HEALTH-RELATED QUALITY OF LIFE IN SOUTH-AFRICAN CHILDREN WHO USE COCHLEAR IMPLANTS

by

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Table of Contents

LIST OF TABLES

LIST OF FIGURES

LIST OF ABBREVIATIONS

CI: Cochlear implant

HRQoL: Health-related quality of life

PCIU: Pretoria Cochlear Implant Unit

DCIP: Durban Cochlear Implant Program

JCIC: Johannesburg Cochlear Implant Centre

CCIPP: Children with cochlear implants: Parental perspectives

FORMATTING

APA referencing style was utilized in this dissertation

RESEARCH OUTPUT

Parts of this dissertation have been presented at the following international conferences:

1. **Brewis, B;** le Roux, T; Schlemmer, K; Nauta, L & Vinck, B. (2018). Health-related quality of life in South-African children who use cochlear implants. Paper presented at the *34th World Congress of Audiology, Cape Town, South Africa* (28 - 31 October 2018).

2. **Brewis, B;** le Roux, T; Schlemmer, K; Nauta, L & Vinck, B. (2018). Health-related quality of life in South-African children who use cochlear implants. Poster presented at the *4th World Congress on Cochlear Implants in Emerging Countries, Cape Town, South Africa* (31 October – 2 November 2018).

ABSTRACT

The aim of this study was to describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South African cohort from the perspectives of parents and to relate parental ratings of HRQoL to a range of demographic variables.

Parents of school-going CI recipients completed the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) CI specific HRQoL questionnaire. The effect of different demographic variables on HRQoL outcomes was also determined. The study sample included 54 parents of school-going (mean age = 12.2 years; 3.6 SD; range = 6.6-18.3 years) CI recipients with at least six months CI experience.

Children's communication and general functioning with a CI received the most positive parental ratings. A number of statistically significant associations between HRQoL outcomes and demographic variables have been confirmed. A mainstream educational setting was associated with better HRQoL outcomes in terms of communication and education, while pre-lingual onset of deafness was associated with better HRQoL outcomes in terms of general functioning and well-being. Shorter duration of deafness and unilateral implantation were associated with higher parental ratings for self-reliance and well-being respectively. Longer duration of CI use was linked to better ratings for general functioning, while shorter duration of CI use was linked to improved ratings for effects of implantation.

Parents assigned positive ratings to their child's HRQoL. This exploration of children's HRQoL related to their CIs contributes to evidence-based pediatric CI services that support optimal psychosocial outcomes.

Keywords: cochlear implant, health-related quality of life, outcomes, parents, pediatric cochlear implantation.

1. INTRODUCTION

Cochlear implantation has progressed over the past years as a successful treatment option for children with severe to profound sensorineural hearing loss (Lin & Niparko, 2006). Reports indicate that some prelingually deafened children who are implanted within the first year of life can achieve speech and language skills comparable to their normal hearing peers (Ching et al., 2009; Niparko et al., 2010; Wie, 2010). The mounting effects that cochlear implantation has on a child's speech production, speech perception and language development extends beyond these measures of improvement (Warner-Czyz, Loy, Tobey, Nakonezny, & Roland, 2011). Cochlear implantation has a larger positive effect on the psychological well-being of a child, and not only on communication (Sack & Whynes, 2005). The World Health Organization (1998) defined quality of life as an individual's perception of his/ her position in life, affected in a multifaceted way by the psychological state, level of independence, social relationships, personal beliefs, and physical health. In recognizing the need to measure the benefits and limitations of medical interventions such as cochlear implantation on social, emotional and physical well-being, the term health-related quality of life (HRQoL) has been defined (Loeffler et al., 2010). HRQoL is concerned with the effects that a disability or medical condition has on daily functioning (Palermo et al., 2008). This general health status of an individual has been recognized as an all-inclusive measure of medical intervention outcomes (Mo, Lindbæk, & Harris, 2005; Streufert, 2008).

1.1 Health-related quality of life as outcome measure in pediatric cochlear implantation

In order to ensure positive outcomes for pediatric cochlear implant (CI) recipients, the impact of permanent hearing loss and consequent treatment on personal well-being should be assessed through HRQoL measurements (Meserole et al., 2014). Additional to speech, language, and educational outcomes, measuring HRQoL in CI recipients have become an important outcome measure to determine device efficacy, however, these measures do not necessarily represent the CI`s effect on everyday functioning (Clark et al., 2012; Meserole et al., 2014). These traditional measures of auditory and communication outcomes are necessary but do not capture the cascading effects of deafness on a child's emotional, social, and behavioural functioning (Hoffman, Cejas, & Quittner, 2018). HRQoL measures should, therefore, identify and quantify the specific areas of general well-being that are presumably influenced by CI use, in order for families and involved professionals to be able to support implanted children in their areas of concern (Kumar, Warner-Czyz, Silver, Loy, & Tobey, 2015).

HRQoL in pediatric CI recipients can be measured by either generic or condition-specific measures. Generic HRQoL measures focus on information about health independent of a medical condition, while condition-specific measures focus on health aspects related to a particular condition, intervention or population (Warner-Czyz et al., 2011). Generic HRQoL measures assess domains that can be compared to children with normal hearing, but these domains do not necessarily focus on issues specific to cochlear implantation such as restrictiveness, comfort, and

satisfaction that relate to the daily impact of a CI device (Kumar et al., 2015; Nicholas & Geers, 2003; Warner-Czyz et al., 2011). Various studies have assessed HRQoL in pediatric CI recipients by focusing only on generic aspects such as physical, psychological, and social well-being (Barton, Fortnum, Stacey, & Summerfield, 2006; Huber, 2005; Sach & Barton, 2007; Sack & Whynes, 2005; Warner-Czyz, Loy, Roland, Tong, & Tobey, 2009; Warner-Czyz et al., 2011). Condition-specific measures focus on and are more sensitive to positive or negative consequences of a particular illness, condition or intervention (Bjornson & McLaughlin, 2001; Wiebe, Guyatt, Weaver, Matijevic, & Sidwell, 2003). Using condition-specific measures provide valuable information as CI recipients have unique experiences in terms of being deaf and using rehabilitative hearing devices.

Generally, for pediatric CI recipients, HRQoL measures are in questionnaire format and are completed by parents/ primary caregivers. Parents/ primary caregivers are not always able to report on unobservable areas of their child's functioning such as their self-esteem and emotional distress (Meserole et al., 2014; Upton, Lawford, & Eiser, 2008). In spite of the limitations, parent-reported questionnaires are valuable as they provide very necessary information about implanted children's functioning, their additional intervention needs and the limitations that they experience as a result of their CI(s) (Damen, Krabbe, Archbold, & Mylanus, 2007). It has been reported that children with severe to profound sensorineural hearing loss' self-reported HRQoL differ from that of what their parents reported (Meyer et al., 2013). This incompatibility may be relevant due to the fact that 90% of children with hearing loss are born to normal-hearing parents (Mitchell & Karchmer, 2004). Similarly, Warner-

Czyz et al. (2009) also indicated that children using cochlear implants rated their overall HRQoL significantly more positively than their parents.

Self-reported measures allow children to rate their own personal experiences and it has been suggested that children as young as seven years are able to describe themselves reliably (Feeny, Juniper, Ferrie, Griffith, & Guyatt, 1998; Herjanic, Herjanic, Brown, & Wheatt, 1975). However, this may not necessarily be the case for all pediatric CI recipients. As a result of immature communication skills, many pediatric CI recipients may not have the required abilities to express their feelings or describe experiences (Hays et al., 2006). Responses from parents can be used in such cases to describe HRQoL since parents are considered as the closest proxy to a child. Several studies have assessed HRQoL outcomes in pediatric CI recipients by using parent-reported questionnaires (Archbold, Lutman, Gregory, O'Neill, & Nikolopoulos, 2002; Archbold, Sach, O'Neill, Lutman, & Gregory, 2008; Damen et al., 2007; Fortunato-Tavares, Befi-Lopes, Bento, & de Andrade, 2012; Incesulu, Vural, & Erkam, 2003; O'Neill, Lutman, Archbold, Gregory, & Nikolopoulos, 2004).

1.2 Health-related quality of life outcomes in pediatric cochlear implant recipients

Previous research has determined if associations between HRQoL and demographic variables exist. Significant improvement in HRQoL has been documented for pediatric CI recipients after CI surgery (Barton et al., 2006; Beadle, Shores, & Wood, 2000; Sach & Barton, 2007). A variety of generic and CI specific HRQoL domains have been positively reported in the literature across HRQoL measures. Literature

report positive ratings for communication (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Loy, Warner-Czyz, Tong, Tobey, & Roland, 2010; Sack & Whynes, 2005; Warner-Czyz et al., 2009, 2011; Zhang et al., 2016). General functioning received positive ratings (Archbold et al., 2002; Schorr, Roth, & Fox, 2009); together with physical and psychological well-being (Huber, 2005; Loy et al., 2010; Warner-Czyz et al., 2009; Zhao et al., 2018), social relations (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Sack & Whynes, 2005; Zhao et al., 2018) and self-reliance (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Sack & Whynes, 2005; Zhao et al., 2018). However, literature reports less positive ratings for HRQoL domains that relate to education (Kumar et al., 2015; Soleimanifar, Jafari, & Zarandy, 2015), and the two family-related sub-domains, the effects of the CI on the recipient (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Kumar et al., 2015; Sack & Whynes, 2005) and the support required by the implanted child (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Sack & Whynes, 2005). Parental reports also confirmed the concern for device failure (Huttunen et al., 2009; Incesulu et al., 2003), as well as impatience as progress in HRQoL takes time (Archbold et al., 2008; Huttunen et al., 2009).

1.3 Influence of demographic variables on health-related quality of life outcomes in pediatric CI recipients

Various demographic factors have been identified to influence HRQoL ratings in pediatric CI recipients. An understanding of the factors that will have an opposing effect on CI outcomes will enable parents/ primary caregivers to set realistic expectations for their children (Black, Hickson, Black, & Perry, 2011; Kral &

O'Donoghue, 2010; le Roux et al., 2016). More favorable generic HRQoL outcomes in pediatric CI recipients are associated with female gender (Barton et al., 2006; Sach & Barton, 2007), higher levels of parental education (Barton et al., 2006; Sach & Barton, 2007; Spencer, Tomblin, & Gantz, 2012), onset of hearing loss at an older age and the absence of additional developmental conditions (Barton et al., 2006; Sach & Barton, 2007). In a number of studies on HRQoL outcomes in pediatric CI recipients, longer duration of CI use has been associated with more positive HRQoL ratings (Barton et al., 2006; Loy et al., 2010; Sach & Barton, 2007; Schorr, Roth, & Fox, 2009; Spencer, Tomblin, & Gantz, 2012; Yorgun et al., 2015). An association between younger age at implantation and better HRQoL scores was shown in various studies (Barton et al., 2006; Sach & Barton, 2007; Warner-Czyz et al., 2009, 2011), whereas Kumar et al., (2015) and Wong et al., (2017) could not confirm this association. Younger chronological age and lower levels of family stress are also associated with more positive ratings of generic and CI specific HRQoL (Meserole et al., 2014; Sach & Barton, 2007; Warner-Czyz et al., 2009, 2011; Zhang et al., 2016).

1.4 Problem-statement and rationale

Globally the number of pediatric CI surgeries is steadily increasing due to the expansion of selection criteria, including those children with less severe degrees of hearing loss as well as those with additional medical conditions (Black et al., 2011; Fitzpatrick et al., 2009). The measurement of HRQoL outcomes has thus become a fundamental component in pediatric cochlear implantation since it contributes to evidence-based services that eventually endorse optimal outcomes. Despite this recent focus to assess the broader personal impact of permanent hearing loss and

cochlear implantation in children, further investigation is needed to explore the possible influence of different interacting factors on HRQoL outcomes.

Infant hearing loss is one of the most frequently occurring birth disorders, with an estimate of 17 babies born with a hearing loss in South Africa every day (Swanepoel, Störbeck, & Friedland, 2009). Despite this prevalence, reports on overall outcomes for pediatric HL in SA is limited. Since the first multichannel cochlear implantation took place in 1986, more than 1000 children have been implanted at 11 respective CI programs in South Africa (South African Cochlear Implant Group, 2017). With an increase in bilateral implantation and a growing number of children with less severe hearing losses being implanted (Sparreboom, Leeuw, Snik, & Mylanus, 2012; Tait et al., 2010), the number of pediatric CI surgeries is increasing not only globally, but also in South Africa. Pediatric CI outcomes and predictors of these outcomes were only recently described for a South African sample (le Roux et al., 2016). HRQoL outcome was not included as a measure and therefore, no published data yet exist on HRQoL outcomes in South Africa for pediatric CI recipients. The impact of certain demographic factors on HRQoL outcomes has not yet been explored for pediatric CI recipients in South Africa.

A number of studies on HRQoL outcomes in pediatric CI recipients reported that parents are less satisfied with their child's educational outcomes compared to other HRQoL sub-domains (Sack & Whynes, 2005; Huttunen et al, 2009; Kumar et al, 2015; Zhao et al., 2018). This study aimed to investigate why parental expectations in terms of education are often not met. This study, therefore, gave prominence to

those parents of older CI recipients who attend formal schooling and excluded parents of very young CI recipients who are not yet in school.

The aim of this study was to describe HRQoL outcomes of school-going CI recipients in a South African cohort from the perspectives of parents. This study is also one of the first to relate parental ratings of CI-specific HRQoL in pediatric CI recipients to demographic, hearing loss and CI-related variables not yet described in literature.

2. METHODOLOGY

2.1 Research aim

The main aim of this study was to describe HRQoL outcomes of school-going pediatric CI recipients in a South-African cohort from the perspectives of parents and to relate parental ratings of HRQoL to a range of demographic variables.

2.2 Research design

This study employed a descriptive cohort research design. Cohort studies are also known as observational studies since there is no manipulation of any variable (Haynes & Johnson, 2009). Descriptive research describes events as they occur naturally, without changing the situation under investigation (Irwin, Pannbacker, & Lass, 2010). Furthermore, descriptive research can also be defined as non-experimental, since this type of research aims to identify characteristics of an observed occurrence in a clinical or natural setting (Leedy & Ormrod, 2010; Maxwell & Satake, 2006). It can also involve the acquisition of information about an individual's attitudes or opinions by asking questions and tabulating the responses (Leedy & Ormrod, 2010; Maxwell & Satake, 2006). Since this research had the purpose to explain and predict, quantitative data were collected (Leedy & Ormrod, 2010). Data were collected at a single point in time (Leedy & Ormrod, 2013).

2.3 Ethical considerations

This study was initiated and conducted within the framework of the ethical guidelines as set out in the South-African National Health Act (2007). The individual principles presented in these documents are listed and discussed below in Table 2.1 as they were applied to the study.

Table 2.1. Ethical principles applied to the study (South African National Health Act, 2007; Leedy & Ormrod 2010; Leedy & Ormrod, 2013).

