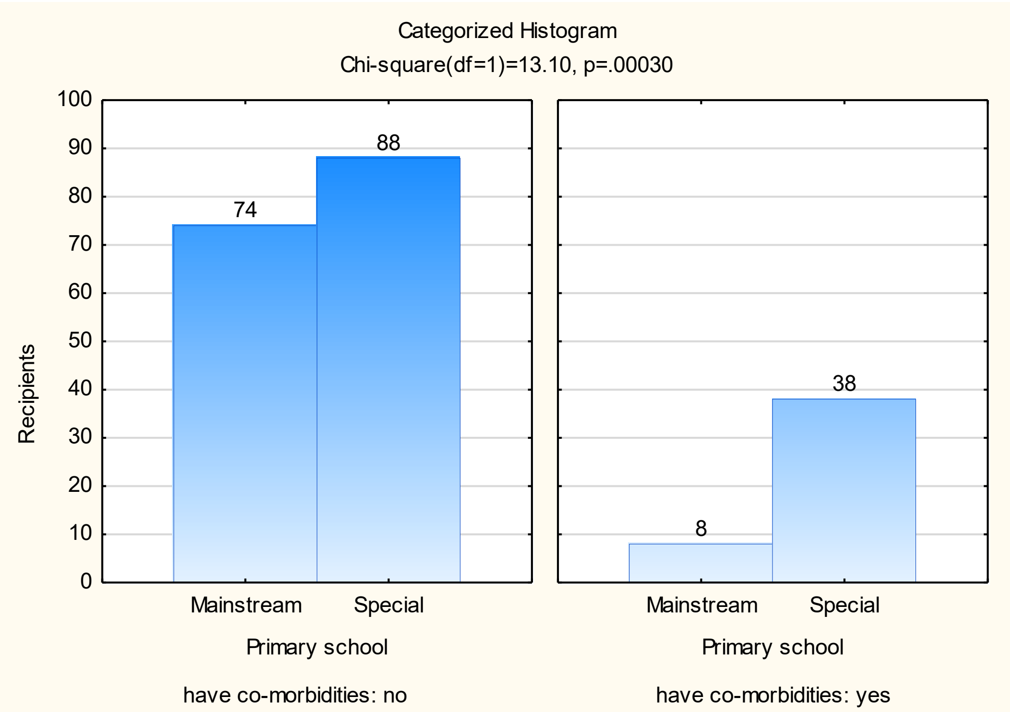

Figure 14. Group differences between recipients in mainstream and special education in terms of the presence of co-morbidities in primary school.

Secondary School

A similar trend was observed at secondary school level. As seen in Figure 15, 51 of the recipients (53%) without a co-morbidity were in mainstream education, while only 5 of the recipients (14%) with a co-morbidity were in the mainstream. The presence of a co-morbidity was negatively associated with mainstream educational placement at secondary school level (Chi-square (df=1)=17.42, p=0,00).

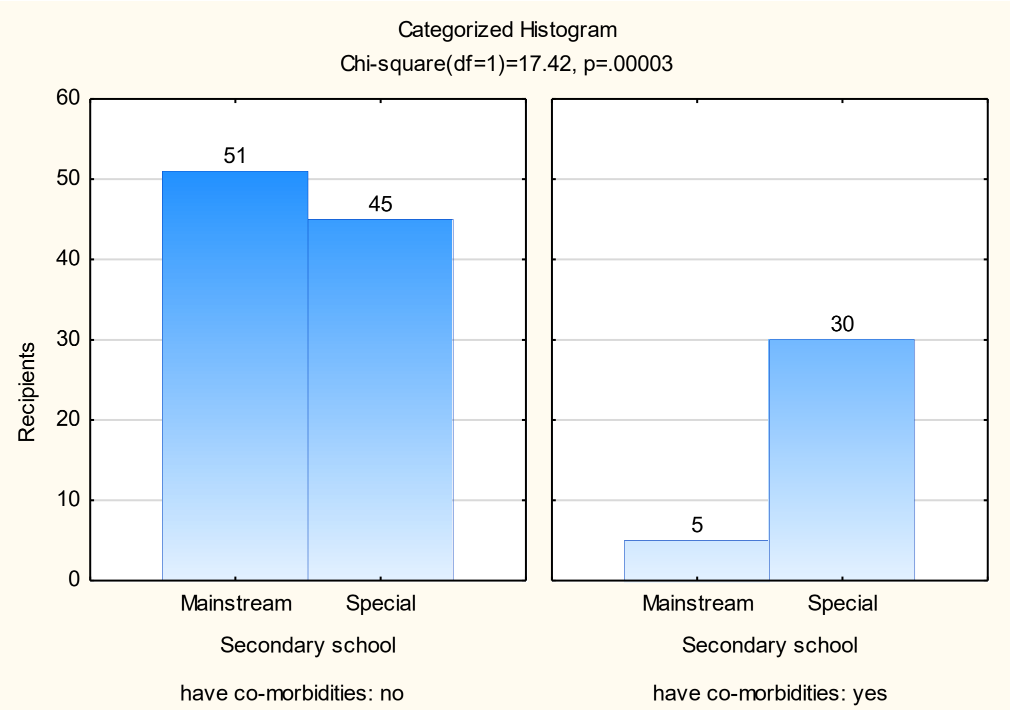

Figure 15. Group differences between recipients in mainstream and special education in terms of the presence of co-morbidities in secondary school.

4.12.1.5. Age at onset of deafness

Primary School

As seen in Figure 16, the mean age at onset of severe to profound SNHL loss or ANSD for the recipients in mainstream schools was 1.59 years (19.08 months). This was later than the mean age at onset of deafness of 1. 06 years (12.7 months) for recipients in special schools. Although recipients in mainstream education had a later onset of hearing loss, than the recipients in special education, this difference was not significant (F (1,204)=1.64, p=0.20). Age at onset of deafness was therefore not significantly associated with mainstream education at primary school level in the study cohort.

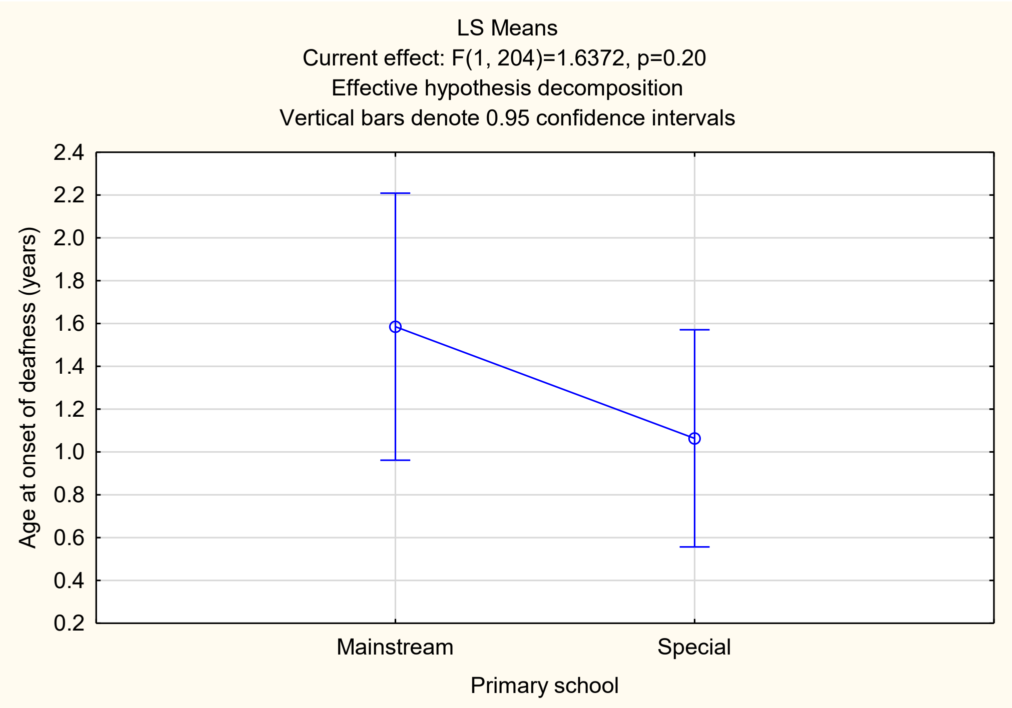

Figure 16. Difference in mainstream and special education groups in terms of the age at onset of deafness in primary school

