

Childhood hearing loss profile, decentralised screening and outcomes in the Western Cape public healthcare system, South Africa

by

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Silva Kuschke has obtained, for the research described in this work, the appropriate research ethics approval.

The author declares that she has observed the ethical standards required in terms of the University of Pretoria's code of ethics for researchers and the policy guidelines for responsible research.

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PLAGIARISM DECLARATION

- Full name: Silva Kuschke
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- Degree: PhD: Audiology
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I declare that this thesis is my own original work. Where secondary material is used, it has been carefully acknowledged and referenced in accordance with university requirements.

I understand what plagiarism is and am aware of university policy and implications in this regard.

Mausina.

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02 December 2021

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Childhood hearing loss is a global epidemic most prevalent in low- and middle-income countries (LMIC) where hearing healthcare services are often inaccessible. The aim of this research project was to describe the profile of childhood hearing loss, to explore a decentralised model of hearing healthcare through district hearing screening, and to describe the hearing aid outcomes of children with bilateral sensorineural hearing loss in the Western Cape public healthcare system, South Africa.

Due to the limited availability of hearing screening programmes and poor data capturing within existing programmes, the nature and associated risk profile of childhood hearing loss in South Africa is largely unknown. Study I of this research project aimed to provide one of the first reports on the profile of childhood hearing loss in the Western Cape Province of South Africa, by describing the nature, associated risk factors, and age of diagnosis for childhood hearing loss in a cohort from the Red Cross War Memorial Children's Hospital (RCWMCH). A retrospective review of clinical data from children under six years with confirmed hearing loss at RCWMCH between 1 January 2019 and 31 July 2019 was conducted. Descriptive statistics were used to analyse data for a sample of 240 children (mean age of 42 months; 21.8 SD; range 2 - 72). More than two thirds (68.3%) of the children presented with bilateral hearing loss. The majority presented with conductive hearing loss (64.6%), followed by

sensorineural (28.7%) and mixed hearing loss (3.3%) or auditory neuropathy spectrum disorder (3.3%). More than half (51.8%) of the bilateral sensorineural hearing losses were profound. The most prominent risk factor for conductive hearing loss was otitis media, for sensorineural hearing loss it was a family history of childhood hearing loss, and for auditory neuropathy spectrum disorder it was hyperbilirubinaemia. Approximately one third of patients (27.1%) with sensorineural hearing loss did not have any associated risk factors. The mean age of diagnosis of permanent congenital or early-onset hearing loss was 31.4 months (22.8 SD; range 2 - 72), with a mean delay of nine months (13.2 SD; range 0 - 60) between age of suspicion and diagnosis of hearing loss (n = 93). The results of Study I highlighted that infant hearing screening services in the public health sector of South Africa should be prioritised alongside primary health care efforts to reduce preventable risks for hearing loss. Age of diagnosis of permanent congenital or early-onset hearing loss was severely delayed, undermining prospects of positive outcomes through early intervention.

In low-resourced contexts, referrals to central hospitals for primary care services like hearing screening add to growing waiting lists for specialised care. Long waiting times delay care for the time-sensitive treatment of childhood hearing loss. Study II aimed to compare a centralised tertiary model of hearing healthcare to a decentralised model through district hearing screening for children in the Western Cape Province of South Africa. A pragmatic quasi-experimental study design, with a seven-month control period of standard service provision at a tertiary hospital (June to December 2018) and a seven-month intervention period where a new hearing screening service, utilising oto-acoustic emissions, was introduced at a district hospital (June to December 2019). The effect of decentralising hearing healthcare for the intervention period was measured by attendance rates for initial hearing screening, patient travelling distance, number of referrals to a tertiary-level hospital, and hearing outcomes. Children referred to the tertiary hospital for initial hearing screening from the district hospital catchment area, and who attended their appointments, were included in the study (315 in the tertiary hospital group and 158 in the district hospital group). Data were collected from patient records and an electronic database at the Department of Audiology at the tertiary hospital and analysed with a combination of descriptive and inferential statistical methods. During the decentralised hearing