Principle	Application to study
A researcher conducting research involving human subjects is obliged to submit their research proposal for approval by an accredited research ethics committee.	Institutional ethics committee approval was obtained from the Research Ethics Committee of the Faculty of Humanities, University of Pretoria, prior to the commencement of data collection (Appendix A).
The right, safety, and well-being of the parent participants are the most important considerations and should prevail over the interest of science and society. Foreseeable risks and inconveniences should be weighed against the anticipated benefit for parent participants/ their implanted children and society. A study should only be initiated and continued if the anticipated benefits justify the risks.	At all times the rights and welfare of the participants were protected. There were no risks involved for the participants of this research.
Freely given informed consent to participate in the research should be obtained from all parties involved prior to the commencement of data collection.	An information letter was provided to the three participating CI programs outlining the purpose of the study and procedures for data collection (Appendix B). Permission to conduct the study and access patient data has been obtained from the CI team coordinators of the Pretoria Cochlear Implant Unit, the Johannesburg Cochlear Implant Centre and the Durban Cochlear Implant Program (Appendix B). Research participants also received an information letter that informed them of the nature of the research as well as what was expected from them in terms of participation (Appendix C). Informed consent was required from each participant prior to the commencement of data collection (Appendix C). Participation was completely voluntary and participants had the right to withdraw from the study without any negative consequences.
The confidentiality of records that could identify participants should be protected, respecting the privacy and confidentiality rules in accordance with the applicable regulatory requirement(s). Participants' rights to privacy and confidentiality should be protected at all times.	Privacy of participants was maintained at all times during data collection and data analyses. The data received from participating CI team coordinators excluded identifying information. Data were presented anonymously for the purpose of data analysis. Each participant was given an alphanumeric research code and hence data were reported using this research code. The identity of the participant represented by this code was unknown, even to the researcher.
Plagiarism	Every attempt was made to avoid plagiarism in this research project. Where secondary work of others was utilized, complete and correct citations was used (Appendix D). The research project is the work of the researcher herself.
Storage of data	According to the policies of the University of Pretoria, data obtained from this project was stored in both hard and digital copy. Data will be archived at the Department of Speech-Language Pathology and Audiology at the University of Pretoria for fifteen years (Appendix E). During this period, data may be used for other research purposes.
Release of findings	The results of this research study will be published in accredited academic journals, as well as in a summative research report.

2.4 Participants

This study included 54 parents of school-going pediatric CI recipients. Three CI programs in South Africa participated in this multicentre study; from which two programs are situated in the Gauteng Province (Pretoria Cochlear Implant Unit and the Johannesburg Cochlear Implant Centre) and one program in the KwaZulu-Natal Province (Durban Cochlear Implant Program).

Inclusion criteria

Eligible participants for this study included parents of school-going pediatric CI recipients with at least one CI and with a minimum of six months implant use at the time of data collection. According to the *National Curriculum Statement* of the Department of Basic Education (2017), formal schooling in South Africa can be defined as Grade R to Grade 12 and starts in the year the child turns six (Department of Basic Education, 2007). Only parents who were proficient in English were considered for participation since the validated HRQoL measure used for data collection (Appendix C) was only available in English.

Exclusion criteria

Parents with implanted children who were diagnosed with visual, cognitive or developmental delays were excluded.

Procedure for participant selection

Permission was obtained from the CI team coordinators of the participating CI programs to access patient data and contact details of the parents who met the

inclusion criteria of this study (Appendix C). Patient registers at the participating CI programs were reviewed to allocate eligible parent participants.

Description of study population

Demographic and clinical characteristics of 54 pediatric CI recipients are presented in Table 2.2.

Table 2.2. Characteristics of school-going pediatric cochlear implant recipients (n=54)

Demographics	***% (n)***	***Hearing loss and cochlear implant characteristics***	***% (n)***
Age at study *(years)*		Unilateral/bilateral cochlear implantation*	
Mean (SD)	12.21 (3.60)	2 cochlear implants (bilateral)	48.20 (26)
Range	6.60-18.30	1 cochlear implant with hearing aid in non-implanted ear (bimodal)	31.50 (17)
		1 cochlear implant without a hearing aid in non-implanted ear	20.40 (11)
Gender *		Age at first cochlear implant *(years) (n=40)***	
Male	53.70 (29)	Mean (SD)	3.90 (2.41)
Female	46.30 (25)	Range	0.62-11.60
Educational setting *		Age at second cochlear implant (*years) (n= 26)*	
Mainstream private school	27.80 (15)	Mean (SD)	6.60 (3.90)
Mainstream public school	20.40 (11)	Range	1.50-13.70
Special needs school	18.52 (10)		
School for the Deaf (South-African Sign Language mode of communication)	16.70 (9)	Time-lapse between first and second implant (years)	
School for the hard of hearing (oral mode of communication)	9.30 (5)	(*n=26*)	3.34 (3.04)
Homeschool	7.41 (4)	Mean (SD)	0.20-10.60
		Range	
Communication mode *		Duration of CI use *(years)*	
Spoken language	79.63 (43)	Mean (SD)	8.21 (4.10)
Total communication (South African Sign Language and spoken language)	20.40 (11)	Range	0.70-15.80
Highest level of parental education *		Age at diagnosis of hearing loss (*years) (n=40)***	
Tertiary qualification (University or other)	63.00 (34)	Mean (SD)	19.83 (12.62)
Matric completed	20.40 (11)	Range	1.00-54.00
High school (Gr8-11)	14.81 (8)		
Primary school (Gr1-7)	1.90 (1)		
		Age at diagnosis of deafness/severe to profound hearing loss *(months) (n=40) **	
		Mean (SD)	20.40 (14.3)
		Range	1.00-60.00
		Duration of deafness prior to cochlear implant *(years)*	
		Mean (SD)	1.9 (2.0)
		Range	0.1-9.7
		Onset of hearing loss	
		Congenital/early onset	74.10 (40)
		Unknown	16.70 (9)
		Acquired/ progressive/ sudden onset	9.30 (5)

* *Data reported by parents*
*** Only congenital/early onset hearing loss were considered n=40*

2.5 Materials for data-collection

Children with Cochlear Implants: Parental Perspectives questionnaire

Parents completed a parental proxy HRQoL questionnaire, the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) (Archbold et al., 2008) (Appendix F). The CCIP is an established HRQoL assessment for children with CIs and is the only CI-specific HRQoL measure that is validated (Almeida, Matas, Couto, & Carvalho, 2015; Archbold et al., 2008; Kumar et al., 2015; Warner-Czyz, Loy, Roland, & Tobey, 2013; Warner-Czyz et al., 2011; Zhang et al., 2016). The development of the CCIPP has been fully described (Archbold et al., 2002, 2008; O'Neill et al., 2004). The CCIPP has been shown to be a valid, reliable and robust HRQoL measure (Nunes, Pretzlik, & Ilicak, 2005; O'Neill et al., 2004). Originally the CCIPP was employed as a parent-directed retrospective interview (Archbold et al., 2002). With consistent results across methodologies, subsequent studies employed semi-structured interviews (Nunes et al., 2005), and also self-administered surveys (Archbold et al., 2008; Huttunen et al., 2009; Incesulu et al., 2003; O`Neill et al., 2004). The CCIPP includes 74 statements in which two main domains of the cochlear implantation process is covered: decision-making (26 items) and outcomes of implantation (48 items). Eight domains are used to categorize the outcomes of implantation, including, communication, general functioning, well-being, self-reliance, social relations, education, effects of implantation, and supporting the child. Parents of school-going CI recipients were asked to rate their response to each statement on a 5-point Likert scale that ranges from strongly agree to strongly disagree. Most (46) statements are worded positively and 28 are worded negatively in order to balance

items for negativity and positivity. The majority of the items of the CCIPP questionnaire relate to children across a broad age range. Five of the CCIPP items for outcomes of implantation relate to educational/ school issues specifically, making this a credible HRQoL outcome measure for school-aged pediatric CI recipients specifically.

Demographic, CI and hearing loss related variables

Additional demographic data were captured for the purpose of this study. The following demographic variables were reported on by parents at the beginning of the CCIPP questionnaire: gender, age at study, communication mode, unilateral or bilateral cochlear implantation, educational setting and highest level of parental education. Team coordinators of the participating CI programs assisted with the capturing of retrospective demographic, CI and hearing loss related data from the clinical patient files (Appendix H). Variables captured from clinical files included demographic variables (age at study, and highest level of parental education), CI related variables (age at first cochlear implant, age at second cochlear implant, duration of cochlear implant use, time-lapse between first and second cochlear implant) and hearing loss related variables (age at diagnosis of hearing loss, age at diagnosis of deafness/ severe to profound hearing loss, duration of deafness prior to cochlear implant, and onset of hearing loss).

2.6 Data collection procedures

Prospective parent participants were contacted by telephone to inform them verbally about the purpose and procedures of the research study. Those parents who were interested to participate received an information letter in hard copy or via email containing the purpose of the study and requirements for participation (Appendix C). Informed consent was required from each participant prior to the initiation of data collection (Appendix C). Parents who consented to participate were requested to complete a self-administered parent-proxy HRQoL questionnaire either electronically (online) or in hard-copy (Appendix F). Participants were requested to complete the questionnaire within seven working days of receiving the questionnaire. For those parents who preferred to complete the HRQoL questionnaire in hard-copy, written consent was required. For those parents who completed the HRQoL questionnaire electronically (online), a website link opened into a website page that reiterated the information contained in the consent slip previously emailed to them together with the information letter. Parents then responded with a virtual "click to accept" key that served as an indication of their consent (Leedy & Ormrod, 2013). Access to the HRQoL questionnaire was only possible once consent has been provided. Responses from the HRQoL questionnaire were returned to the researcher either electronically (online) or in hard-copy. A total of 120 questionnaires were distributed among the three participating CI programs and 54 questionnaires were returned (response rate of 45%).

Team coordinators of the participating CI programs were requested to assist with the capturing of retrospective demographic, CI and hearing loss related data from the

clinical patient files of the implanted children of the parents who completed the HRQoL questionnaire. Data on demographic variables that were captured included date of birth, gender, educational setting, communication mode and highest level of parental education. Hearing loss related data included age at diagnosis of hearing loss, age at diagnosis of deafness/severe to profound hearing loss, duration of deafness prior to cochlear implant, and onset of deafness. Cochlear implant related data included, unilateral or bilateral implantation, age at first cochlear implant, age at second cochlear implant and duration of CI usage.

Questionnaire and demographic, CI and hearing loss related data were captured on an Excel spreadsheet for the purpose of data analysis. Data was collected over a period of four months data collection period.

2.7 Data analysis procedures

SAS version 9.4 was used for the statistical analysis. The eight domains outlined in the overarching domain of outcomes of implantation in the CCIPP were used for the HRQoL analysis in this study. These HRQoL domains included communication, education, effects of implantation, general functioning, well-being, self-reliance, social relations, and supporting the child. Parents rated their responses on a 5-point Likert scale ranging from 1 (strongly disagree) to 5 (strongly agree). Non-responses were categorized as missing values. Scoring of negative statements were reversed in order for higher values to correspond to a more positive response that represent better HRQoL (Huttunen et al., 2009; Kumar et al, 2015.; Zhao et al, 2018). Scores were categorized and averaged by HRQoL domains to yield a domain mean for each participant. An overall HRQoL mean score was also calculated for the eight

subdomains together. Descriptive statistics were used to define the children of parent participants in terms of demographic, cochlear implant and hearing loss characteristics (Table 2).

The criteria used to differentiate between pre- and post-lingual deafness in pediatric CI recipients was the age at diagnosis of severe-profound hearing loss before and after three years of age respectively (Fryauf-Bertschy, Tyler, Kelsay, Gantz, & Woodworth, 1997; Dowell et al., 2004; Ruffin et a.l, 2013). For bilateral implantation, only the implanted children who had at least six months experience with their bilateral implant at the time of data collection (completion of the CCIPP) were considered as bilateral implant users. All 26 children with bilateral implants whose parents participated had at least six months experience with their bilateral implants at the time of data collection and were considered as bilateral implant users for data analysis.

Distribution-free nonparametric statistics were used to accommodate for the smaller sample size and the use of ordinal data. In order to determine if associations exist among the respective HRQoL sub-domains, Spearman correlation coefficients were calculated. Spearman correlation coefficients were also used to assess possible associations between continuous variables *(chronological age, age at diagnosis of deafness, duration of CI use, age at first CI switch-on* and *duration of deafness*) and HRQoL sub-domain and overall ratings. For the *age at diagnosis of deafness* and the *age at first CI activation* variables, only the children with congenital/ early-onset hearing loss were considered (n=40) in order to reflect the current status of early hearing detection and intervention services in South Africa. In order to investigate

the influence of categorical variables on HRQoL sub-domain and overall ratings, general linear models were constructed. These categorical variables included *gender*, *educational setting*, unilateral or bilateral *cochlear implantation*, *onset of deafness* and *highest level of parental education*. A Bonferonni adjustment was used to reduce the likelihood of Type 1 error because of multiple comparisons. Statistical significance was accepted at the traditional $p < 0.05$ level.

3. RESEARCH ARTICLE

HEALTH-RELATED QUALITY OF LIFE IN SOUTH-AFRICAN CHILDREN WHO USE COCHLEAR IMPLANTS

Authors: Bianca Brewis, Talita le Roux, Kurt Schlemmer, Leone Nauta, Bart Vinck

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3.1 Abstract

Objective: To describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South African cohort from the perspectives of parents, and to relate parental ratings of HRQoL to a range of demographic variables.

Design: Parents of school-going CI recipients completed the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) CI specific HRQoL questionnaire. The effect of different demographic variables on HRQoL outcomes was also determined.

Study sample: The study sample included 54 parents of school-going (mean age = 12.2 years; 3.6 SD; range = 6.6-18.3 years) CI recipients with at least six months CI experience.

Results: Children's communication and general functioning with a CI received the most positive parental ratings. A number of statistically significant associations between HRQoL outcomes and demographic variables have been confirmed. A mainstream educational setting was associated with better HRQoL outcomes in terms of communication and education, while pre-lingual onset of deafness was associated with better HRQoL outcomes in terms of general functioning and well-being. Shorter duration of deafness and unilateral implantation were associated with higher parental ratings for self-reliance and well-being respectively. Longer duration of CI use was linked to improved ratings for effects of implantation.

Conclusion: Parents assigned positive ratings to their child's HRQoL. This exploration of children's HRQoL related to their CIs contributes to evidence-based pediatric CI services that promote optimal psychosocial outcomes.

3.2 Introduction

Cochlear implantation has become a well-established treatment option for children with severe to profound hearing loss who obtain insufficient benefit from acoustic amplification. Recent reports indicate that some prelingually deafened children who are implanted before the age of one can achieve speech and language skills comparable to their normal hearing peers (Ching et al., 2009; Niparko et al., 2010; Wie, 2010). Not only does cochlear implantation have a positive effect on the language acquisition skills, speech perception, speech production, and communication performance in young children, but it also has a broader impact on

their psychosocial well-being (Sack & Whynes, 2005; Warner-Czyz et al., 2013). In recognizing the need to actualize and measure the benefits and limitations of medical interventions such as cochlear implantation on an individual's social, emotional and physical well-being, the term health-related quality of life (HRQoL) has been defined (Loeffler et al., 2010). HRQoL refers to an individual's general health status and perception of his/ her position in life, affected by psychological state, social relationships and level of independence (WHO, 1998). In order to provide an all-inclusive account of cochlear implant (CI) outcomes for an individual, the functional impact of permanent hearing loss and consequent treatment on that individual's personal well-being should be assessed through HRQoL measures (Zaidman Zait & Smith, 2010).