Secondary School

As seen in Figure 17, the mean age at onset of severe to profound SNHL or ANSD for the recipients in mainstream secondary schools was 2.22 years (26.64 months) and for the recipients in special education it was 1.28 years (15.36 months). Once again, the average age at onset of deafness was later for mainstream participants in secondary schools compared to the recipients in special education, but the difference between the groups was not significant (F=2.4768, p=0.12). The age at onset of deafness was therefore not significantly associated with mainstream placement in secondary school either.

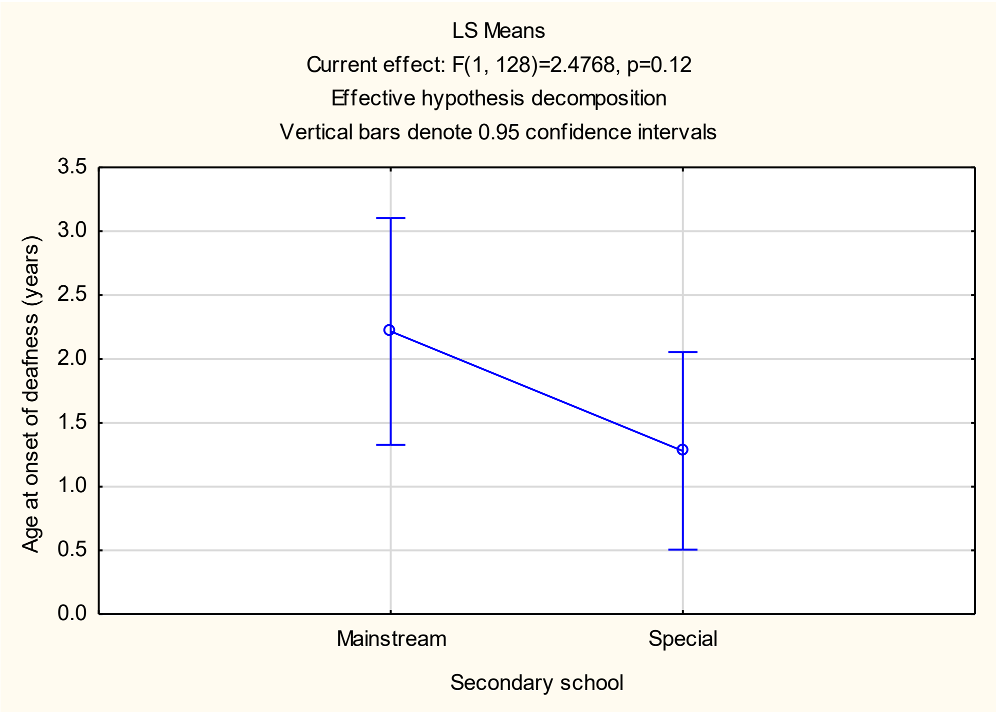

Figure 17. Difference in mainstream and special education groups in terms of the age at onset of deafness in secondary school

5.12.1.6. Duration of deafness prior to hearing aid fitting

Primary School

As seen in Figure 18, the mean duration from the onset of deafness to hearing aid fitting for the recipients in mainstream schools was 0.89 years (10.68 months) and for the recipients in special schools, was 1.31 years (15.72 months). The duration to amplification was significantly shorter for the recipients in mainstream education (F (1,201) = 9.77, p=<0.01). A shorter duration of deafness to hearing aid fitting was therefore significantly associated with mainstream placement in the primary school.

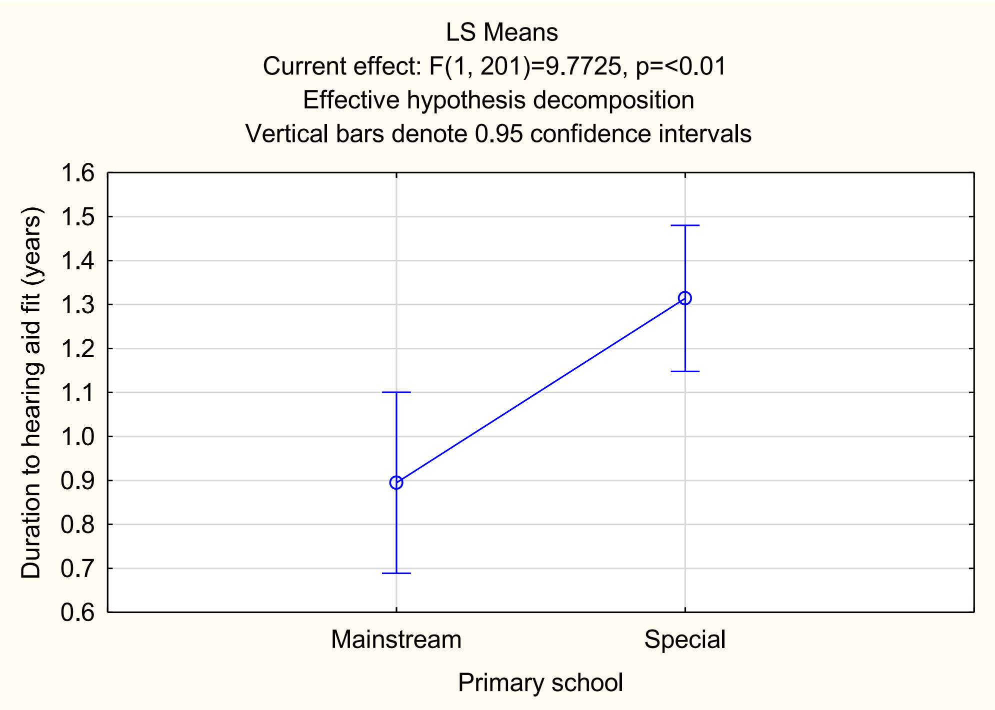

Figure 18. Difference in mainstream and special education groups in terms of the duration of deafness prior to hearing aid fitting at primary school level

Secondary School

As seen in Figure 19, the mean duration between the onset of deafness and hearing aid fitting for the recipients in secondary school mainstream education was 0.97 years (11.64 months). The duration to amplification for the recipients in special schools was 1.21 years (14.52 months). As was observed for the primary school recipients, the duration to amplification was shorter for recipients in mainstream education. This was however not a significant difference (F (1,125) = 1.98, p=0.16). A shorter duration of hearing loss to hearing aid fitting was therefore not significantly associated with mainstream placement at secondary school level.