Clinical assessments of CI efficacy typically relate to communication and auditory skills and these measures do not necessarily represent the CI`s effect on everyday functioning (Clark et al., 2012; Meserole et al., 2014). As a result, HRQoL has recently become a widespread outcome measure to quantify and monitor the effects of cochlear implantation not only for adults, but also for children. HRQoL measures should identify and quantify the specific areas of general well-being that are presumably influenced by CI use, in order for families and involved professionals to be able to support implanted children in their areas of concern (Kumar et al., 2015).

The impact of a CI on a child's well-being can be measured by using generic or condition-specific HRQoL measures. Condition-specific measures are sensitive to both positive and negative consequences of a specific disorder or treatment, while generic measures focus on broader HRQoL domains such as social well-being and

self-esteem, independent of a disorder or medical condition (Bjornson & McLaughlin, 2001; Hinderink, Krabbe, & Van Den Broek, 2000; Kumar et al., 2015; Wiebe et al., 2003). Obtaining HRQoL scores for the pediatric CI population usually, involve either the parents (parent-report) or the CI recipients themselves (self-report). Some pediatric CI recipients may not necessarily have the required skills to express their feelings or describe experiences as a result of immature communication skills (Hays et al., 2006). Responses from parents can be used in such cases to describe HRQoL, since parents are considered as the closest proxy to a child. Not only do parents provide valuable information about their child's general functioning, additional intervention needs and experienced benefits from cochlear implantation (Damen et al., 2007), but they also provide useful insight into the socio-emotional and physical well-being of their children (Warner-Czyz et al., 2009).

Significant improvement in HRQoL has been documented for pediatric CI recipients after CI surgery (Barton et al., 2006; Beadle et al., 2000; Sach & Barton, 2007). Positive ratings for a variety of generic and CI specific HRQoL domains have been reported in the literature across HRQoL measures, including communication (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Loy et al., 2010; Sack & Whynes, 2005; Warner-Czyz et al., 2009), general functioning (Almeida et al., 2015; Zhang et al., 2016); physical and psychological well-being (Huber, 2005; Warner-Czyz et al., 2009; Loy et al., 2010), social relations (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Sack & Whynes, 2005) and self-reliance (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Sack & Whynes, 2005). However, literature reports less positive ratings for HRQoL domains that relate to education (Kumar et al., 2015,

Soleimanifar et al., 2015), the effects of the CI on the recipient (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Kumar et al., 2015; Sack & Whynes, 2005) and the support required by the implanted child (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Sack & Whynes, 2005).

Various demographic factors have been identified to influence HRQoL ratings in pediatric CI recipients. Female gender, higher levels of parental education, onset of hearing loss at an older age and the absence of additional special needs are associated with more favorable generic HRQoL outcomes in pediatric CI recipients (Barton et al., 2006; Sach & Barton, 2007). Younger chronological age, earlier implantation, longer duration of CI use and lower levels of family stress are also associated with more positive ratings of generic and CI specific HRQoL (Meserole et al., 2014; Sach & Barton, 2007; Warner-Czyz et al., 2009, 2011; Zhang et al., 2016)

Globally the number of pediatric CI surgeries is steadily increasing due to the expansion of selection criteria including those children with less severe degrees of hearing loss as well as those with additional medical conditions (Black et al., 2011; Fitzpatrick et al., 2009). Therefore the measurement of HRQoL outcomes has become a fundamental component in pediatric cochlear implantation since it contributes to evidence-based services that eventually endorse optimal outcomes. Despite this recent focus to assess the broader personal impact of permanent hearing loss and cochlear implantation in children, further investigation is needed to explore the possible influence of different interacting factors on HRQoL outcomes.

At the end of 2017, just more than 1000 children have been implanted at 11 respective CI programs in South Africa (South African Cochlear Implant Group, 2017). A previous report on hearing loss diagnosis and age of intervention of 264 pediatric CI recipients in South Africa, indicated that hearing loss was typically diagnosed late (15.3 months), resulting in delayed initial hearing aid fitting (18.8 months), enrolment in early intervention services (19.5 months) and eventual cochlear implantation (43.6 months) (le Roux et al, 2015). Within this sample, the majority of children (74%) were communicating orally, while 13% used Total Communication, 6% used South African Sign Language, and the remaining 7% used alternative manual modes of communication (le Roux et al, 2015). Only recently has preliminary data been published on pediatric CI outcomes and predictors of these outcomes within a South African sample of 301 pediatric CI recipients (le Roux et al., 2016). However, HRQoL was not included as an outcome measure and as a result, no published data yet exists on HRQoL outcomes in pediatric CI recipients in South Africa. Also, the impact of specific demographic factors on HRQoL outcomes has not yet been explored for pediatric CI recipients in South Africa. A number of studies on HRQoL outcomes in pediatric CI recipients reported that parents are less satisfied with their child's education compared to other HRQoL sub-domains (Huttunen et al., 2009; Kumar et al., 2015; Sack & Whynes, 2005). In addition, limited information on educational setting and including very young children (not yet in a formal educational setting) has been acknowledged as limitations in recent studies that used a validated CI-specific measures to report on HRQoL in children with CIs (Kumar et al, 2015; Zhao et al, 2018). In an attempt to investigate why parent expectations are often not met in terms of education, this study excluded parents of very young (<6 years of age) CI recipients and gave prominence to parents of older CI recipients who attend

formal schooling. Therefore, this study aims to describe HRQoL outcomes of school-going CI recipients in a South African cohort from the perspectives of parents. This study is also one of the first to relate parental ratings of CI-specific HRQoL in pediatric CI recipients to demographic, hearing loss and CI related variables not yet described in literature.

3.3 Methods

Study population

This study included 54 parents of school-going pediatric CI recipients. Three CI programs in South Africa participated in this multicentre study, from which two programs are situated in the Gauteng Province (Pretoria Cochlear Implant Unit and the Johannesburg Cochlear Implant Centre) and one program in the KwaZulu-Natal Province (Durban Cochlear Implant Program). Eligible participants for this study included parents of school-going pediatric CI recipients with at least one CI and with a minimum of six months implant use at the time of data collection. According to the National Curriculum Statement of the Department of Basic Education (2017), formal schooling in South Africa can be defined as Grade R to Grade 12 and starts in the year the child turns six (Department of Basic Education,2007). Only parents who were proficient in English were considered for participation since the validated HRQoL measure used for data collection was used in its original English format. Parents with implanted children who were diagnosed with visual, cognitive or developmental delays were excluded. Permission was obtained from the CI team coordinators of the participating CI programs to access contact details of parents and

patient data from clinical files of the children of parents who met the inclusion criteria of this study. Patient registers at the participating CI programs were reviewed to allocate eligible parent participants. Demographic and clinical characteristics of 54 pediatric CI recipients are presented in Table 3.1. All children were active users of their cochlear implants and were implanted for at least six months. Nearly half of the children (48.2%) were implanted bilaterally, with the interval between first and second implant ranging from 0.2 to 10.6 years (M = 3.3 years; 3.0 SD; n=26). The majority of children in this sample were oral communicators (79.6%) with the remaining 20.4% using a combination of oral communication and South African Sign Language (n=54). In terms of educational setting, almost half of the children were in either private or public mainstream settings (48.2%), with 18.5% being placed in a special needs school, 16.7% in schools for the Deaf, 9.3% in schools for the hard-of-hearing and 7.4% home-schooled. (n=54). Most children had pre-lingual deafness (74%), compared to 26% who were post-lingually deafened.

Table 3.1. Characteristics of school-going pediatric cochlear implant recipients (n=54)

Demographics	***% (n)***	***Hearing loss and cochlear implant characteristics***	***% (n)***
Age at study *(years)*		Unilateral/bilateral cochlear implantation*	
Mean (SD)	12.21 (3.60)	2 cochlear implants (bilateral)	48.20 (26)
Range	6.60-18.30	1 cochlear implant with hearing aid in non-implanted ear (bimodal)	31.50 (17)
		1 cochlear implant without a hearing aid in non-implanted ear	20.40 (11)
Gender *		Age at first cochlear implant *(years) (n=40)***	
Male	53.70 (29)	Mean (SD)	3.90 (2.41)
Female	46.30 (25)	Range	0.62-11.60
Educational setting *		Age at second cochlear implant (*years) (n= 26)*	
Mainstream private school	27.80 (15)	Mean (SD)	6.60 (3.90)
Mainstream public school	20.40 (11)	Range	1.50-13.70
Special needs school	18.52 (10)		
School for the Deaf (South-African Sign Language mode of communication)	16.70 (9)	Time-lapse between first and second implant (years)	
School for the hard of hearing (oral mode of communication)	9.30 (5)	(*n=26*)	3.34 (3.04)
Homeschool	7.41 (4)	Mean (SD)	0.20-10.60
		Range	
Communication mode *		Duration of CI use *(years)*	
Spoken language	79.63 (43)	Mean (SD)	8.21 (4.10)
Total communication (South African Sign Language and spoken language)	20.40 (11)	Range	0.70-15.80
Highest level of parental education *		Age at diagnosis of hearing loss (*years) (n=40)***	
Tertiary qualification (University or other)	63.00 (34)	Mean (SD)	19.83 (12.62)
Matric completed	20.40 (11)	Range	1.00-54.00
High school (Gr8-11)	14.81 (8)		
Primary school (Gr1-7)	1.90 (1)		
		Age at diagnosis of deafness/severe to profound hearing loss *(months) (n=40) **	
		Mean (SD)	20.40 (14.3)
		Range	1.00-60.00
		Duration of deafness prior to cochlear implant *(years)*	
		Mean (SD)	1.9 (2.0)
		Range	0.1-9.7
		Onset of hearing loss	
		Congenital/early onset	74.10 (40)
		Unknown	16.70 (9)
		Acquired/ progressive/ sudden onset	9.30 (5)

* *Data reported by parents*
*** Only congenital/early onset hearing loss were considered n=40*

Materials for data-collection

Parents completed a parental proxy CI specific HRQoL questionnaire, the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) (Archbold et al., 2008). The CCIP is an established and validated HRQoL assessment for children with CIs (Almeida et al., 2015; Archbold et al., 2008; Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Kumar et al., 2015; Warner-Czyz et al., 2013, 2009, 2011; Zhang et al., 2016). The development of the CCIPP has been fully described (Archbold et al., 2002, 2008; O'Neill et al., 2004). Originally the CCIPP was employed as a parent-directed retrospective interview (Archbold et al., 2002). Demonstrating consistent results across methodologies, subsequent studies made use of semi-structured interviews (Nunes et al., 2005), and also self-administered surveys (Archbold et al., 2008; Huttunen et al., 2009; Incesulu et al., 2003; Kumar et al., 2015; O'Neill et al., 2004). The CCIPP has been shown to be valid, reliable and robust HRQoL measure (Nunes et al., 2005; O'Neill et al., 2004). It includes 74 statements in which two main domains of the cochlear implantation process is covered: decision-making (26 items) and outcomes of implantation (48 items). Eight sub-domains are used to categorize the outcomes of implantation, consisting of five child related sub-domains (*communication, general functioning, well-being, self-reliance, social relations,* and *education*) and two family-related sub-domains (*effects of implantation* and *supporting the child*). Parents were asked to rate their response to each statement on a 5-point Likert scale (ranging from strongly agree to strongly disagree). Most (46) statements are worded positively and 28 are worded negatively. Additional demographic, hearing loss and CI related data were captured for the purpose of this study. Data on the following demographic variables were obtained

from parents in conjunction to completing the CCIPP questionnaire: their child's gender, communication mode, unilateral or bilateral cochlear implantation and current educational setting, as well as the parents' highest level of education. Team coordinators of the participating CI programs assisted with the capturing of additional retrospective CI and hearing loss related data from the clinical patient files. Data were captured for CI related variables (unilateral or bilateral cochlear implantation, age at first CI, age at second CI, duration of CI use) and hearing loss related variables (age at diagnosis of hearing loss, age at diagnosis of deafness/ severe to profound hearing loss, duration of deafness prior to CI, and onset of hearing loss).

Data collection

Parents who met the inclusion criteria of this study were contacted by telephone or were approached during clinical consultations by the researchers (authors 1-4) to inform them verbally about the purpose and procedures of the study. Permission was obtained from CI team coordinators of the participating CI programs to access patient data and contact details of the parents who met the inclusion criteria. Those parents willing to participate received an information letter in hard copy or via email containing the purpose of the study and requirements for participation. Informed consent was required from each parent participant prior to the initiation of data collection. A total of 120 questionnaires were distributed among the three participating CI programs and 54 questionnaires were returned (response rate of 45%). Parents who consented to participate were requested to complete the CCIPP questionnaire either electronically (online) or in hard-copy, within seven working days of receiving the questionnaire. Team coordinators of the participating CI programs

were requested to assist with the capturing of retrospective demographic, CI and hearing loss related data from the clinical patient files. Results of the CCIPP questionnaire, as well as demographic, CI and hearing loss related data, were captured on an Excel spreadsheet for the purpose of data analysis. Data were collected over a four months data collection period.

Statistical analysis

A commercially available statistical software package (SAS version 9.4) was used for the analysis. Values were assigned to responses obtained on a 5-point Likert scale (1 = strongly disagree to 5 = strongly agree), making it possible to calculate standard deviations and estimate the dispersion in scores (O'Neill et al., 2003). Non-responses were processed as missing values. Scoring of negative statements were reversed in order for higher values to correspond to a more positive response that represent better HRQoL (Huttunen et al, 2009; Kumar et al, 2015; Zhao et al, 2018). Scores for each HRQoL domain were averaged to yield a domain and overall HRQoL mean for each participant.

Frequency tables and descriptive statistics were used to describe the implanted children of parent participants in terms of demographic, CI and hearing loss characteristics (Table 1). The criteria used to differentiate between pre- and post-lingual deafness in pediatric CI recipients was the age at diagnosis of severe-profound hearing loss before and after three years of age respectively (Fryauf-Bertschy et al, 1997; Dowell et al., 2004; Ruffin et al., 2013). All 26 children with bilateral implants whose parents participated had at least six months experience with

their bilateral implants at the time of data collection and were considered as bilateral implant users for data analysis.

Distribution-free nonparametric statistics were used to accommodate for the smaller sample size and the use of ordinal data. Multiple comparisons of HRQoL domains were done by means of post hoc testing. By using the Wilcoxon signed rank test, pairwise comparisons were performed to determine if significant differences exist between HRQoL domains. In order to determine if associations exist among the respective HRQoL sub-domains, Spearman correlation coefficients were calculated. Spearman correlation coefficients were also used to assess possible associations between continuous variables *(chronological age, age at diagnosis of deafness, duration of CI use, age at first CI activation* and *duration of deafness*) and HRQoL sub-domain and overall ratings. For the *age at diagnosis of deafness* and the *age at first CI activation* variables, only the children with congenital/ early-onset hearing loss were considered (n = 40), in order to reflect the current status of early hearing detection and intervention services in South Africa. In order to investigate the influence of categorical variables on HRQoL sub-domain and overall ratings, general linear models were constructed. These categorical variables included *gender*, *educational setting*, *cochlear implantation*, *onset of deafness* and *highest level of parental education*. Statistical significance was accepted at the traditional $p < 0.05$ level. A Bonferonni adjustment was performed to reduce the likelihood of Type 1 error owing to multiple comparisons.

3.4 Results

Comparisons among health-related quality of life domains

All eight HRQoL domains` mean ratings together with the mean rating of the overall HRQoL score (M= 3.79; SD = 3.77; range = 2.60-4.46) surpassed three on a 5-point Likert scale, demonstrating that parents regarded their child's quality of life as being more positive than negative.

Table 3.2 presents descriptive statistics for sub-domain and overall HRQoL ratings of the CCIPP questionnaire Highest mean scores were obtained for *communication* (M= 4.15, SD = 0.62, range = 2.60-4.46) and *general functioning* (M= 4.05, SD = 0.51, range = 2.83-5.00). Parents also rated *well-being* (M= 3.81, SD = 0.51, range = 2.40-5.00), *self-reliance* (M= 3.88, SD = 0.63, range = 2.00-5.00), *social relations* (M= 3.87, SD = 0.52, range = 2.71-5.00) and *education* (M= 3.70, SD = 0.64, range = 2.29-5.00) positively (mean scores higher than 3.7 out of 5). Lowest mean scores were obtained for *effects of implantation* (M= 3.49, SD = 0.62, range = 2.00-4.83) and *supporting the child* (M= 3.46, SD = 0.74, range = 2.17- 4.33).

Wilcoxon Signed Ranks Test was conducted to determine if significant differences ($p < 0.05$) exist between the rating parents have assigned to the different HRQoL sub-domains. Communication had the highest number of significant interdomain differences. C*ommunication* obtained significantly more positive ranks than *well-being* (Z= -4.040, *p*= 0.001*), self-reliance* (Z= -3.019, *p*= 0.003*)*, *social relations (*Z= -3.910, *p*= 0.001*), education* (Z= -4.800, *p*= 0.001*)*, *effects of implantation* (Z= -5.202, *p*= 0.001*)* and *supporting the child* (Z= -5.335, *p*= 0.001*). General-functioning* also obtained significantly more positive ranks than *well-being* (Z= -2.752, *p*= 0.006*)*,

social relations (Z= -2.644, *p*= 0.008*), education (*Z= -2.975*, p*= 0.003*)*, *effects of implantation* (Z= -4.476, *p*= 0.001*)* and *supporting the child* (Z= -5.231, *p*= 0.001*)*. Significantly more positive ranks were achieved for *well-being* than for the *effects of implantation* (Z= -2.691, *p*= 0.007*)* and *supporting the child* (Z= -2.933, *p*= 0.003*)*. Both *self-reliance* and *social relations* obtained significantly more positive ranks than *effects of implantation* (Z= -3.457, *p*= 0.001*)* and *supporting the child* (Z= -3.898, *p*= 0.001*)*. Lastly, *education* obtained more positive ranks than *supporting the child* (Z= -2.420, *p*= 0.016*).*

Table 3.2. Health-related quality of life scores depicted from CCIPP results (n= 54)

CCIPP questionnaire	*Description of sub-domain**	*Mean (SD)*	*Median*	*Range*
Total HRQoL score		3.79 (0.37)	3.77	2.60-4.46
CCIPP sub-domains				
Communication	Ease, quality, and quantity of communication and conversation	4.15 (0.62)	4.14	2.00-5.00
General Functioning	Changes in attention, safety, and engagement	4.05 (0.51)	3.92	2.83-5.00
Well Being	Happiness and frustration	3.81 (0.60)	3.80	2.40-5.00
Self-Reliance	Indicators of confidence and independence	3.88 (0.63)	4.00	2.00-5.00
Social Relations	Relationships within and outside the family	3.87 (0.52)	3.86	2.71-5.00
Education	Performance of the child at school; placement and responsiveness within the school district	3.70 (0.64)	3.71	2.29-5.00
Effects of implantation	Progress with the cochlear implant, future concerns regarding device function, and child reaction to the device	3.49 (0.62)	3.54	2.00-4.83
Supporting the child	Amount and effects of help required by child before and after implantation	3.46 (0.47)	3.50	2.17-4.33

**Description according to Archbold et al., 2008; Kumar et al., 2015*

Associations among health-related quality of life sub-domains

In order to determine if associations exist among the respective HRQoL sub-domains, Spearman correlation coefficients were calculated (Table 3.3). *Communication* achieved the highest number of highly significant ($p < 0.05$) interdomain associations. Ratings of *communication* positively correlated with five domains, (ordered from greatest to least magnitude), namely *social relations* (r=0.65), *self-reliance* (r=0.59), *general-functioning* (r=0.53), *education* (r=0.53) and *well-being* (0.52). Moderate positive correlations that were highly significant ($p<0.05$) were found between *well-being* and *education* (r=0.52, $p<0.001$) and between *self-reliance* and *education* (r=0.53, $p<0.001$).

General functioning achieved significant ($p< 0.05$) positive correlations with three domains (listed in order from greatest to least magnitude), namely *self-reliance* (r=0.42), *social relations* (r=0.35) and *well-being* (r=0.28). Likewise, *well-being* positively correlated significantly ($p<0.05$) with *self-reliance* (r=0.42) and *social relations* (r=0.35). Listed in order from greatest to least magnitude, significant ($p<0.05$) positive correlations were also found between *social relations* and *education* (r=0.42), *education* and e*ffects of implantation* (r=0.33), as well as *effects of implantation* and *supporting the child* (r=0.27).

Table 3.3. Spearman correlations among health-related quality of life sub-domains (n=54)

Health-related quality of life sub-domain	1	2	3	4	5	6	7	8
1. Communication		0.53 <.0001**	0.52 <.0001**	0.59 <.0001**	0.65 <.0001**	0.53 <.0001**	0.25 0.0653	0.18 0.1988
2. General functioning			0.28 0.0405*	0.42 0.0014*	0.41 0.0022*	0.13 0.3380	0.06 0.6607	0.26 0.0597
3. Well-being				0.42 0.0017*	0.35 0.0100*	0.52 <.0001**	0.15 0.2816	-0.03 0.8222
4. Self-reliance					0.37 0.0054**	0.53 <.0001**	0.21 0.1253	0.17 0.2158
5. Social relations						0.42 0.0017*	0.29 0.0354*	0.10 0.4675
6. Education							0.33 0.0162*	0.10 0.4747
7. Effects of implantation								0.27 0.0478*
8. Supporting the child								

* $p < 0.05$

** $p < 0.001$

Associations between continuous demographic variables and health-related quality of life ratings

In order to assess associations between continuous demographic variables and HRQoL sub-domain and overall ratings, Spearman correlations coefficients were computed. Five continuous variables were included in the analyses namely age at study (chronological age), age at first CI activation, duration of CI use, age at diagnosis of deafness/severe to profound hearing loss and duration of deafness prior to cochlear implantation (Table 3.4). A significant positive correlation was found between *duration of CI use* and the *general functioning* domain, implying that parents of children who had longer CI experience assigned more positive ratings for their children's general functioning (r= 0.31; *p*= 0.024). In contrast, a significant negative correlation was found between *duration of CI use* and *effects of*

implantation, indicating that parents of children who had longer CI experience were less satisfied with the *effects of implantation* relative to parents of children who had less CI experience (r= -0.33, *p*= 0.014). *Duration of deafness* correlated negatively with the *self-reliance domain*, suggesting that parents of children who had a longer duration of deafness prior to cochlear implantation, assigned lower ratings to their children's self-reliance relatively to parents of children who had a shorter duration of deafness prior to implantation (r= -0.31, *p*= 0.022).

Table 3.4. Associations between continuous demographic variables and health-related quality of life ratings

Health-related quality-of-life sub-domain	Age at study (n=54)	Age at first CI activation (n=40)	Duration of CI use (n=54)	Age at diagnosis of deafness (n=40)	Duration of deafness (n=54)
Communication	0.08	-0.31	0.14	-0.19	-0.16
	0.5863	0.0529	0.3245	0.2311	0.2383
General functioning	0.09	-0.25	0.31	-0.14	-0.23
	0.5317	0.1230	0.0241*	0.3790	0.0989
Well-being	-0.20	-0.03	-0.13	-0.15	0.00
	0.1482	0.8339	0.3410	0.3580	0.9719
Self-reliance	-0.03	-0.31	0.12	-0.15	-0.31
	0.8153	0.0522	0.4057	0.3480	0.0215*
Social relations	-0.17	-0.08	-0.16	-0.14	0.01
	0.2101	0.6099	0.2374	0.3807	0.9426
Education	-0.21	-0.16	-0.22	-0.08	-0.03
	0.1318	0.3208	0.1106	0.6452	0.8455
Effects of implantation	-0.19	-0.23	-0.33	0.24	0.13
	0.1621	0.1613	0.0141*	0.1331	0.3507
Supporting the child	0.00	-0.01	0.04	-0.01	-0.12
	0.9795	0.6591	0.7777	0.9548	0.4021
Overall HRQoL	-0.15	-0.17	-0.09	-0.13	-0.10
	0.2877	0.2929	0.5064	0.4320	0.4814

Spearman correlation coefficient

** p< 0.05*

Associations between categorical demographic variables and health-related quality of life ratings

A general linear model was constructed to investigate the influence of categorical demographic variables on overall HRQoL ratings. Likewise, general linear models were also constructed for each of the HRQoL sub-domains. Five categorical variables were considered for the modeling and included *gender* (male/ female), *educational setting* (mainstream/ non-mainstream), *cochlear implantation* (unilateral/ bilateral), *onset of deafness* (pre-/ post-lingual) and *highest level of parental education* (tertiary/ high school or matric).

No significant associations were found between categorical variables and overall HRQoL ratings. Table 3.5 presents the general linear regression analysis results in terms of the four HRQoL sub-domains that yielded significant (p<0.05) associations with categorical variables. Significant associations were only obtained for the *communication, general functioning, well-being*, and *education* sub-domains.

Table 3.5. Associations between categorical demographic variables and health-related quality of life sub-domain ratings (n=54)

Health-related quality of life subdomain	Categorical demographic variable	Pr>F (p-value)	Categories	Mean score (SD)
Communication	Education setting	0.0374	Mainstream (n=20) Non-mainstream (n=34)	4.33 (SD: 0.54) 3.98 (SD: 0.66)
General functioning	Onset of deafness	0.0214	Pre-lingually deafened (n=40) Post-lingually deafened (n=14)	4.16 (SD: 0.45) 3.71 (SD: 0.57)
Well-being	Cochlear implantation	0.0345	Bilateral (n=26) Unilateral (n=28)	3.66 (SD: 0.61) 3.94 (SD: 0.57)
	Onset of deafness	0.0465	Pre-lingually deafened (n=40) Post-lingually deafened (n=14)	3.88 (SD: 0.60) 3.61 (SD: 0.57)
Education	Educational setting	0.0161	Mainstream (n=26) Non-mainstream (n=28)	3.93 (SD: 0.58) 3.48 (SD: 0.62)

Educational setting was significantly associated with the *communication* sub-domain (p= 0.037), showing that parents of children in mainstream educational settings on average score their children's communication higher (M= 4.33, SD = 0.54) than parents whose children are not in a mainstream educational setting (M= 3.98, SD = 0.66). Onset of deafness was significantly associated with the *general functioning* sub-domain (*p*= 0.021), indicating that parents whose children were pre-lingually deafened on average score there children's general functioning higher (M= 4.16, SD = 0.45) than those parents whose children were post-lingually deafened (M= 3.71, SD = 0.57). Cochlear implantation and onset of deafness were significantly associated with the *well-being sub-domain* (*p*= 0.035 and *p*= 0.047 respectively). Parents of children implanted bilaterally on average score their children's well-being more negatively (M= 3.66, SD = 0.61) in comparison to parents of children implanted unilaterally (M= 3.94, SD = 0.57). Furthermore, parents of children with pre-lingual deafness on average score their children's well being higher (M= 3.88, SD = 0.60) than parents of children with post-lingual deafness (M= 3.61, SD = 0.57). Educational setting was significantly associated with the *education sub*-domain (*p*= 0.016), indicating that parents of children in mainstream educational settings on average score their children's general performance in school higher (M= 3.93, SD = 0.58) than parents of children in non-mainstream educational settings (M= 3.48. SD = 0.62).

3.5 Discussion

Parents of school-going pediatric CI recipients in this study perceived their children's HRQoL to be more positive than negative, by assigning positive ratings (on average exceeding three on a sub-scale of five) to overall HRQoL and all HRQoL sub-domains. Post-operative average ratings exceeding three out of five for all eight HRQoL sub-domains of the CCIPP have been confirmed by a number of studies (Huttunen et al., 2009; Kumar et al., 2015; Loy et al., 2010).

The *communication* sub-domain (referring to the ease, quality, and quantity of communication and conversation of the implanted child) achieved the highest mean score. It is clear from this study and from reports formerly published that parents consider their child's communication as one of the most advantageous aspects of HRQoL post-implantation (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Kumar et al., 2015; Sack & Whynes, 2005; Warner-Czyz et al., 2009, 2011). The second highest mean score was obtained for the *general-functioning* sub-domain (reflecting changes in attention, safety, and engagement of the implanted child*)*. Overall, parents perceived their child's increased dependence on auditory information and functional hearing in everyday environments as a positive outcome of implantation (Huttunen et al., 2009).

The two family-related sub-domains, namely *effects of implantation* (reflecting the child's progress with the CI, future concerns regarding device functioning, and the child's reaction to the device) and *supporting the child* (referring to the amount and

effects of help required by the child before and after implantation) received least positive ratings by parents in this study. Lower ratings for *effects of implantation* (Almeida et al., 2015; Huttunen et al., 2009; Kumar et al., 2015; Zhang et al., 2016) as well as *supporting the child* (Almeida et al., 2015; Huttunen et al., 2009) was also obtained in other studies on parent-perceived HRQoL in pediatric CI recipients. These findings indicate that parents, in general, are not only concerned about their child's future device function and reactions to the CI but also about the assistance their child will require in everyday life of the family. It is, therefore, necessary to further explore why cochlear implantation negatively affects ratings of family-related HRQoL.

The highest number of significant inter-domain associations was also achieved for the *communication* sub-domain, indicating positive correlations between children's communication and their social relations, self-reliance, general-functioning, education and well-being. According to parent perceptions, this implies that improved communication abilities in children with CIs lead to better relationships, greater independence, more reliance on auditory information, steady progress in academics and improved overall happiness. Significant positive correlations were indicated between the *education* and *well-being* sub-domains, as well as the *education* and *self-reliance* sub-domains. In agreement with previously published reports (Sack & Whynes, 2005; Huttunen et al, 2009; Kumar et al, 2015), the *education* sub-domain on average received modest parental ratings in this study. Irrespective, higher parental satisfaction with education strongly influenced overall well-being and independence. Parents who feel they cannot offer comprehensive support to their child, but rather only in certain areas, should be encouraged by these inter-domain

associations (Sack & Whynes, 2005). Since there is for instance a positive correlation between communication and social relations, parents' efforts to support their child's communication (attend regular device programming and speech-language therapy sessions) could also be beneficial to their child social well-being, even if they feel less adequate to assist their child in terms of social skills (Kumar et al, 2015).