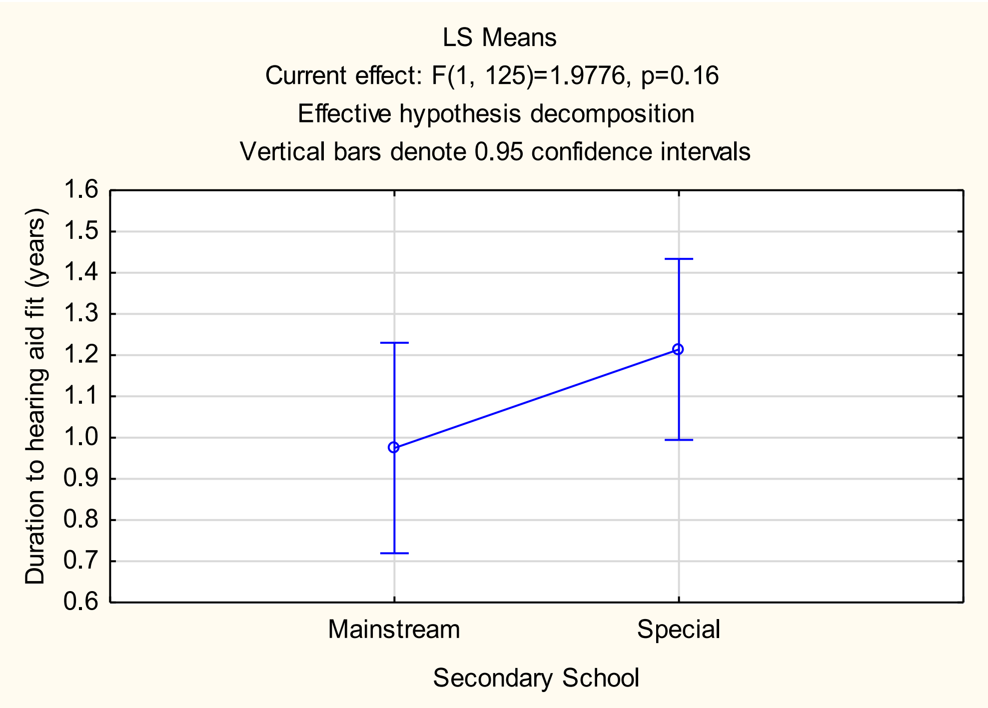

Figure 19. Difference in mainstream and special education groups in terms of the duration of deafness prior to hearing aid fitting at secondary school level

5.12.1.7. Duration of deafness prior to cochlear implantation

Primary School

As seen in Figure 20, the duration of deafness prior to cochlear implantation was 3.6 years (43.2 months) for the recipients in mainstream education and it was 4.69 years (56.28 months) for the recipients in special education. The duration of deafness prior to cochlear implantation was significantly shorter for the recipients in mainstream education ($F(1,204) = 3.99$, $p=0.05$). A shorter duration of hearing loss to implantation was significantly associated with mainstream placement in the primary school.

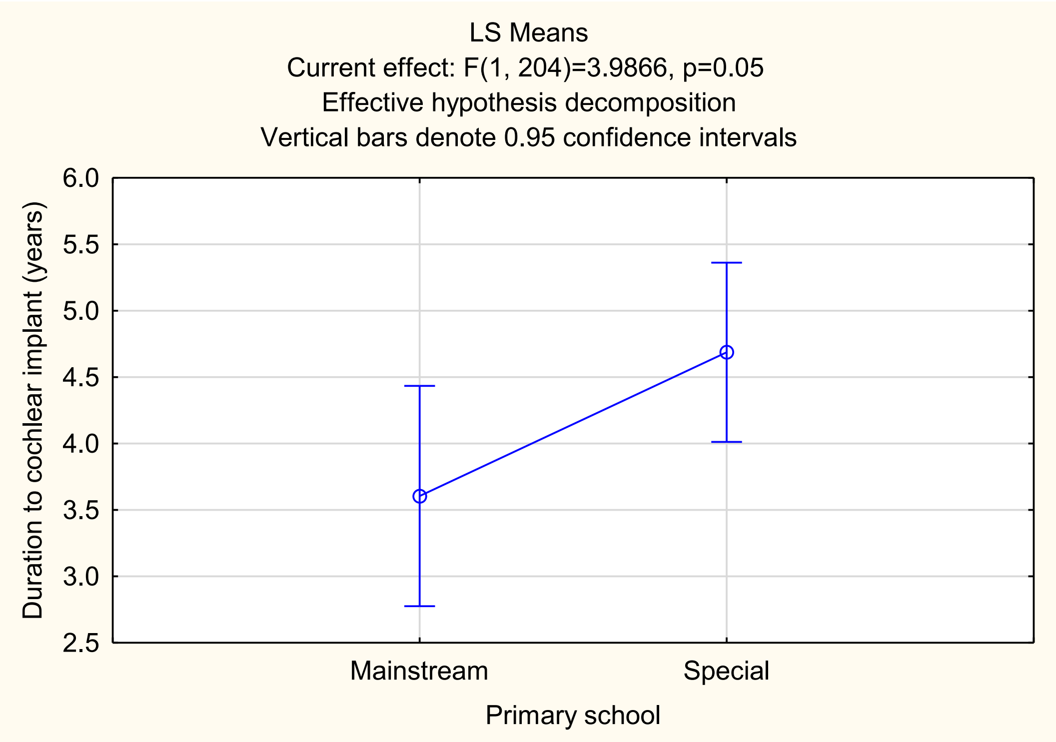

Figure 20. Difference in mainstream and special education groups in terms of the duration of deafness prior to cochlear implantation at primary school level

Secondary School

As seen in Figure 21, the mean duration of deafness prior to cochlear implantation was 4.64 years (55.68 months) for the recipients in mainstream education and it was 5.42 years (65.05 months) for the recipients in special education. Although the duration of deafness prior to implantation was shorter for recipients in mainstream education, this difference was not significant (F (1,128) = 1.00; p=0.32) A shorter duration of deafness to cochlear implantation was therefore not significantly associated with mainstream placement at secondary school level.

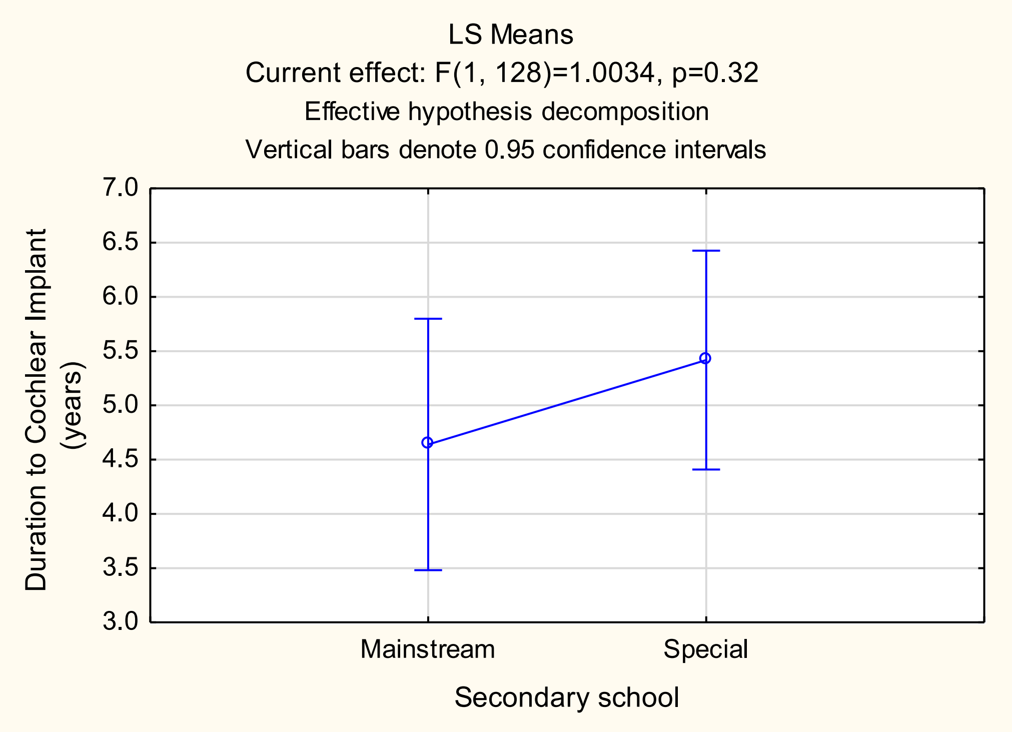

Figure 21. Difference in mainstream and special education groups in terms of duration of deafness prior to cochlear implantation at secondary school level

5.12.1.8. Unilateral or bilateral cochlear implant use

Primary School

As seen in Figure 22, 46 recipients (63%) of bilateral cochlear implant recipients were in mainstream education at primary school level, relative to the 36 unilateral recipients (27%). Bilateral recipients represented a significantly higher percentage of cases in mainstream education (Chi-square (df = 1) = 26.20, p=0.00). Bilateral implant use was significantly associated with mainstream placement in primary school.