In addition to describing CI-specific HRQoL outcomes from the perspectives of parents, this study also related HRQoL outcomes to a broad range of demographic, hearing loss and CI related variables. Duration of CI use correlated positively with the *general functioning* HRQoL sub-domain, indicating that parents of children who had longer CI experience assigned higher ratings to their child's general functioning in terms of positive changes in attention, safety, and engagement (functional hearing). Using an ad hoc, self-reported CI-specific questionnaire, Schorr et al., (2009) also confirmed that a longer duration of CI use correlated with higher overall HRQoL in a group of 37 pediatric CI recipients (ages 5-14 years). In contrast, a significant negative correlation was found between duration of CI use and the *effects of implantation* sub-domain in this study. This negative correlation suggests that parents of children who had longer CI experience were less satisfied with the effects of implantation relative to parents of children who had less CI experience. More positive HRQoL with less CI experience was also confirmed by Warner-Czyz et al. (2009) for a group of pre-schools (ages 4-7 years) CI recipients. With a broader range of ages (6.8 - 18.3 years) and duration of CI experience (0.7 - 15.8 years) in this study, this finding could suggest that the positive effects of implantation become less apparent to parents the longer the child is implanted. Since parents over time

become more concerned about their child's future device function and reactions to the CI, involved professionals should offer on-going counselling and sustained support to parents for extended periods of time after implantation.

Duration of deafness correlated negatively with the *self-reliance* HRQoL sub-domain (signifying the child's confidence and independence), indicating that parents of children who had a shorter duration of deafness prior to cochlear implantation assigned better ratings to their child`s *self-reliance* relatively to parents of children with longer duration of deafness prior to implantation. Since the duration of deafness/ severe-to-profound hearing loss implies the duration of auditory deprivation prior to cochlear implantation, it is known to be a critical predictor of implantation success (le Roux et al, 2017). Therefore, study results suggest that the shorter the period of auditory deprivation prior to implantation is, the more confident and independent parents will perceive their children to be.

As also confirmed by the significant association between the *communication* and *self-reliance* sub-domains in this study, pediatric CI recipients are more self-reliant and independent when they are able to communicate effectively (Huttunen et al, 2009).

The *communication* sub-domain, as well as the *education* sub-domain (referring to the performance of the child at school, as well as placement and responsiveness within the educational setting) were significantly associated with educational setting. These results imply that parents of children in mainstream educational settings on

average scored their children's communication abilities and school performance higher than parents of children in non-mainstream educational settings. Even though mainstream education is likely to be a realistic outcome for children implanted early and without additional developmental difficulties (Archbold et al, 2002; Damen et al, 2007), the emphasis of educational placement for pediatric CI recipients should rather be on the appropriateness of the educational setting to each child's specific needs (le Roux et al, 2016). Irrespective, the relationship between mainstream schooling and parent satisfaction with educational placement, performance, and progress was evident in this study.

Within this study sample, the onset of deafness had a significant influence on both *general functioning* and *well-being* HRQoL sub-domains. Parents of prelingually deafened children on average perceived their children's general functioning with the help of hearing and their overall well-being and happiness to be more positive than parents of post-lingually deafened children. This could relate to the fact that earlier age at onset of deafness concur with better psychosocial adjustment in school-going deaf children (Polat, 2003).

Contrary to expectations, parents of children with bilateral CIs on average scored their children's *well-being* more negatively in comparison to parents of children implanted unilaterally. However, this assosciation between bilateral implantation and HRQoL outcomes was only evident for one of the HRQoL sub-domains, and not for overall HRQoL outcomes. Irrespective, tthis finding is in contrast to a number of studies confirming the strong association between bilateral implantation and improved HRQoL outcomes in adult (Olze et al, 2012; Härkönen et al, 2015; le Roux

et al, 2017) and pediatric (Samuel et al, 2016) CI recipients . Evidence on whether bilateral implantation significantly improve broader outcomes such as HRQoL, is lacking for pediatric CI recipients (Johnston et al, 2009; Sparreboom et al, 2010). This study therefore provides some evidence that the expectations of parents of bilaterally implanted children are not necessarily met in terms of the overall comfort and happiness of their children. Also, since family financial resources remains a decisive factor for bilateral implantation in South Africa, not all children have equal opportunity to access a second implant (le Roux et al, 2016). It could be that the parental expectations of this selective (more privileged) sub-group of bilateral pediatric CI recipients in this study are very high and not easily met. It should however be noted that only three of the eleven CI programs in South Africa participated in this multicenter study, resulting in a relatively small sample size. Consequently the study sample for this research could not be considered as representative of parents of pediatric CI recipients in South Africa and results should not be generalized.

3.6 Conclusion

Parents of school-going pediatric CI recipients in this study assigned positive ratings to overall HRQoL and all HRQoL sub-domains. A mainstream educational setting was associated with better HRQoL outcomes in terms of communication and education, while pre-lingual onset of deafness was associated with better HRQoL outcomes in terms of general functioning and well-being. Longer duration of deafness and bilateral implantation were associated with lower parental ratings for self-reliance and well-being respectively. Longer duration of CI use was linked to

better ratings for general functioning, but poorer ratings for effects of implantation. This study provided valuable insights into parental perceptions of CI outcomes in terms of HRQoL. This investigation of children's HRQoL associated with their CIs contributes to evidence-based pediatric CI services that promote optimal psychosocial outcomes and assist professionals to make the best decisions about the required care and support for pediatric CI recipients and their parents.

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4. GENERAL DISCUSSION, CLINICAL IMPLICATIONS AND CONCLUSION

4.1 Discussion of results

In order to provide a wide-ranging interpretation of pediatric cochlear implantation outcomes, the functional impact of permanent hearing loss on pediatric CI recipients' personal well-being should be assessed through HRQoL measures. Professionals who provide services to pediatric CI recipients need to be sensitive to psychosocial issues beyond communication in order to optimize functional outcomes in implanted children (Kumar et al., 2015). Currently, published data on HRQoL outcomes in pediatric CI recipients in South Africa are non-existing. Furthermore, the impact of specific demographic factors on HRQoL outcomes has not yet been explored for South African pediatric CI recipients in South Africa. The current study aimed to describe HRQoL outcomes of school-going pediatric CI recipients in a South African cohort from the perspectives of parents and to relate parental ratings of HRQoL to a range of demographic variables.

Health-related quality of life outcome profile

Average ratings of overall HRQoL and all HRQoL sub-domains exceeded three on a 5-point Likert scale, indicating that parents viewed their child’s HRQoL on the positive side of the rating scale. Average post-operative ratings exceeding three out of five for all eight HRQoL sub-domains of the CCIPP have been confirmed by a number of studies (Huttunen et al., 2009; Kumar et al., 2015; Loy et al., 2010). Highest mean scores were obtained for the *communication* and *general functioning*

HRQoL sub-domains, confirming that parents consider their child's communication and increased dependence on auditory information in everyday environments as a positive outcome of implantation (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Incesulu et al., 2003; Kumar et al., 2015; Loy et al., 2010; Sack & Whynes, 2005; Warner-Czyz et al., 2009). In this study, the two family-related sub-domains, namely *effects of implantation* and *supporting the child* received the least positive ratings by parents. Lower ratings for both the *effects of implantation* (Almeida et al., 2015; Huttunen et al., 2009; Kumar et al., 2015; Zhang et al., 2016) and *supporting the child* (Almeida et al., 2015; Huttunen et al., 2009) sub-domains was also shown in other studies on parent-perceived HRQoL in pediatric CI recipients. In general, these findings point to the fact that parents are not only concerned about their child's future device function and reactions to the CI but also about the assistance their child will require in everyday life of the family.

Associations among health-related quality of life sub-domains

The *communication* sub-domain achieved the highest number of significant inter-domain associations, indicating positive correlations between children's communication and their social relations, self-reliance, general-functioning, education, and well-being. Significant positive correlations were indicated between the *education* and *well-being* sub-domains, as well as the *education* and *self-reliance* sub-domains. The *education* sub-domain on average received modest parental ratings in this study, as confirmed by previously published reports (Huttunen et al., 2009; Kumar et al., 2015; Sack & Whynes, 2005; Zhao et al., 2018). Irrespective, higher parental satisfaction with education strongly influenced overall well-being and

independence. Parents who feel they cannot offer comprehensive support to their child, but rather only in certain areas, should be encouraged by these inter-domain associations (Sack & Whynes, 2005).

Associations between continuous demographic variables and health-related quality of life ratings

Parents of children who had longer CI experience assigned higher ratings to their child's general functioning in terms of positive changes in attention, safety, and engagement. This was indicated by the positive correlation between duration of CI use and the *general functioning* HRQoL sub-domain. Schorr et al. (2009), by means of an ad-hoc self-reported CI-specific questionnaire, also confirmed that longer duration of CI use correlated with higher overall HRQoL. In contrast, a significant negative correlation was found between duration of CI use and the *effects of implantation* sub-domain in this study. This negative correlation suggests that parents of children who had longer CI experience were less satisfied with the effects of implantation relative to parents of children who had less CI experience. More positive HRQoL with less CI experience was also confirmed by Warner-Czyz et al (2009) for a group of pre-schools (ages 4-7 years) CI recipients. Professionals involved in the CI process should offer on-going counselling and sustained support to parents for extended periods of time after implantation, as parents become more concerned about their child's future device function and reactions to the CI over time.

Parents of children who had a shorter duration of deafness prior to cochlear implantation allocated better ratings to their child`s *self-reliance* compared to parents

of children with longer duration of deafness prior to implantation. This was confirmed by a negative correlation between duration of deafness and the *self-reliance* HRQoL sub-domain. Since the duration of deafness/ severe-to-profound hearing loss implies the duration of auditory deprivation prior to cochlear implantation, it is known to be a critical predictor of implantation success (le Roux et al., 2017). The shorter the period of auditory deprivation prior to implantation is, the more confident and independent parents will perceive their children to be. Thus, study results suggest that the shorter the period of auditory deprivation prior to implantation is, the more confident and independent parents will perceive their children to be.

Associations between categorical demographic variables health-related quality of life ratings

The relationship between mainstream schooling and parent satisfaction with educational placement, performance, and progress was evident in this study. Both the *communication* and *education* sub-domains were significantly associated with educational setting. Parents of children in mainstream educational settings on average scored their children's communication abilities and school performance higher than parents of children in non-mainstream educational settings.

Onset of deafness had a significant influence on both *general functioning* and *well-being* HRQoL sub-domains in this study sample. On average, parents of prelingually deafened children perceived their children's general functioning with the help of hearing and their overall well-being and happiness to be more positive than parents of post-lingually deafened children. This could relate to the fact that earlier age at

onset of deafness concurs with better psychosocial adjustment in school-going deaf children (Polat, 2003).

Research emphasizes the strong association between bilateral implantation and improved HRQoL outcomes in adult (Olze et al, 2012; Härkönen et al, 2015; le Roux et al, 2017) and pediatric (Samuel et al., 2016) CI recipients. In contrast to these published reports and contrary to expectations, parents of children with bilateral CIs on average scored their children's *well-being* more negatively in comparison to parents of children implanted unilaterally. Evidence on whether bilateral implantation significantly improve broader HRQoL outcomes is lacking for pediatric CI recipients (Johnston et al., 2009; Van Schoonhoven et al., 2013). This study provides some evidence that the expectations of parents of bilaterally implanted children are not necessarily met in terms of the overall comfort and happiness of their children.

4.2 Clinical implications and recommendations

- Because of the impact of CI use on psychosocial outcomes, the assessment of HRQoL, in particular, is a suitable way to ensure the most positive outcomes for pediatric CI recipients (Kumar et al., 2015). Consequently, specific areas of general well-being that are influenced by CI device use can be identified and professionals can support implanted children in these areas of concern (Kumar et al., 2015). Therefore national pre- and post-operative clinical protocols should include HRQoL as a required outcome measure for pediatric cochlear implantation. HRQoL data should be captured at regular intervals for all pediatric CI recipients in order to monitor progress over time

and to determine the developmental trajectories of HRQoL in the pediatric CI population in South Africa. Routine measurements of HRQoL will contribute to evidence-based pediatric CI services that promote optimal psychosocial outcomes.

- Parents of pediatric CI recipients are in a unique position to evaluate the impact a CI has on their child's psychosocial well-being within the context their child grows up (O'Neill et al., 2004). Exploring parental views during clinical contact sessions (by means of self-report questionnaires and discussion) can inform and guide the professionals involved in the rehabilitation process (Nunes et al., 2005; O'Neill et al., 2004). Obtaining the perspectives from parents also provide involved professionals with the opportunity to review their practices in order to provide families with useful information (Incesuluet al., 2003). Therefore it is imperative to understand parental perspectives and their experiences, in order to improve the quality of counseling and support provided at each stage of service delivery (Dev, Lohith, Pascal, Dutt, & Dutt, 2018).

- Early implanted pediatric CI recipients have an increased probability to develop age-appropriate language skills when compared to those implanted at older ages (Manrique, Cervera-Paz, Huarte, & Molina, 2004; Tobey et al., 2013; Wie, 2010). The average age for cochlear implantation for children with congenital/ early onset hearing loss in this study sample was 3.9 years, exceeding three and a half years of age. Possible contributing factors to late implantation may include funding constraints, lack of prompt referral to

specialized CI services, parental barriers such as delayed or missed appointments, complex medical conditions, family indecision and geographical region (Fitzpatrick, Ham, & Whittingham, 2015; le Roux et al., 2016).

- The two family-related HRQoL sub-domains, namely *effects of implantation* and *supporting the child* received least positive ratings by parents in this study. Kumar et al. (2015) indicated that parents have a strong desire for more support from professionals in order to equip them as parents to support their child with a CI.

- The *education* HRQoL sub-domain only received modest parental ratings in this study. This finding was confirmed by a number of other studies (Fortunato-Tavares et al., 2012; Huttunen et al., 2009; Kumar et al., 2015; Sack & Whynes, 2005). Pediatric CI recipients are often seen by educators in high functioning educational settings as an inconvenience due to their hearing disability (Sack & Whynes, 2005). Also, the lack of educational settings that are suitable for the unique needs of CI recipient is a concern often expressed by parents (Sach & Whynes, 2005; Zhao et al., 2018). Professionals involved in pediatric cochlear implantation should provide ongoing educational support and guidance, not only to the parents of pediatric CI recipients but also to their educators.

- A mainstream educational setting was associated with improved HRQoL outcomes in terms of the *communication* and *education* HRQoL sub-domains.

Some of the benefits of pediatric cochlear implantation include the acquisition and development of spoken language, as well as the integration into mainstream school environments for many CI recipients (Peixoto et al., 2013). Current South African educational policy has the long-term goal to develop an inclusive education system for children with severe-profound hearing loss, which will address barriers to learning such as language, communication and inflexible curriculums (Department of Education, 2007). Irrespective, numerous persistent challenges, such as limited access to specialist support in public ordinary schools, currently impede the progress that is being made toward an inclusive education system (Department of Education, 2015). It is possible that not all pediatric CI recipients in South Africa have equal access to supportive mainstream education due to socioeconomic and geographical constraints. Professionals involved in pediatric cochlear implantation should liaise with relevant authorities and role-players in order to address the challenges that are associated with inclusive education for children with severe-profound hearing loss.

4.3 Study strengths and limitations

A critical evaluation of this research project was conducted to evaluate its strengths and weaknesses.

Study strengths

- Limited published outcome data exist for pediatric cochlear implantation in South Africa (le Roux et al., 2016). This study was the first in South Africa to analyze parental perspectives of CI-specific HRQoL in pediatric CI recipients. Furthermore, this study was also one of the first to relate parental ratings of HRQoL in pediatric CI recipients to demographic, hearing loss and CI related variables not yet described in literature.

- Most published studies that used a validated CI-specific questionnaire to report on HRQoL in children with CIs, included very young children (not yet in a formal educational setting) in their samples with ages ranging between 1.8 - 18 years (Damen et al., 2007; Kumar et al., 2015; Zhao et al., 2018). As a result, information on educational aspects in these studies has been limited. In order to give prominence to parents of older CI recipients who attend formal schooling, this study included only parents of school-going children (6-18 years old) and excluded parents of very young (<6 years of age) CI recipients. The only validated parent proxy CI specific HRQoL questionnaire available at the time of data collection for this study, namely the CCIPP, (Archbold et al., 2002; Archbold et al., 2008; Schorr, Roth, & Fox, 2009; Warner-Czyz et al.,

2013, 2011) has been used for data collection. As no published data yet exist in South Africa, this study is the first to determine parental perspectives using a validated CI-specific HRQoL questionnaire.

- In this study, parental ratings of HRQoL were not only related to a range of demographic variables, but it was also determined if associations exist among the respective HRQoL sub-domains. Exploration of the interconnectivity among HRQoL sub-domains affords an opportunity for parents and professional to influence broader outcomes in pediatric cochlear implantation (Kumar et al., 2015).

Study limitations

Three CI programs in South Africa participated in this multicenter study, resulting in a relatively small dataset of 54 parents of pediatric CI recipients. Consequently, the study sample for this research could not be considered as representative of CI recipients in South Africa. With all the implanted children in this study representing only the private healthcare sector, this research sample is not representative of the larger South African population and results could not be generalized. Furthermore, a broader and larger study sample with greater variability might have produced different results.

The CCIPP was used in its original English format for the purposes of this study since it has not yet been translated and validated into other South African languages. Only parents who were proficient in English were, therefore, requested to complete

the questionnaire. As a result, some parents who were not proficient in English had to be excluded, leading to a predisposed study sample.

HRQoL outcome data were collected only at a single point in time (cross-sectionally) for the purpose of this study. Longitudinal outcome data, captured at fixed predetermined intervals (repeated measures), would allow for the monitoring of outcomes over time and for the assessment of development trajectories.

4.4 Future perspectives

Only until very recently, no CI–specific HRQoL measures have been developed using the Food and Drug Administration Guidance on patient-reported outcomes. Hoffman et al. (2018) developed the first CI-specific, self-report HRQoL instrument for school-age children with CIs, with an accompanying parent-proxy version. Future studies should consider the translation and adaption of such self-report CI-specific HRQoL measures in order to ensure suitability of its use for the South African population of pediatric CI recipients.

Once HRQoL outcome data are systematically captured at fixed intervals among South African CI programs for pediatric CI recipients, a prediction analysis can be valuable to report on the predictors of HRQoL outcomes, the evolution of HRQoL outcomes over the time and the consistency of predictors over time.

4.5 Conclusion

Parents of school-going pediatric CI recipients in this study assigned positive ratings to overall HRQoL and all HRQoL sub-domains. A mainstream educational setting was associated with better HRQoL outcomes in terms of communication and education, while pre-lingual onset of deafness was associated with better HRQoL outcomes in terms of general functioning and well-being. The *communication* and general functioning HRQoL sub-domains achieved the highest mean scores. Shorter duration of deafness and unilateral implantation were associated with higher parental ratings for self-reliance and well-being respectively. Longer duration of CI use was linked to better ratings for general functioning, but poorer ratings for effects of implantation. This study provided valuable insights into parental perceptions of CI outcomes in terms of HRQoL. This investigation of children's HRQoL associated with their CIs contributes to evidence-based pediatric CI services that promote optimal psychosocial outcomes and assist professionals to make the best decisions about the required care and support for pediatric CI recipients and their parents.

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6. APPENDIXES

Appendix A – Ethical clearance letter

UNIVERSITEIT VAN PRETORIA
UNIVERSITY OF PRETORIA
YUNIBESITHI YA PRETORIA

Faculty of Humanities
Research Ethics Committee

2 March 2017

Dear Prof Vinck

Project:	**Health related quality of life in South African children who use cochlear implants**
Researcher:	**B Brewis**
Supervisor:	**Ms T le Roux, Dr K Schlemmer and Prof B Vinck**
Department:	**Speech-Language Pathology and Audiology**
Reference number:	**13030303 (GW20170217HS)**

Thank you for the response to the Committee's correspondence of 27 February 2017.

I have pleasure in informing you that the Research Ethics Committee formally **approved** the above study at an *ad hoc* meeting held on 2 March 2017. Data collection may therefore commence.

Please note that this approval is based on the assumption that the research will be carried out along the lines laid out in the proposal. Should your actual research depart significantly from the proposed research, it will be necessary to apply for a new research approval and ethical clearance.

The Committee requests you to convey this approval to the researcher.

We wish you success with the project.

Sincerely

Prof Maxi Schoeman
Deputy Dean: Postgraduate and Research Ethics
Faculty of Humanities
UNIVERSITY OF PRETORIA
e-mail: tracey.andrew@up.ac.za

Appendix B - Information letter to and consent slips from cochlear implant teams

Pretoria Cochlear Implant Unit (PCIU)

UNIVERSITEIT VAN PRETORIA
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Faculty of Humanities
Department of Speech-Language Pathology and Audiology

Attention: Mrs Nicolize Cass

Team coordinator: Pretoria Cochlear Implant Unit

August 2016

Dear Mrs Cass,

RE: Permission to conduct a research study with parents of school-going cochlear implant recipients from the Pretoria Cochlear Implant Unit

I am a Master's degree student from the Department of Speech-Language Pathology and Audiology at the University of Pretoria. I have chosen to conduct my research in the field of cochlear implants (CIs). The aim of this study is to describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South African cohort from the perspectives of parents, and to relate parental ratings of HRQoL to a range of demographical variables. This multicentre study attempts to collect HRQoL outcome data of school-going pediatric CI recipients from the Durban Cochlear Implant Program (DCIP), the Pretoria Cochlear Implant Unit (PCIU) and Johannesburg Cochlear Implant Centre (JCIC).

Title: *Health-related quality of life in South African children who use cochlear implants*

Researcher: Bianca Brewis

Study leaders: Mrs Talita le Roux, Dr Kurt Schlemmer and Prof Bart Vinck

Design and procedure:

This study will follow a cross-sectional cohort design (descriptive research, collecting mainly quantitative data). Parents/ primary caregivers of school-going pediatric CI recipients between the ages of 6 and 18 years would be considered as research participants. Pediatric CI recipients with diagnosed or suspected cognitive, visual or developmental delay will be excluded as participants. Parents/ primary caregivers should be proficient in English.

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Fax: 012 420 3517

bart.vinck@up.ac.za
www.up.ac.za
talita.leroux@up.ac.za

Parent/ primary caregivers of pediatric CI recipients will be expected to complete a CI specific parent-proxy, self-administered HRQoL questionnaire

- Children with Cochlear Implants: Parental Perspectives (Archbold et al., 2002)

Additional to questionnaire data, information on demographical variables (such as age at identification of hearing loss, duration of CI use, bilateral/ unilateral implantation etc.) will be captured from clinical patient files .

Confidentiality: Identifying data of all participants will not be disclosed and data obtained from clinical patient files will be handled with strict confidentiality. Participants will be assigned an identifying code which will be used for data processing. Anonymity of all participants will be guaranteed at all times.

Data storage: All the research data/ and or documents referring to the above mentioned study will be stored in the Department of Speech-Language Pathology and Audiology (University of Pretoria). The storage of above mentioned data and/or documents will be maintained for a minimum of 15 years from the commencement of this study.

Written consent and assent: Parents of pediatric CI recipients will receive an information letter containing the purpose and procedures of the study. Freely given informed consent will be obtained from parents/ primary caregivers as well as assent provided by pediatric CI recipients. Participation is voluntary and all participants have the right to withdraw from the study at any time without any negative consequences.

Risks: No risks are associated with this study.

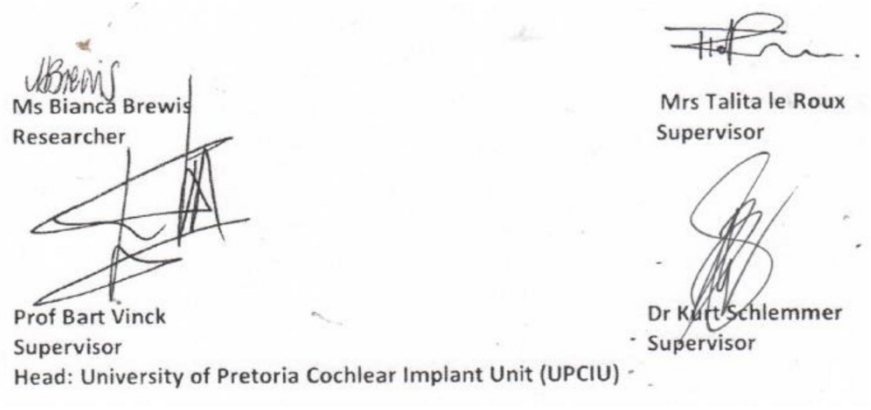
Release of findings: The results of this research study will be published in accredited academic journals, as well as in a summative research report.

Co-authorship of manuscripts to be submitted for publications: One representative from each CI-team will act as co-author for subsequent publications for this research study. Co-authors will be expected to give input to the manuscript drafts and oversee the data-collection procedure as their respective CI program for the purpose of this study.

In order to conduct this study, clinical data of pediatric CI recipients and contact details of parents are required. If permission for this is granted from you as team coordinator of the PCIU, you are requested to sign this letter of consent.

Please contact us should you require more information. Thank you in advance for your time and co-operation.

Yours sincerely,



Ms Bianca Brewis
Researcher

Mrs Talita le Roux
Supervisor

Prof Bart Vinck
Supervisor
Head: University of Pretoria Cochlear Implant Unit (UPCIU)

Dr Kurt Schlemmer
Supervisor

PCIU
PRETORIA COCHLEAR IMPLANT UNIT

PERMISSION FOR USE OF INFORMATION OF PEDIATRIC COCHLEAR IMPLANT RECIPIENT OF PCIU

Herewith I, Mrs Nicolize Cass, give permission that the information of pediatric Cochlear Implant recipients from the Pretoria Cochlear Implant Unit (PCIU) may be used for the research project titled: *Health-related quality of life in South African children who use cochlear implants.* I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

PRETORIA COCHLEAR IMPLANT UNIT
NPC 2015/24147/08

Mrs Nicolize Cass

Team coordinator: Pretoria Cochlear Implant Unit Date: 8/3/2017

Communication Pathology Building, Room 3-32, Lynnwood Road, Pretoria, 0002
Phone: 012 420 3684 | Fax: 012 420 3517 | Email: ptacoch@up.ac.za |

Johannesburg Cochlear Implant Centre (JCIC)

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Faculty of Humanities
Department of Speech-Language Pathology and Audiology

Attention: Mrs Leone Nauta

Team coordinator: Johannesburg Cochlear Implant Centre

August 2016

Dear Mrs Nauta,

RE: Permission to conduct a research study with parents of school-going cochlear implant recipients from the Johannesburg Cochlear Implant Centre

I am a Master's degree student from the Department of Speech-Language Pathology and Audiology at the University of Pretoria. I have chosen to conduct my research in the field of cochlear implants (CIs). The aim of this study is to describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South African cohort from the perspectives of parents, and to relate parental ratings of HRQoL to a range of demographical variables. This multicentre study attempt to collect HRQoL outcome data of pediatric CI recipients and their parents/ primary caregivers from the Durban Cochlear Implant Program (DCIP), the Pretoria Cochlear Implant Unit (PCIU) and Johannesburg Cochlear Implant Centre (JCIC).

Title: *Health-related quality of life in South African children who use cochlear implants*

Researcher: Bianca Brewis

Study leaders: Mrs Talita le Roux, Dr Kurt Schlemmer and Prof Bart Vinck

Design and procedure:

This study will follow a cross-sectional cohort design (descriptive research, collecting mainly quantitative data). Parents/ primary caregivers of school-going pediatric CI recipients between the ages of 6 and 18 years would be considered as research participants. Pediatric CI recipients with diagnosed or suspected cognitive, visual or developmental delay will be excluded as participants. Parents/ primary caregivers should be proficient in English.

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- Children with Cochlear Implants: Parental Perspectives (Archbold et al., 2002)

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Risks: No risks are associated with this study.

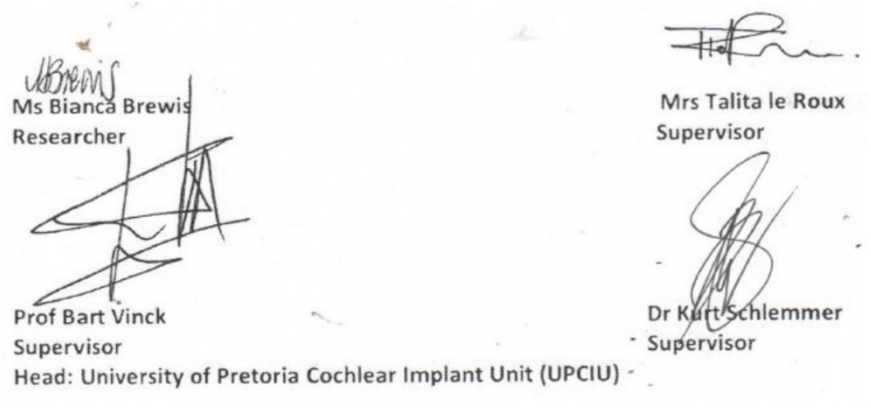
Release of findings: The results of this research study will be published in accredited academic journals, as well as in a summative research report.

Co-authorship of manuscripts to be submitted for publications: One representative from each CI-team will act as co-author for subsequent publications for this research study. Co-authors will be expected to give input to the manuscript drafts and oversee the data-collection procedure as their respective CI program for the purpose of this study.

In order to conduct this study, clinical data of pediatric CI recipients and contact details of parents are required. If permission for this is granted from you as team coordinator of the JCIC, you are requested to sign this letter of consent.

Please contact us should you require more information. Thank you in advance for your time and co-operation.

Yours sincerely,



Ms Bianca Brewis
Researcher

Mrs Talita le Roux
Supervisor

Prof Bart Vinck
Supervisor
Head: University of Pretoria Cochlear Implant Unit (UPCIU)

Dr Kurt Schlemmer
Supervisor

JCIC

JCIC
Lower Level
18 Eton Rd

011 482 6141
011 356 6510

2 June 2017

PERMISSION FOR USE OF INFORMATION OF PEDIATRIC COCHLEAR IMPLANT RECIPIENT
OF THE JOHANNESBURG COCHLEAR IMPLANT CENTRE

Herewith I, Mrs Leone Nauta give permission that the information of pediatric CI recipients from the Johannesburg Cochlear Implant Centre may be used for the research project titled: Health-related quality of life in South African children who use cochlear implants

The representative from JCIC who will act as co-author for subsequent publications for this research study will be:

Leoné Nauta

I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

L Nauta

Mrs Leone Nauta
Team coordinator: Johannesburg Cochlear Implant Centre

Date: 2/6/2017

UNIVERSITEIT VAN PRETORIA
UNIVERSITY OF PRETORIA
YUNIBESITHI YA PRETORIA
Denkleiers • Leading Minds • Dikgopolo tša Dihlalefi

Faculty of Humanities
Department of Speech-Language Pathology and Audiology

Attention: Dr Kurt Schlemmer

Team coordinator: Durban Cochlear Implant Program

August 2016

Dear Dr Schlemmer,

RE: Permission to conduct a research study with parents of school-going cochlear implant recipients from the Durban Cochlear Implant Program

I am a Master's degree student from the Department of Speech-Language Pathology and Audiology at the University of Pretoria. I have chosen to conduct my research in the field of cochlear implants (CIs). The aim of this study is to describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South African cohort from the perspectives of parents, and to relate parental ratings of HRQoL to a range of demographical variables. This multicentre study attempts to collect HRQoL outcome data of school-going pediatric CI recipients from the Durban Cochlear Implant Program (DCIP), the Pretoria Cochlear Implant Unit (PCIU) and Johannesburg Cochlear Implant Centre (JCIC).