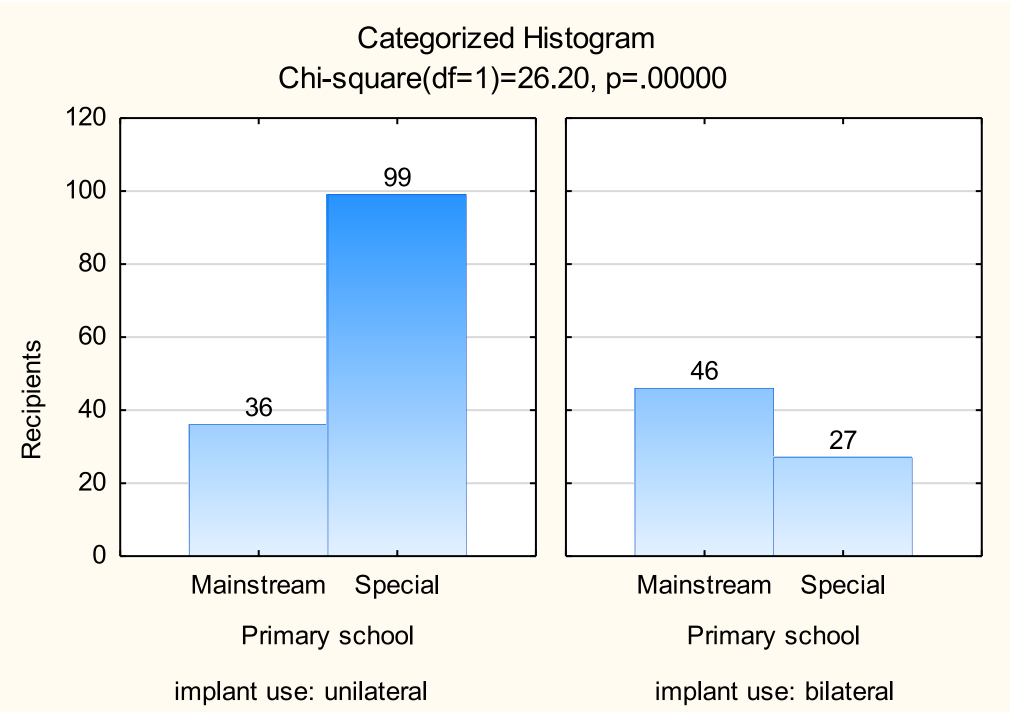

Figure 22. Difference in mainstream and special education groups in terms of bilateral or unilateral implant use in primary school

Secondary School

As seen in Figure 23, 19 bilateral recipients (63%) were in mainstream education relative to the 37 unilateral users (27%).Once again bilateral users represented a significantly higher percentage of cases in mainstream education (Chi-square (df=1)=6.70, p=.01. Bilateral cochlear implant use was therefore significantly associated with mainstream placement at secondary school level as well.

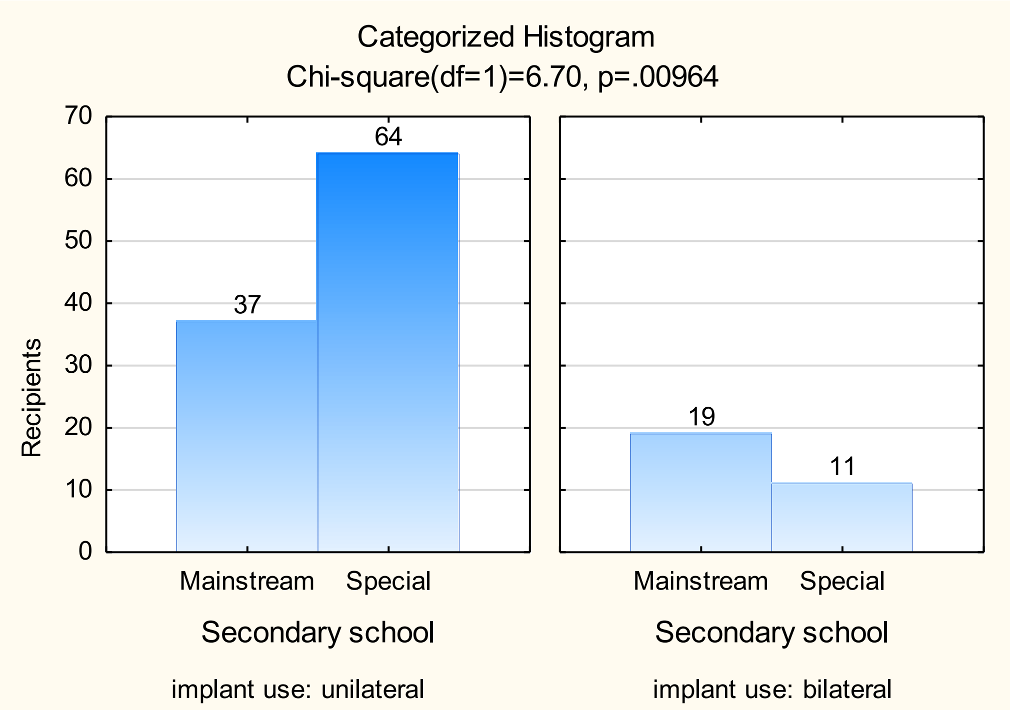

Figure 23. Difference in mainstream and special education groups in terms of bilateral or unilateral implant use in secondary school

5.12.1.9. Speech perception development

Primary School

The mean speech perception category reached at the first three annual intervals after implantation for recipients in mainstream education compared to the recipients in special school education are summarized in Figure 24. At every annual test interval, the mean category achieved by the recipients in mainstream education was significantly higher than that of the recipients in special education. The most significant improvement in mean speech perception category for recipients in mainstream education was observed between year one and year two. On average, the recipients in this group had reached category 7 at the second year test interval and their performance remained stable after this interval. In contrast, the speech perception development for the recipients in special education was on average, more gradual over the three year period.