Title: *Health-related quality of life in South African children who use cochlear implants*

Researcher: Bianca Brewis

Study leaders: Mrs Talita le Roux, Dr Kurt Schlemmer and Prof Bart Vinck

Design and procedure:

This study will follow a cross-sectional cohort design (descriptive research, collecting mainly quantitative data). Parents/ primary caregivers of school-going pediatric CI recipients between the ages of 6 and 18 years would be considered as research participants. Pediatric CI recipients with diagnosed or suspected cognitive, visual or developmental delay will be excluded as participants. Parents/ primary caregivers should be proficient in English.

Communication Pathology Building
Dept. of Speech-Language Pathology and Audiology
Corner of Lynnwood Road and Roper Street, Hatfield
Private Bag X20, Hatfield, 0028
University of Pretoria
PRETORIA
Republic of South Africa

Tel: 012 420 2355
Fax: 012 420 3517

bart.vinck@up.ac.za
www.up.ac.za
talita.leroux@up.ac.za

Parent/ primary caregivers of pediatric CI recipients will be expected to complete a CI specific parent-proxy, self-administered HRQoL questionnaire:

- *Children with Cochlear Implants: Parental Perspectives* (Archbold et al., 2002)

Additional to questionnaire data, information on demographical variables (such as age at identification of hearing loss, duration of CI use, bilateral/ unilateral implantation etc.) will be captured from clinical patient files .

Confidentiality: Identifying data of all participants will not be disclosed and data obtained from clinical patient files will be handled with strict confidentiality. Participants will be assigned an identifying code which will be used for data processing. Anonymity of all participants will be guaranteed at all times.

Data storage: All the research data/ and or documents referring to the above mentioned study will be stored in the Department of Speech-Language Pathology and Audiology (University of Pretoria). The storage of above mentioned data and/or documents will be maintained for a minimum of 15 years from the commencement of this study.

Written consent and assent: Parents of pediatric CI recipients will receive an information letter containing the purpose and procedures of the study. Freely given informed consent will be obtained from parents/ primary caregivers. Participation is voluntary and all participants have the right to withdraw from the study at any time without any negative consequences.

Risks: No risks are associated with this study.

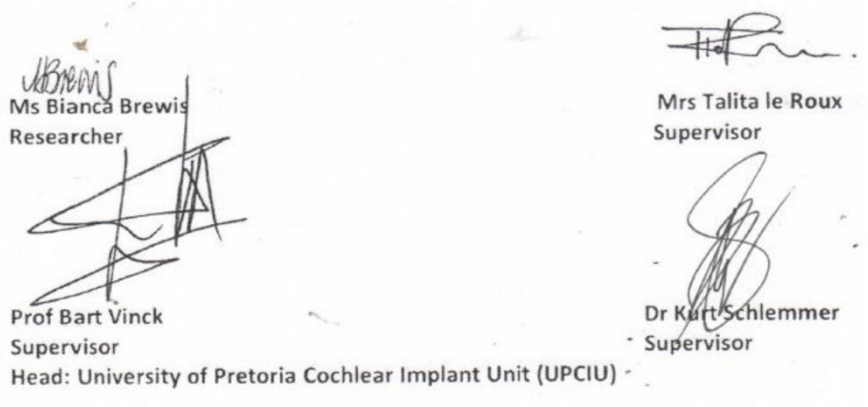
Release of findings: The results of this research study will be published in accredited academic journals, as well as in a summative research report.

Co-authorship of manuscripts to be submitted for publications: One representative from each CI-team will act as co-author for subsequent publications for this research study. Co-authors will be expected to give input to the manuscript drafts and oversee the data-collection procedure as their respective CI program for the purpose of this study.

In order to conduct this study, clinical data of pediatric CI recipients and contact details of parents are required. If permission for this is granted from you as team coordinator of the DCIP, you are requested to sign this letter of consent.

Please contact us should you require more information. Thank you in advance for your time and co-operation.

Yours sincerely,



Ms Bianca Brewis
Researcher

Mrs Talita le Roux
Supervisor

Prof Bart Vinck
Supervisor
Head: University of Pretoria Cochlear Implant Unit (UPCIU)

Dr Kurt Schlemmer
Supervisor

dcip
DURBAN COCHLEAR IMPLANT PROGRAMME

PERMISSION FOR USE OF INFORMATION OF PEDIATRIC COCHLEAR IMPLANT RECIPIENT OF DCIP

Herewith I, Dr Kurt Schlemmer give permission that the information of paediatric CI recipients from the Durban Cochlear Implant Program (DCIP) may be used for the research project titled: *Health-related quality of life in South African children who use cochlear implants.* I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

Dr Kurt Schlemmer

Teamcoordinator:
Durban Cochlear Implant Program (DCIP) **Date:** 7/3/17

Appendix C – Parental information letter and consent slip

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Faculty of Humanities
Department of Speech-Language Pathology and Audiology

May 2017

Dear parent of cochlear implant recipient,

RE: Permission to take part in research study on health-related quality of life in pediatric cochlear implant recipients in a South African cohort

I am a Master's degree student from the Department of Speech-Language Pathology and Audiology at the University of Pretoria. I have chosen to conduct my research in the field of cochlear implants (CIs). The main aim of my study is to describe the health-related quality of life (HRQoL) outcomes of school-going pediatric CI recipients from the perspectives of parents. Furthermore, this study aims to determine if associations exist between demographic variables and HRQoL outcomes. This multicentre study attempts to collect HRQoL outcome data from the parents of school-going pediatric CI recipients from the Durban Cochlear Implant Program (DCIP), the Pretoria Cochlear Implant Unit (PCIU), Tygerberg Hospital University of Stellenbosch Cochlear Implant Unit (TH-US-CIU) and Johannesburg Cochlear Implant Centre (JCIC).

Title: *Health-related quality of life in South African children who use cochlear implants*

Researcher: Bianca Brewis

Study leaders: Dr Talita le Roux, Dr Kurt Schlemmer, and Prof Bart Vinck

Design and procedure:

This study will follow a cross-sectional cohort design (descriptive research, collecting mainly quantitative data). The parents of school-going pediatric CI recipients between the ages of 7 and 18 years would be considered as research participants. Pediatric CI recipients with diagnosed or suspected cognitive, visual or developmental delay will be excluded as participants. Parent participants should be proficient in English.

Parent/ primary caregivers of pediatric CI recipients will be expected to complete a CI specific parent-proxy, self-administered HRQoL questionnaire:

- *Children with Cochlear Implants: Parental Perspectives (Archbold et al., 2002)*

Added to this validated questionnaire, parents/ primary caregivers will be asked to complete some general questions about cochlear implantation.

Information on demographical variables (such as age at identification of hearing loss, duration of CI use, bilateral/ unilateral implantation etc.) will be captured from patient clinical files additional to questionnaire data.

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Republic of South Africa

Tel: 012 420 2355
Fax: 012 420 3517

bart.vinck@up.ac.za
www.up.ac.za

talita.leroux@up.ac.za

Confidentiality: Identifying data of all participants will not be disclosed and data obtained from clinical patient files will be handled with strict confidentiality. Participants will be assigned an identifying code which will be used for data processing. The anonymity of all participants will be guaranteed at all times.

Data storage: All the research data and/ or documents referring to the above-mentioned study will be stored in the Department of Speech-Language Pathology and Audiology (University of Pretoria). The storage of the mentioned data and/or documents will be maintained for a minimum of 15 years from the commencement of this study.

Written consent and assent: Parents/ primary caregivers will receive this information letter containing the purpose and procedures of the study. Freely given informed consent are required from parents/ primary caregivers. Participation is voluntary and all participants have the right to withdraw from the study at any time without any negative consequences.

Risks: There are no risks associated with this study.

Release of findings: The results of this research study will be published in accredited academic journals, as well as in a summative research report.

If you agree to participate in this study, you are requested to sign this letter of consent.

Please contact us should you require more information. Thank you in advance for your time and co-operation.

Yours sincerely,

Ms Bianca Brewis
Researcher

Mrs Talita le Roux
Supervisor

Prof Bart Vinck
Supervisor
Head: University of Pretoria Cochlear Implant Unit (UPCIU)

Dr Kurt Schlemmer
Supervisor

PERMISSION FOR PARENT/ PRIMARY CAREGIVER TO TAKE PART IN RESEARCH STUDY

Herewith I, _____ (parent/ primary caregiver) of _____ (pediatric cochlear implant recipient) agree to partake in this research study (entitled: *Health-related quality of life in South African children who use cochlear implants)* by completing the attached parent proxy questionnaire on HRQoL.

I give permission to the researcher to have access to my child with a cochlear implant(s)'s clinical records. This information may be used for the purpose of this research study and for publication in scientific literature. I understand that patient confidentiality will be maintained at all times. Should the data obtained for this study be used for future studies, written consent will again be obtained from me.

I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

_____

Signature of parent/ primary caregiver **Date:** _____

Appendix D – Declaration of originality

DECLARATION OF ORIGINALITY

UNIVERSITY OF PRETORIA

The Department of Speech-Language Pathology and Audiology places great emphasis upon integrity and ethical conduct in the preparation of all written work submitted for academic evaluation.

While academic staff teach you about referencing techniques and how to avoid plagiarism, you too have a responsibility in this regard. If you are at any stage uncertain as to what is required, you should speak to your lecturer before any written work is submitted.

You are guilty of plagiarism if you copy something from another author's work (eg a book, an article or a website) without acknowledging the source and pass it off as your own. In effect you are stealing something that belongs to someone else. This is not only the case when you copy work word-for-word (verbatim), but also when you submit someone else's work in a slightly altered form (paraphrase) or use a line of argument without acknowledging it. You are not allowed to use work previously produced by another student. You are also not allowed to let anybody copy your work with the intention of passing if off as his/her work.

Students who commit plagiarism will not be given any credit for plagiarised work. The matter may also be referred to the Disciplinary Committee (Students) for a ruling. Plagiarism is regarded as a serious contravention of the University's rules and can lead to expulsion from the University.

The declaration which follows must accompany all written work submitted while you are a student of the Department of Speech-Language Pathology and Audiology. No written work will be accepted unless the declaration has been completed and attached.

Full names of student: Bianca Brewis

Student number: 13030303

Topic of work: Health-related quality of life in South African children who use cochlear implants

Declaration

1. I understand what plagiarism is and am aware of the University's policy in this regard.

2. I declare that this dissertation (eg essay, report, project, assignment, dissertation, thesis, etc) is my own original work. Where other people's work has been used (either from a printed source, Internet or any other source), this has been properly acknowledged and referenced in accordance with departmental requirements.

3. I have not used work previously produced by another student or any other person to hand in as my own.

4. I have not allowed, and will not allow, anyone to copy my work with the intention of passing it off as his or her own work.

SIGNATURE 

Appendix E – Data storage

UNIVERSITEIT VAN PRETORIA
UNIVERSITY OF PRETORIA
YUNIBESITHI YA PRETORIA

FACULTY OF HUMANITIES
RESEARCH ETHICS COMMITTEE

Declaration for the storage of research data and/or documents

I/ We, the principal researcher(s) Bianca Brewis

and supervisor(s) Mrs Talita Le Roux; Dr Kurt Schlemmer; Professor Bart H.M.E Vinck

of the following study, titled Health-related quality of life in South African children who use cochlear implants

will be storing all the research data and/or documents referring to the above-mentioned study in the following department: Speech-Language Pathology and Audiology

We understand that the storage of the mentioned data and/or documents must be maintained for a minimum of 15 years from the commencement of this study.

Start date of study: January 2017

Anticipated end date of study: November 2017

Year until which data will be stored: 2032

Name of Principal Researcher(s)	Signature	Date
Bianca Brewis	JBrewis	2017-02-02

Name of Supervisor(s)	Signature	Date
Mrs Talita Le Roux		2017-02-03
Dr Kurt Schlemmer		

Name of Head of Department	Signature	Date
Professor Bart H.M.E Vinck		3/2/2017

Appendix F – Children with Cochlear Implants: Parental perspective questionnaire (CCIPP)

Girl version

Children with cochlear implants: Parental perspectives

Devised by Sue Archbold and Mark Lutman

Child's Name:

We would like your help in completing this questionnaire

We recognize that you are in the best position to describe what cochlear implants have meant to your child and family. The questionnaire consists of statements with which you can agree or disagree. You are asked to tick one of five boxes to indicate your opinion: *strongly agree, agree, neither agree nor disagree, disagree, strongly disagree.*

Please give your initial response to the statements rather than thinking about each one for a long time.