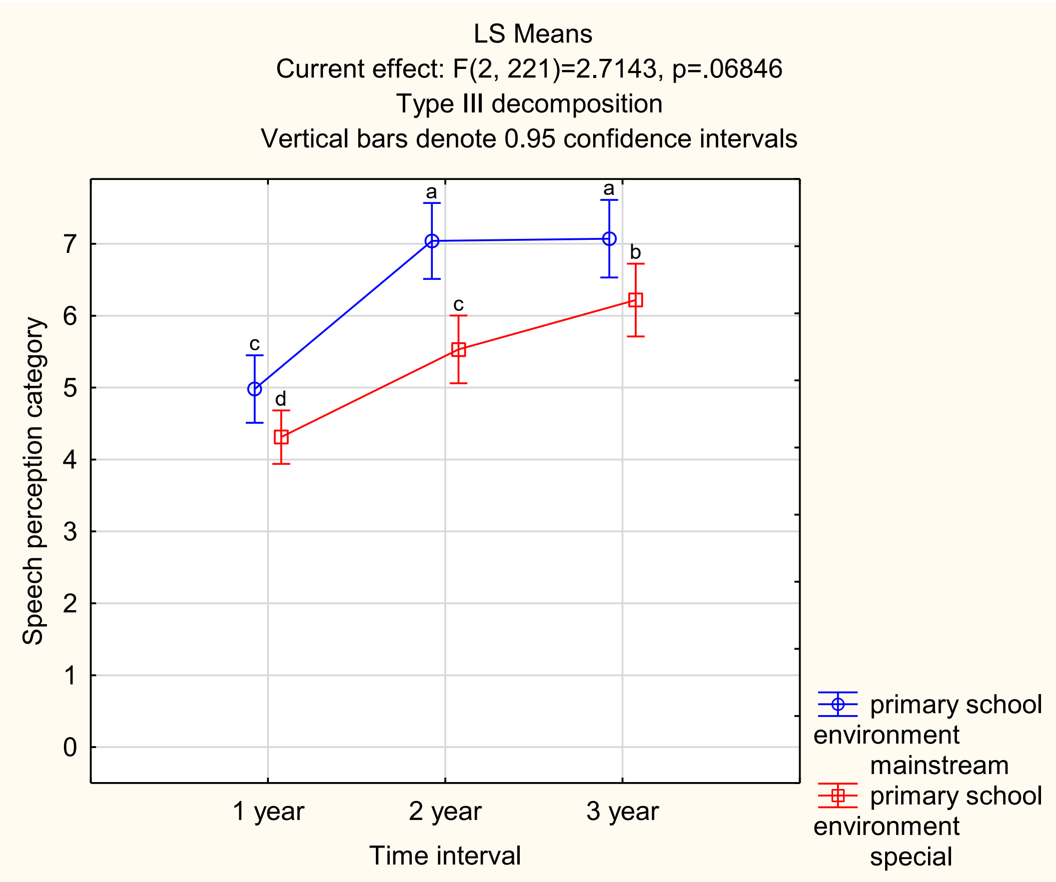

Figure 24. Speech perception development trajectory of recipients in mainstream education compared to recipients in special education at primary school level

Note: The letters in Figure 25 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. In this way, any mean on the graph can be compared to any other mean. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be >0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be <0.05)

Secondary School

The development in mean speech perception category for recipients in mainstream education and recipients in special education for the first three years after implantation are displayed in Figure 25. The mean speech perception category of recipients in mainstream at the first and second year intervals was significantly higher than the average performance of special school recipients. Again, the most significant improvement in speech perception category for mainstream recipients was between year 1 and 2, while the recipients in special education made a more gradual progress over the 3 years.

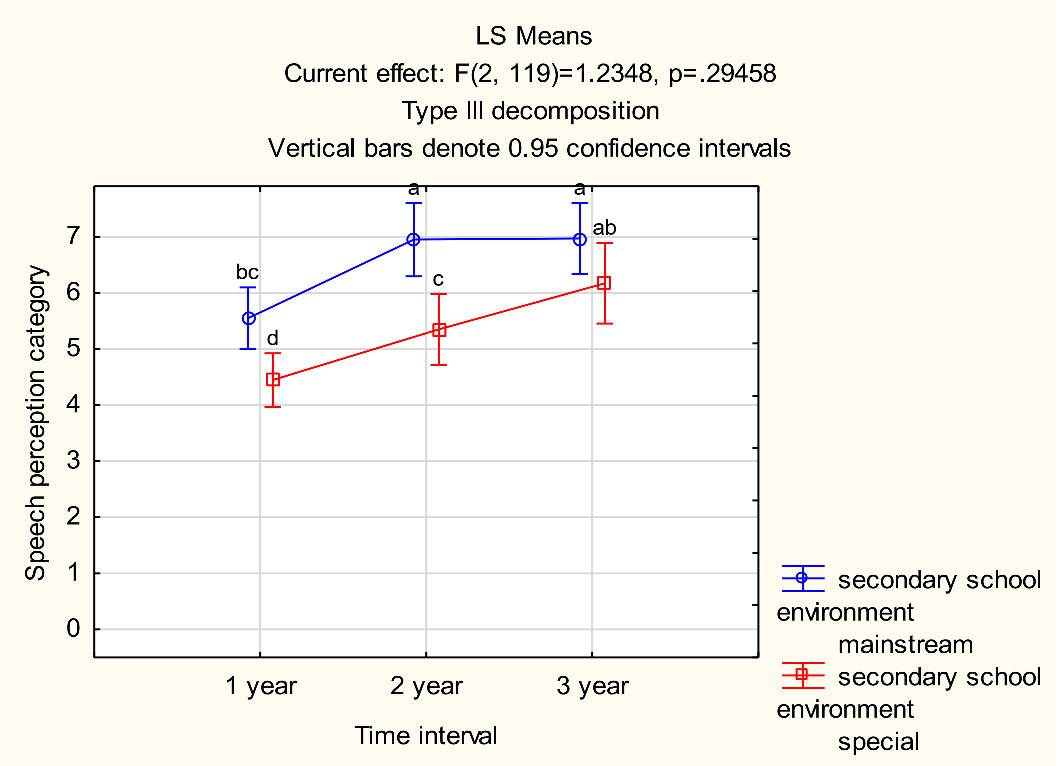

Figure 25 Speech perception development trajectory of recipients in mainstream education compared to recipients in special education.

Note: The letters in Figure 26 represent the results of the post hoc tests where all means were compared pairwise to determine possible significant differences. In this way, any mean on the graph can be compared to any other mean. If the annotations share one letter (e.g. a vs. a), then the corresponding *p*-value comparing the two means will be >0.05. If the annotations share no letters (e.g. a vs. b), then the corresponding *p*-value comparing the two means will be <0.05)

The univariate analysis of variables, indicated that access to private health insurance was significantly associated with mainstream placement at primary school level, but not in secondary school. A higher maternal education level was significantly associated with mainstream placement at primary and secondary school level. The presence of a co-morbidity was negatively associated with mainstream placement in primary and secondary school. The association between age at onset of deafness and mainstream placement in primary and secondary school level was not significant. A shorter duration of hearing loss prior to amplification and cochlear implantation was associated with mainstream placement in primary, but not in secondary school. Bilateral implant use was associated with mainstream placement in primary and secondary school. The most significant improvement in mean speech perception category for participants in mainstream education occurred after two years of implant use.

5.12.2. Logistic regression analysis

During the univariate analysis of variables, associations were determined between potential predictor variables and the dependent variables, mainstream or special school placement. In order to identify variables that differed for recipients in mainstream and special education, independently, in the presence of other variables 2 logistic regressions were conducted.

The first examined for variables that differentiate recipients in mainstream education from recipients in special education at primary school level and the second for variables that differentiate recipients in mainstream education from recipients in special education at secondary school level. Possible predictor variables identified during the univariate analysis of variables, namely, family income, maternal education level, the presence of co-morbidities, bilateral implant use, the duration of hearing loss to amplification and cochlear implantation and the mean speech perception category at the first and second year after implantation were included in the regression. Although age of onset of hearing loss was not associated with educational placement in our study population, it was included in the logistic regressions as well.

The results of the logistic regressions for primary school recipients are summarized in Table 20 and for secondary school recipients in Table 21.

Table 20: Logistic regression model of predictors for mainstream placement in primary school

Variable	**B**	**Wald Stat χ²**	***p***	**Odds Ratio**
Family income classified as private	0.06	0.03	0.85	1.13
Maternal education level	*0.65*	*7.21*	*0.01*	*1.92*
Co-morbidities present	*-1.24*	*11.56*	*0.00*	*12.5* *
Age at onset of deafness	-0.04	0.13	0.72	0.96
Duration of deafness prior to hearing aid fitting	-0.41	2.58	0.11	0.66
Duration of deafness prior to CI	-0.13	2.2	0.13	0.88
Bilateral implant use	0.46	2.8	0.09	2.52
Speech perception category 1 year after CI	-0.01	0.00	0.94	0.99
Speech perception category 2 years after CI	*0.60*	*4.24*	*0.04*	*1.82*

Note. CI=cochlear implant

*Odds with special school placement as reference. All other odds are for mainstream placement as reference.

It was observed that an improvement in maternal education level with one category, as described in section 4.6.2.2, increased the odds of mainstream placement with 1.92 times. The presence of a co-morbidity increased the odds of special school placement with 12.5 times. The speech perception category reached 2 years after implantation was a more significant predictor of educational placement than the result 1 year after implantation. An improvement in speech perception with one category score at 2 years, increased the odds of mainstream placement with 1.82 times.

Table 21: Logistic regression model of predictors for mainstream placement in secondary school

Variable	B	Wald Stat χ^2	*p*	Odds Ratio
Family income classified as private	-0.8	2.37	0.12	0.21
Maternal education level	*1.37*	*7.21*	*0.01*	*3.93*
Co-morbidities present	*-2.06*	*12.81*	*0.00*	*50**
Age at onset of deafness	0.03	0.07	0.8	1.03
Duration of deafness to hearing aid fitting	-0.16	0.17	0.68	0.87
Duration of deafness to cochlear implantation	-0.01	0.01	0.92	0.99
Bilateral cochlear implant use	0.72	2.01	0.16	4.24
Speech perception category 1 year after CI	-0.27	1.58	0.21	0.77
Speech perception category 2 years after CI	2.08	7.78	0.01	8.04

Note. CI=cochlear implant

The same 3 independent variables were associated with mainstream placement in secondary school as was observed at primary school level. An increment in maternal education level with one category, increased the odds of mainstream placement with 3.93 times. Once again, the presence of a comorbidity was negatively associated with mainstream placement. The presence of a co-morbidity increased the odds of special school placement with 50 times. An improvement in speech perception with one category score at the second year test interval increased the odds of mainstream education with 8.04 times.

Summary and discussion

The educational placement of recipients and variables associated with mainstream educational placement were documented in sections 5.9 and 5.12.

In summary, it was found that less than half of the recipients in basic education, were placed in mainstream schools. Results from previous studies varied in terms of the percentage of learners being mainstreamed: 32% (Beadle et al., 2005); 36% (Illg et al., 2017); 52% (Le Roux et al., 2016);

58% (Uziel et al., 2007). Although the majority of recipients were not able to attend mainstream schools, a significant percentage of recipients in primary (91.3%) and secondary school (77%) were able to follow a mainstream curriculum in a special school setting due to access to individualized and rehabilitative support. This highlights the level of educational support that many cochlear implant recipients may require and the importance that appropriate educational placement should be accessible to a child before cochlear implantation should be considered (Müller & Wagenfeld, 2003). This consideration is particularly important in a low resource country like South Africa (Müller & Wagenfeld, 2003). The specialized support that these learners may require, should be a consideration in the long-term goal of the Department of Education to develop an inclusive education system for learners with special needs (Department of Education, 2001).

The absence of a co-morbidity, maternal education level and the speech perception ability after two years of implant use were the only independent variables associated with mainstream educational placement at primary and secondary school level in the study cohort. The association between these variables and outcomes of pediatric cochlear implantation has been documented in the literature. Birman et al. (2012), Boons et al. (2012) and Ching et al. (2018) reported a negative relationship between the presence of co-morbidities and language development. Le Roux et al. (2016) documented that recipients with co-morbidities were less likely to attend mainstream education. Ching et al. (2018) reported that a higher maternal education level will result in improved language outcomes after pediatric cochlear implantation and Daya, Ashley, Gysin and Papsin (2000) documented that children in mainstream education demonstrated better speech perception.

Although the benefit of home language education for educational development is acknowledged in the hearing population (Prinsloo, 2007), home language education was not associated with educational placement in the study cohort. As discussed in section 5.1, only a 9% of recipients in the study cohort did not receive home language education. It is therefore possible that an association could not have been established, due to this small number.

Although bilateral cochlear implantation is associated with improved outcomes in language development (Boons et al., 2002; Sarant et al., 2014), it was not identified as an independent variable for mainstream placement in the present study. As documented in section 5.5, the majority of the bilateral recipients in the study received the second implant during a sequential procedure. Sarant et al. (2014) demonstrated that a shorter duration of time between the first and second implant is associated with improved language outcomes. The duration between the first and second implant was not investigated in the present study. .It is therefore possible that a possible association between bilateral implant use and mainstream placement could not be established, due to this limitation.

It was a very unexpected finding that age at onset of deafness, duration of deafness prior to amplification and duration of deafness prior to implantation were not identified as independent variables associated with mainstream placement. The evidence from the literature is undisputable that early cochlear implantation is associated with improved language outcomes (Arterières et al., 2009; Boons et al., 2012; Ching et al., 2018; Niparko et al., 2010; Tobey et al., 2013) and mainstream educational placement (Semenov et al., 2013). It is possible that the following two methodological limitations in the study contributed to this unexpected finding:

First, the pre-operative residual hearing of the recipients was not included in the bivariate and logistic regression analysis. As documented in section 5.4.2, variation in the pre-operative residual hearing for the recipients with SNHL was observed, indicating that some recipients in the study sample had a pre-operative hearing loss that fell in the severe range. As the pre-operative pure tone average for only 69% of the recipients with a prelingual onset of deafness was available, this variable was not included in the bivariate analysis and logistic regression analysis. It could be argued that onset of deafness and duration of deafness to intervention may have a different association with educational placement, when recipients had more residual hearing.

Second, although the age at amplification and cochlear implantation was associated with educational placement at primary school level, it was not identified as independent variables. The inclusion of children with perilingual and postlingual onset of deafness in the study cohort, most likely influenced this result.

5.13 General discussion and summary of findings

In sections 5.1 to 5.12 of this chapter the demographic, socio-economic and pre-operative audiological characteristics of a cohort of pediatric cochlear implant recipients, the long-term post-operative outcomes in the domains of implant use, mode of communication, speech perception development, educational placement, occupational status and the variables associated with mainstream educational placement were reported and discussed.

Firstly, the ethnic distribution of the recipients in this study population was not representative of the ethnic distribution in the Western Cape Province. The black population was the most under represented ethnic group. In terms of family income and maternal education level, the socio-economic status of the majority of the recipients was higher than what is documented to be the provincial or national average (Blecher et al., 2011; Statistics South Africa, 2011). It was clear that cochlear implantation was more accessible for recipients from the private health sector in South Africa.