Please tick only one box per statement and do not leave any unanswered. If any statement does not apply to your situation, please write N/A (meaning not applicable)

Your name: (person filling in the form)

Your relationship with child: (father, guardian, etc)

Date of implantation:

Today's date:

Parent's views and perspectives

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
1 Communication is difficult even with people she knows well.	☐	☐	☐	☐	☐
2 Immediately after implantation her ability to communicate was poorer.	☐	☐	☐	☐	☐
3 The help I give her has become more productive now she has her implant.	☐	☐	☐	☐	☐
4 Before implantation she obtained no benefit at all from her hearing aids.	☐	☐	☐	☐	☐
5 She does not have a close relationship with her grandparents.	☐	☐	☐	☐	☐
6 She is totally reliant on her implant all the time.	☐	☐	☐	☐	☐
7 She knows when I want her attention because she can hear me call.	☐	☐	☐	☐	☐
8 I worry that the implant will break down.	☐	☐	☐	☐	☐
9 She is unable to cope with mainstream schooling.	☐	☐	☐	☐	☐
10 It has been a problem getting someone to look after the family when we go to the Implant Centre.	☐	☐	☐	☐	☐
11 Progress during the first few months seemed very slow.	☐	☐	☐	☐	☐
12 I can seldom leave her to do something on her own.	☐	☐	☐	☐	☐

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
26 Progress after implantation has exceeded my expectations.	☐	☐	☐	☐	☐
27 We can now chat even when she cannot see my face.	☐	☐	☐	☐	☐
28 Making the decision to proceed with implantation was the most difficult part for me.	☐	☐	☐	☐	☐
29 It was a difficult time waiting for the assessments before implantation.	☐	☐	☐	☐	☐
30 She was socially isolated before getting her implant.	☐	☐	☐	☐	☐
31 The local school and support services adequately meet all our needs concerning the use of her implant.	☐	☐	☐	☐	☐
32 A significant change has been improvement in her confidence.	☐	☐	☐	☐	☐
33 She was very dependent on us before the implantation.	☐	☐	☐	☐	☐
34 We feel the need for advice from the Implant Centre concerning her future.	☐	☐	☐	☐	☐
35 She can amuse herself listening to music or watching TV or playing games.	☐	☐	☐	☐	☐
36 We are reliant on the Implant Centre for technical advice about her implant.	☐	☐	☐	☐	☐
37 I am concerned about her future school placement.	☐	☐	☐	☐	☐
38 The process of implantation was no more intrusive that expected.	☐	☐	☐	☐	☐

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
39 She does not make friends easily outside the family.	☐	☐	☐	☐	☐
40 It is essential that she is encouraged to wear the processor at all time.	☐	☐	☐	☐	☐
41 She is sociable within the family.	☐	☐	☐	☐	☐
42 A positive attitude is a great help towards successful use of the implant.	☐	☐	☐	☐	☐
43 Regular tuning and checking of the implant system are essential.	☐	☐	☐	☐	☐
44 At least one visit per year by Implant Centre staff to school is essential.	☐	☐	☐	☐	☐
45 She shares in family situations more than before implantation.	☐	☐	☐	☐	☐
46 Before proceeding with implantation, parents should obtain as much information and advice as possible.	☐	☐	☐	☐	☐
47 She is as independent as most other children of her age.	☐	☐	☐	☐	☐
48 Parents should have a choice in the use of sign language at school.	☐	☐	☐	☐	☐
49 It was useful to meet another family with an implanted child before deciding on an implant.	☐	☐	☐	☐	☐
50 I am happy about her progress at school	☐	☐	☐	☐	☐
51 I can now let her play outside as she is aware of the sound of traffic.	☐	☐	☐	☐	☐

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
52 The most important factor in choosing an implant device is its reliability.	☐	☐	☐	☐	☐
53 She is still unable to cope in new situations.	☐	☐	☐	☐	☐
54 I am confident that long-term electrical stimulation will not be a problem.	☐	☐	☐	☐	☐
55 The whole process of implantation is still stressful.	☐	☐	☐	☐	☐
56 I expected her to learn to talk once she had her implant.	☐	☐	☐	☐	☐
57 I worry that ultimately she may be neither part of the hearing or deaf world.	☐	☐	☐	☐	☐
58 It was important to me that my child could hear sounds from traffic for safety reasons.	☐	☐	☐	☐	☐
59 Her behaviour has improved since her implant	☐	☐	☐	☐	☐
60 I believe now that my child will have reasonable prospects for employment.	☐	☐	☐	☐	☐
61 She has become argumentative since getting her implant.	☐	☐	☐	☐	☐
62 A parent of a child with an implant needs to be patient as benefits may take time to show.	☐	☐	☐	☐	☐
63 It has been hard to take time off work for the appointments at the Implant Centre.	☐	☐	☐	☐	☐

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
64 She is less frustrated than before she had the implant.	☐	☐	☐	☐	☐
65 She takes part in family relationships on an even footing with other members.	☐	☐	☐	☐	☐
66 I find it easier to communicate with her by speaking than by signing.	☐	☐	☐	☐	☐
67 I give the same amount of help as before her implant.	☐	☐	☐	☐	☐
68 I chose implantation for my child so she would have a chance to become part of the hearing world.	☐	☐	☐	☐	☐
69 She is totally reliant on her implant at school.	☐	☐	☐	☐	☐
70 She continues to be a happy child and good fun to be with.	☐	☐	☐	☐	☐
71 Her use of spoken language has developed greatly.	☐	☐	☐	☐	☐
72 Now she is talkative and engages others in conversation.	☐	☐	☐	☐	☐
73 Other children in the family resented the time and attention taken up by the implant.	☐	☐	☐	☐	☐
	(Tick here ( ) if no other children)				
74 Her relationship with brothers and sisters has improved. (Tick here ( ) if no other children)	☐	☐	☐	☐	☐

Thank you very much for taking the time to complete this questionnaire

Boy version

Children with cochlear implants: parental perspectives

Devised by Sue Archbold and Mark Lutman.
Development support by The Royal National Institute for Deaf People.

Child's name

We would like your help in completing this questionnaire.

We recognise that you are in the best position to describe what cochlear implants have meant to your child and family.

This questionnaire is designed for parents at least one year after implantation. It consists of statements with which you can agree or disagree. You are asked to tick one of five boxes to indicate your opinion: *strongly agree, agree, neither agree nor disagree, disagree, strongly disagree.*

The statements are based on interviews with parents who have a child with a cochlear implant.

Please give your initial response to the statements rather than thinking about each one for a long time.

Please tick only one box per statement and do not leave any unanswered. If any statement does not apply to your situation, for example if there are no brothers or sisters, please write 'N/A' (meaning not applicable) so that we know you haven't overlooked it.

Thank you very much for taking the time to complete the questionnaire.

Parents' views and experiences

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strong disagr
1. Communication is difficult even with people he knows well.	☐	☐	☐	☐	☐
2. Immediately after implantation his ability to communicate was poorer.	☐	☐	☐	☐	☐
3. The help I give him has become more productive now he has his implant.	☐	☐	☐	☐	☐
4. Before implantation he obtained no benefit at all from his hearing aids.	☐	☐	☐	☐	☐
5. He does not have a close relationship with his grandparents.	☐	☐	☐	☐	☐
6. He is totally reliant on his implant all the time.	☐	☐	☐	☐	☐
7. He knows when I want his attention because he can hear me call.	☐	☐	☐	☐	☐
8. I worry that the implant will break down.	☐	☐	☐	☐	☐
9. He is unable to cope with mainstream schooling.	☐	☐	☐	☐	☐
10. It has been a problem getting someone to look after the family when we go to the Implant Centre.	☐	☐	☐	☐	☐
11. Progress during the first few months seemed very slow.	☐	☐	☐	☐	☐
12. I can seldom leave him to do something on his own.	☐	☐	☐	☐	☐
13. The programme at the Implant Centre should emphasise speaking and listening.	☐	☐	☐	☐	☐
14. I worry that he will blame me for my decision for him to have an implant.	☐	☐	☐	☐	☐
15. He has needed more help from me since he received his implant.	☐	☐	☐	☐	☐
16. He still shows signs of frustration in his behaviour.	☐	☐	☐	☐	☐
17. I am concerned that my child will be rejected by the Deaf Community because of the implant.	☐	☐	☐	☐	☐
18. The quality of his speech gives me cause for concern.	☐	☐	☐	☐	☐
19. A lot of help at first means a child needs less help later.	☐	☐	☐	☐	☐

Parents' views and experiences

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
20. I get more time to myself because of his increased independence.	☐	☐	☐	☐	☐
21. Only experienced teams should carry out cochlear implantation.	☐	☐	☐	☐	☐
22. The costs of travel to the Implant Centre are a problem.	☐	☐	☐	☐	☐
23. He is keeping up well with children of his own age at school.	☐	☐	☐	☐	☐
24. Signing support is helpful for a considerable time after implantation.	☐	☐	☐	☐	☐
25. I wish to participate in meetings with other families who have an implanted child.	☐	☐	☐	☐	☐
26. Progress after implantation has exceeded my expectations.	☐	☐	☐	☐	☐
27. We can now chat even when he cannot see my face (for example in the car or in the dark).	☐	☐	☐	☐	☐
28. Making the decision to proceed with implantation was the most difficult part for me.	☐	☐	☐	☐	☐
29. It was a difficult time waiting for the results of the assessments before implantation.	☐	☐	☐	☐	☐
30. He was socially isolated before getting his implant.	☐	☐	☐	☐	☐
31. The local school and support services adequately meet all our needs concerning the use of his implant at school.	☐	☐	☐	☐	☐
32. A significant change has been improvement in his confidence.	☐	☐	☐	☐	☐
33. He was very dependent on us before the implantation.	☐	☐	☐	☐	☐
34. We feel the need for advice from the Implant Centre concerning his future.	☐	☐	☐	☐	☐
35. He can now amuse himself listening to music or watching TV or playing games.	☐	☐	☐	☐	☐
36. We are reliant on the Implant Centre for technical advice about his implant.	☐	☐	☐	☐	☐
37. I am concerned about his future school placement.	☐	☐	☐	☐	☐

Parents' views and experiences

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strong disagr
38. The process of implantation was no more intrusive than expected.	☐	☐	☐	☐	☐
39. He does not make friends easily outside the family.	☐	☐	☐	☐	☐
40. It is essential that he is encouraged to wear the processor all the time.	☐	☐	☐	☐	☐
41. He is sociable within the family.	☐	☐	☐	☐	☐
42. A positive attitude is a great help towards successful use of the implant.	☐	☐	☐	☐	☐
43. Regular tuning and checking of the implant system are essential.	☐	☐	☐	☐	☐
44. At least one visit per year by Implant Centre staff to home/school is essential.	☐	☐	☐	☐	☐
45. He shares in family situations more than before implantation.	☐	☐	☐	☐	☐
46. Before proceeding with implantation, parents should obtain as much information and advice as possible.	☐	☐	☐	☐	☐
47. He is as independent as most other children of his age.	☐	☐	☐	☐	☐
48. Parents should have a choice in the use of sign language at school.	☐	☐	☐	☐	☐
49. It was useful to meet another family with an implanted child before deciding on an implant.	☐	☐	☐	☐	☐
50. I am happy about his progress at school.	☐	☐	☐	☐	☐
51. I can now let him play outside as he is aware of the sound of traffic.	☐	☐	☐	☐	☐
52. The most important factor in choosing an implant device is its reliability.	☐	☐	☐	☐	☐
53. He is still unable to cope in new situations.	☐	☐	☐	☐	☐
54. I am confident that long-term electrical stimulation will not be a problem.	☐	☐	☐	☐	☐
55. The whole process of implantation is still stressful.	☐	☐	☐	☐	☐
56. I expected him to learn to talk once he had his implant.	☐	☐	☐	☐	☐

Parents' views and experiences

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strong disagr
57. I worry that ultimately he may be neither part of the deaf nor the hearing world.	☐	☐	☐	☐	☐
58. It was important to me that my child could hear sounds from traffic for safety reasons.	☐	☐	☐	☐	☐
59. His behaviour has improved since he had his implant.	☐	☐	☐	☐	☐
60. I believe now that my child will have reasonable prospects for employment.	☐	☐	☐	☐	☐
61. He has become argumentative since getting his implant.	☐	☐	☐	☐	☐
62. A parent of a child with an implant needs to be patient as benefits may take time to show.	☐	☐	☐	☐	☐
63. It has been hard to take time off work for the appointments at the Implant Centre.	☐	☐	☐	☐	☐
64. He is less frustrated than before he had the implant.	☐	☐	☐	☐	☐
65. He takes part in family relationships on an equal footing with other members.	☐	☐	☐	☐	☐
66. I find it easier to communicate with him by speaking than by signing.	☐	☐	☐	☐	☐
67. I give the same amount of help as before his implant.	☐	☐	☐	☐	☐
68. I chose implantation for my child so he would have a chance to become part of the hearing world.	☐	☐	☐	☐	☐
69. He is totally reliant on his implant at school.	☐	☐	☐	☐	☐
70. He continues to be a happy child and good fun to be with.	☐	☐	☐	☐	☐
71. His use of spoken language has developed greatly.	☐	☐	☐	☐	☐
72. Now he is talkative and engages others in conversation.	☐	☐	☐	☐	☐
73. Other children in the family resented the time and attention taken up by the implant. (Tick here ☐ if no other children.)	☐	☐	☐	☐	☐
74. His relationship with brothers and sisters has improved. (Tick here ☐ if no other children.)	☐	☐	☐	☐	☐

Thank you very much for taking the time to complete this questionnaire

Appendix G – Click to accept on Survey Monkey

Girl version

Children with cochlear implants: Parental perspectives (girl)

Informed consent

Permission to partake in study (please see email regarding informed consent).

* 1. Herewith I, parent/ primary caregiver of a child with a cochlear implant(s) agree to partake in this research study: by completing two **parent-report** questionnaires on Health-Related Quality of Life (HRQoL). I give permission that my child with a cochlear implant(s) may also partake in this research study by completing the **self-report** HRQoL for children with cochlear implants. I also give permission that my other child/ children may partake in this research by completing the **sibling-report** HRQoL questionnaire.

I give permission to the researcher to have access to my child with a cochlear implant(s)'s clinical records. This information may be used for the purpose of this research study and for publication in scientific literature. I understand that patient confidentiality will be maintained at all times.

Should the data obtained for this study be used for future studies, written consent will again be obtained from me.

I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

Herewith I give consent to take part in this research study. I also give consent that my child with a cochlear implant(s) and his/ her siblings take part in this study.

- ◯ Yes
- ◯ No

Boy version

Children with cochlear implants: Parental perspectives (boy)

Informed consent

Permission to partake in study (please see email regarding informed consent).

* 1. Herewith I, parent/ primary caregiver of a child with a cochlear implant(s) agree to partake in this research study: by completing one **parent-report** questionnaire on Health-Related Quality of Life (HRQoL).

I give permission to the researcher to have access to my child with a cochlear implant(s)'s clinical records. This information may be used for the purpose of this research study and for publication in scientific literature. I understand that patient confidentiality will be maintained at all times. Should the data obtained for this study be used for future studies, written consent will again be obtained from me.

I have received the necessary information about this study and I have had the opportunity to ask questions regarding this project.

Herewith I give consent to take part in this research study.

- ◯ Yes
- ◯ No

Appendix H – Variables captured from patient files/ clinical records

Continuous variables captured:

Current age (age at study)

Age at 1st CI

Age at 2nd CI

Duration of CI use

Age at diagnosis of hearing loss

Age at diagnosis of deafness

Duration of deafness

Categorical variables captured:

Cochlear implant program

Gender

School environment

Unilateral/bilateral cochlear implantation

Communication mode

Etiology of hearing loss

Pre/post-lingually deafened

Relationship to child with CI

Parental education

Other siblings

Other children with hearing loss

Variables provided by cochlear implantation units, namely, PCIU, JCIC and DCIP:

Age at 1st CI

Age at 2nd CI

Duration of CI use

Age at diagnosis of hearing loss

Age at diagnosis of deafness

Duration of deafness

Cochlear implant program

Gender

Etiology of hearing loss

Pre/post-lingually deafened

Variables provided by the parents:

Current age

School environment

Communication mode

Unilateral/bilateral cochlear implantation

Relationship to child with CI

Parental education

Other siblings

Other children with hearing loss

Appendix I – Proof of submission to International Journal of Audiology

From: **International Journal of Audiology**
<onbehalfof@manuscriptcentral.com>
Date: Wed, 23 May 2018 at 12:40
Subject: International Journal of Audiology - Manuscript ID TIJA-2018-05-0144
To: <talita.leroux@up.ac.za>, <gpleroux1@gmail.com>

MS: Health-related quality of life in South-African children who use cochlear implants
MS#: TIJA-2018-05-0144

23-May-2018

Dear Dr. le Roux:

This letter will acknowledge the successful online submission of the above listed manuscript to the International Journal of Audiology. The manuscript will soon be forwarded to an associate editor to oversee the review and obtain editorial comments from expert reviewers. Should you have questions, feel free to contact the editorial office (editor-ija@utdallas.edu). Please reference the manuscript number in any correspondence. You can also view the status of your manuscript at any time by checking your Author Center after logging in to https://mc.manuscriptcentral.com/tija.

It is the goal of the International Journal of Audiology to provide each manuscript with a comprehensive and expeditious peer review. You will be contacted upon completion of the initial review.

Thank you for considering IJA for your submission.

Sincerely,

Ross J. Roeser, Ph.D.
Editor-in-Chief