For nearly 50% of the recipients in the dataset, the aetiology of deafness was unknown and 22% presented with an additional co-morbidity. The majority of the recipients suffered from a congenital onset of SNHL. On average the children with a SNHL suffered from a bilateral profound degree of hearing loss. The majority of the recipients in the dataset used bilateral hearing aids prior to cochlear implantation, but could not perform open set speech perception tasks or demonstrated limited open-set speech perception development with acoustic amplification. On average the age at hearing aid fitting and cochlear implantation for recipients with a prelingual onset of SNHL or ANSD exceeded the optimal timeframes for intervention (Dettman et al., 2007; Dettman et al., 2016; Joint Committee on Infant Hearing, 2007)

This long-term retrospective record review shows that all the recipients who had received a cochlear implant system during childhood, used their devices long-term. The majority of the recipients developed open set speech perception and were able to use oral language as mode of communication. A more gradual speech perception development trajectory was observed for some recipients with a prelingual and perilingual onset of deafness.

The recipients attended a variety of educational settings during basic education. The majority of the recipients required some form of special education, but were able to follow a mainstream curriculum. More than half of the recipients who had completed basic education attended or completed higher education.

The majority (73%) of recipients seeking employment were employed .The recipients were employed in a range of occupations. Although variation in the data was observed, on average their occupations required training at a NQF lever of 4 or lower.

Maternal education level, the absence of a co-morbidity and speech perception ability two years after implantation, were identified as independent variables associated with mainstream educational placement at primary and secondary school level.

5.14. Contribution: Clinical implications of the study for service delivery in South Africa

The description of the demographic and socio-economic characteristics of the recipients in the study cohort elucidated the inequity of cochlear implant provision in the Western Cape Province. This highlights the continuous responsibility of professionals in the field to inform parents, referrers, funders and the Department of Health about cochlear implantation as an intervention option and the evidence behind the expected outcome for carefully selected candidates with severe to profound SNHL or ANSD. In close liaison with cochlear implant manufacturers a continuous effort should be made to improve the affordability of cochlear implant systems in low resource settings.

Cochlear implant systems are expensive devices but it was observed that recipients who were implanted in childhood continued to use their devices. The majority of recipients in this cohort suffered from a profound degree of hearing loss and prior to cochlear implantation proficiency in spoken language development would not have been a realistic expectation for these children (O’Donoghue & Pisoni, 2014).The results of the study revealed that it was possible for the majority of the children in the study cohort to develop open set speech perception and use oral language as the mode for communication.

The majority of recipients in the study cohort were able to follow a mainstream education curriculum in a special education environment. This highlights the level of educational support that many cochlear implant recipients may require and the importance that appropriate educational placement should be accessible to a child before cochlear implantation should be considered (Müller & Wagenfeld, 2003).

On average the age at hearing aid fitting and cochlear implantation for recipients with a prelingual onset of SNHL or ANSD exceeded the optimal timeframes for intervention (Dettman et al., 2007; Dettman et al., 2016; Joint Committee on Infant Hearing, 2007). As discussed in section 2.9, the age at implantation in the child with congenital deafness is a critically important parameter with high prognostic value for clinical outcome and earlier cochlear implantation will improve the cost effectiveness of the intervention (Semenov et al., 2013). The present study highlight the need for earlier identification of childhood hearing loss in South Africa, by means of improved newborn hearing screening services (NHS) in both the public and private health sectors (Meyer et al., 2012; Theunisen & Swanepoel, 2008). It is evident that the outcome and potentially the cost effectiveness of pediatric cochlear implantation in South Africa will have to be evaluated in context of the availability of NHS.

5.15. Limitations of the study and recommendations for future research

5.15.1. Limitations

In order to establish the long-term trends in outcome after pediatric cochlear implantation, a retrospective record review of children who received cochlear implants over a period of 24 years and 10 months was performed. The chosen study method and study sample introduced limitations:

The first limitation of the study is inherent to retrospective record reviews. As discussed in section 4.1, the data in patient records were not originally recorded for research purposes and it is therefore not possible to prove the accuracy of information recorded in the patient records. Further, miscoding or misinterpretation of data may have occurred during the abstraction process. Data stored in case history forms, medical reports and audiograms were accessible. However, information regarding school placement and occupational status and employment were not captured in a consistent manner and had to be obtained from the managing audiologist during personal interviews. A more standardized manner of recording this information has subsequently been included in patient recordkeeping.

The second limitation related to the data abstraction process. All the records were reviewed by a single abstractor, who was not blind to the aims of the study and may have introduced researcher-induced biases.

The third limitation related to the study sample selection. As discussed in section 4.3.3 recipients were implanted over a very long period of 24 years and 10 months. During this period the selection criteria for cochlear implant recipients (Zwolan & Sorkin, 2016) the average age at intervention and cochlear implant technology (Birman et al., 2012) have changed considerably. The study cohort were therefore not homogenous. Furthermore, the inclusion of recipients with a prelingual, perilingual and postlingual hearing loss obscured the prognostic indicators for mainstream educational placement.

5.15.2. Recommendations for future research

As discussed in the previous section, the study population was not homogenous in terms of the age at onset of deafness. It is advised that future studies should confine the study sample to prelingually deafened children.

As discussed in section 2.8.2, the language ability of pediatric cochlear implant recipients will be an important prognostic indicator for educational placement (Kral & O'Donoghue, 2010).The present study did not report on this variable and it is advised that language development after pediatric cochlear implantation should be included in future studies.

As discussed in section 2.7, funding for cochlear implants is not easily available to candidates in the public health sector in South Africa. The TH-SU-CIU is however managing an increasing number of recipients funded by Tygerberg Hospital. The outcomes of this sub-group should be monitored and shared with Department of Health.

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Chapter 6. Conclusion

As discussed in chapters 1 and 2, the outcome of pediatric cochlear implantation is multi-faceted, is likely to develop over many years of a person's life and may be influenced by a multitude of factors. The parents of deaf children often choose cochlear implantation because they want their children to develop spoken language and experience the consequent educational and occupational implications (Fink et al., 2007; O'Donoghue & Pisoni, 2014). Great variability however exists in the outcomes achieved by pediatric cochlear implant recipients (Boons et al., 2012). Cochlear implantation is an elective procedure and the expected outcomes should be available to parents of candidates and the funders of these systems to enable them to make informed decisions. Although unilateral pediatric cochlear implantation is proven to be cost effective in high income countries (Cheng et al., 2000; Barton et al., 2006c), cochlear implant systems are very expensive devices (Barton et al., 2006b) and therefore not easily available in low and middle income countries (Magro et al., 2018). Positive cost-analysis reports of unilateral cochlear implantation in the Global North conclude that the expected financial benefits associated with spoken language development, such as mainstream educational placement and the increased earning potential of the individual (Cheng et al., 2000) will contribute to the cost effectiveness of this intervention option. An understanding of the prognostic factors that will contribute to the outcome of pediatric cochlear implantation and potentially the cost effectiveness is therefore vital in the selection of suitable candidates (Black et al., 2014), especially in low resource settings (Magro et al., 2018).

In the review of the data in pediatric cochlear implantation records, recorded over a period of 27 years and 11 months, it was established that recipients who received cochlear implant systems during childhood continued to use their devices. The majority of recipients developed open set speech perception three years after implantation and were able to communicate via spoken language. Although the majority of recipients at primary and secondary school level attended special education, they were able to access a mainstream curriculum with the necessary educational support. The majority of recipients seeking employment after secondary school or higher education were employed. In this study, prognostic factors positively associated with mainstream educational placement were: a higher maternal education level, the absence of a co-morbidity and the speech perception category reached two years after cochlear implantation. It was however possible that the inclusion of recipients with a prelingual, perilingual and postlingual onset of hearing loss in the study cohort influenced the result and obscured other potential prognostic indicators. Future outcome studies should focus on more homogenous study groups.

The average age at intervention in the study cohort, highlights the need for improvement of NHS services in the public and state health sector in South Africa (Meyer et al., 2012; Theunisen & Swanepoel, 2008). The ethnic and socio-economic distribution of recipients elucidated the inequity in cochlear implant provision in the Western Cape Province. This highlights the continuous responsibility of professionals in the field to address the inequality and provide evidence for the expected outcome for carefully selected cases to the Department of Health. In close liaison with cochlear implant manufacturers a continuous effort should be made to improve the affordability of cochlear implant systems in low resource settings.

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Appendices

Appendix A:Tygerberg Hospital Income Classification System (April 2019)

Category	Criteria
H0	No income or grant only
H1	Family annual income: less than R100 000
H2	Family annual income: equal or more than R100 0000, but less than R350 000
H3	Family annual income: equal to or more than R350 000
P	Belonged to a medical aid scheme

Appendix B: Implant systems used (Cochlear ®, 2017).

(Cochlear ®, 2017).

Type of implant	Year introduced
Cochlear Nucleus Profile with Contour Advance Electrode: CI512	2014
Cochlear Nucleus CI422 Cochlear Implant: CI422	2011
Nucleus Freedom with Straight Electrode: CI24RE(CA)	2005
Nucleus Freedom with Straight Electrode: CI24RE(S)	2005
Nucleus 24 with Contour Advance Electrode: CI24R(CA)	2002
Nucleus 24 with Contour Electrode: CI24R(CS)	2000
Nucleus 24k with Straight Electrode: CI24R ST)	2000
Nucleus 24 with Double Array	1999
Nucleus 24 with Straight Electrode: CI24M	1997
Nucleus 22: CI22M	1986

Appendix C: South African Standard Classification of Occupations (SASCO)

(Statistics South Africa, 2012)

Skill level	Major group	Description
3+4	1	Managers • Chief executives, senior officials and legislators • Administrative commercial managers • Production and specialized services managers • Hospitality, retail and other services managers
4	2	Professionals • Science and engineering professionals • Health professionals • Teaching professionals • Business and administration professionals • Information and communications technology professionals
3	3	Technicians and associate professionals • Science and engineering associate professionals • Health associate professionals • Business and administration professionals • Legal, social, cultural and related associate professionals • Information and communication technicians
2	4	Technicians and associate professionals • Science and engineering associate professionals • Health associate professionals • Business and administration professionals • Legal, social, cultural and related associate professionals • Information and communication technicians
2	5	Service- and sales workers and armed forces • Personal service workers • Sales workers • Personal care workers • Protective service workers and armed forces occupations
2	6	Skilled agricultural, forestry and fishery workers • Market-oriented skilled agricultural workers • Market-oriented skilled forestry, fishery and hunting workers • Subsistence farmers, fishers, hunters and gatherers
2	7	Craft and related trades workers • Building and related trades workers, excluding electricians • Metal, machinery and related trades workers • Handicraft and printing workers • Electrical and electronic trades workers

		• Food processing, wood working, garment and other craft and related trades workers
2	8	Plant and machine operators and assemblers • Stationary plant and machine operations • Assemblers • Drivers and mobile plant operators
1	9	Elementary occupations • Cleaners and helpers • Agricultural, forestry and fishery labourers • Labourers in mining, construction, manufacturing and transport • Food preparation assistants Elementary occupations continue • Street and related sales and service workers • Refuse workers and other elementary workers • Underground economy and related activities

Appendix: D

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Approved with Stipulations

New Application

26-Nov-2013
Lombaard, Suryn S

Ethics Reference #: S13/10/221

Title: **Review of cochlear implantation records at the Tygerberg Hospital University of Stellenbosch cochlear implant unit since 1986.**

DearMs Suryn Lombaard,

The **New Application** received on **30-Oct-2013**, was reviewed by members of **Health Research Ethics Committee 2** via Minimal Risk Review procedures on **11-Nov-2013**.

Please note the following information about your approved research protocol:

Protocol Approval Period: **26-Nov-2013 -26-Nov-2014**

The Stipulations of your ethics approval are as follows:

1. Waiver of consent granted as retrospective data will be used only.

Please remember to use your **protocol number** (**S13/10/221**) on any documents or correspondence with the HREC concerning your research protocol.

Please note that the HREC has the prerogative and authority to ask further questions, seek additional information, require further modifications, or monitor the conduct of your research and the consent process.

After Ethical Review:
Please note a template of the progress report is obtainable on www.sun.ac.za/rds and should be submitted to the Committee before the year has expired. The Committee will then consider the continuation of the project for a further year (if necessary). Annually a number of projects may be selected randomly for an external audit.
Translation of the consent document to the language applicable to the study participants should be submitted.

Federal Wide Assurance Number: 00001372
Institutional Review Board (IRB) Number: IRB0005239

The Health Research Ethics Committee complies with the SA National Health Act No.61 2003 as it pertains to health research and the United States Code of Federal Regulations Title 45 Part 46. This committee abides by the ethical norms and principles for research, established by the Declaration of Helsinki, the South African Medical Research Council Guidelines as well as the Guidelines for Ethical Research: Principles Structures and Processes 2004 (Department of Health).

Provincial and City of Cape Town Approval

Please note that for research at a primary or secondary healthcare facility permission must still be obtained from the relevant authorities (Western Cape Department of Health and/or City Health) to conduct the research as stated in the protocol. Contact persons are Ms Claudette Abrahams at Western Cape Department of Health (healthres@pgwc.gov.za Tel: +27 21 483 9907) and Dr Helene Visser at City Health (Helene.Visser@capetown.gov.za Tel: +27 21 400 3981). Research that will be conducted at any tertiary academic institution requires approval from the relevant hospital manager. Ethics approval is required BEFORE approval can be obtained from these health authorities.

We wish you the best as you conduct your research.
For standard HREC forms and documents please visit: www.sun.ac.za/rds

If you have any questions or need further assistance, please contact the HREC office at 0219389207.

Appendix: E

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Progress Report Approval Letter

21/08/2018

Project ID: 7372

Ethics Reference #: S13/10/221

Title: Review of cochlear implantation records at the Tygerberg Hospital University of Stellenbosch cochlear implant unit since 1986

Dear Miss Suryn Lombaard,

Your request for extension/annual renewal of ethics approval dated 28/05/2018 17:37 refers.

The Health Research Ethics Committee reviewed and approved the annual progress report you submitted through an expedited review process.

The approval of this project is extended for a further year.

Approval date: 21 August 2018

Expiry date: 20 August 2019

Kindly be reminded to submit progress reports two (2) months before expiry date.

Where to submit any documentation

Kindly note that the HREC uses an electronic ethics review management system, *Infonetica*, to manage ethics applications and ethics review process. To submit any documentation to HREC, please click on the following link: https://applyethics.sun.ac.za.

Please remember to use your **Project ID** [7372] and Ethics Reference Number [S13/10/221] on any documents or correspondence with the HREC concerning your research protocol.

National Health Research Ethics Council (NHREC) Registration Numbers: REC-130408-012 for HREC1 and REC-230208-010 for HREC2

Federal Wide Assurance Number: 00001372

Institutional Review Board (IRB) Number: IRB0005240 for HREC1

Institutional Review Board (IRB) Number: IRB0005239 for HREC2

The Health Research Ethics Committee complies with the SA National Health Act No. 61 of 2003 as it pertains to health research and the United States Code of Federal Regulations Title 45 Part 46. This committee abides by the ethical norms and principles for research, established by the Declaration of Helsinki and the South African Medical Research Council Guidelines as well as the Guidelines for Ethical Research: Principles, Structures and Processes 2015 (Department of Health).

Yours sincerely,

Francis Masiye,

HREC Coordinator,

Health Research Ethics Committee 2 (HREC2).