screening project for Study II, attendance rates during the intervention period at the district hospital were significantly higher than attendance rates at the tertiary hospital during the same period one year prior to the intervention (p < 0.001). Travel distance for 158 patients to the district hospital was significantly shorter compared to the tertiary hospital (p < 0.001). Number of referrals to the tertiary hospital for initial hearing screening decreased significantly during the intervention period (p < 0.001). The majority of paediatric patients in both the tertiary and district groups (78.7% and 80.4%, respectively) passed initial hearing screening via oto-acoustic emissions bilaterally. Diagnostic assessment results indicated mild conductive hearing loss for the majority of patients in both groups. Decentralised hearing screening should be conducted at the appropriate level of care to increase access to hearing healthcare, reduce patient travelling distances and associated costs, and reduce the burden on tertiary-level hospitals.

Measuring hearing aid outcomes in children is a complex process because no single measurement exists to determine outcomes on the multidimensional aspects of auditory behaviour in children. This process becomes even more complicated due to barriers such as lack of standardised outcomes assessment tools in a multi-lingual and multi-cultural context typical of most South African children. Study III described hearing aid outcomes and potential factors associated with hearing aid use in South African children with bilateral sensorineural hearing loss (SNHL) accessing the public health care system. A retrospective review of clinical data and caregiver reported outcomes of children aged 0 - 13 years with bilateral SNHL at one-month and threemonths post-hearing aid fitting was used for this study. Oral/aural performance was measured by the *Parents' Evaluation of Aural/Oral Performance of Children* (PEACH) questionnaire. Multiple linear regression was used to evaluate factors associated with hearing aid use. Thematic analysis was applied for qualitative caregiver-reported outcomes. Sixty-eight children with confirmed bilateral SNHL, who were fitted with binaural air-conduction hearing aids at RCWMCH between January 2017 and December 2019, were included in Study III. Average daily hearing aid use increased significantly (p < 0.05) from one-month (5.0; 3.0 SD; range 0.3 - 14.0) to three-months post-fitting (5.9; 3.4 SD; range 1.1 – 16.8). Average PEACH scores were higher in Quiet (73.4%) than in Noise (69.6%). More than half (52.2%) of children required

review based on their overall percentage PEACH scores. Higher average daily hearing aid use was significantly associated with higher overall PEACH scores (p < 0.05). Neuro-typically developing children had significantly higher hearing aid use than children with additional disabilities (p < 0.001). Qualitative caregiver feedback revealed themes pertaining to advantages and barriers to hearing aid use. Outcomes of children with SNHL fitted with binaural hearing aids at RCWMCH demonstrated increased average daily hearing aid use from one-month to three-months post-fitting. Children with additional disabilities had significantly poorer hearing aid use and aural/oral performance requiring more support for this vulnerable group to realize sufficient benefit from hearing aid use.

The results of this research project highlighted that preventable causes of childhood hearing loss were very common in a cohort from the Western Cape Province, and age of diagnosis of hearing loss was severely delayed. Newborn hearing screening and timeous treatment for preventable causes of hearing loss at primary level healthcare need to be prioritised in South Africa to minimise adverse effects on speech- and language development in children. Lack of access to hearing healthcare services remains a barrier in LMICs like South Africa. Strategies, like decentralisation, are feasible (as demonstrated by Study II), and should be implemented to improve equitable access for vulnerable children. Populations who are at risk for poorer hearing aid use, like children with additional disabilities, should be identified and receive support from a multi-disciplinary team to ensure sufficient benefit from hearing aids. In South Africa, it is important to address barriers to timeous and accurate diagnoses of childhood hearing loss, to improve access to hearing healthcare, and to ultimately provide successful intervention with hearing technology.

KEYWORDS

Age of diagnosis Childhood hearing loss Data logging Decentralisation Hearing aid outcomes Hearing healthcare Low-and middle-income countries Oto-acoustic emissions PEACH questionnaire Risk factors Sensorineural hearing loss

ABBREVIATIONS

ABR	Auditory Brainstem Response
ANSD	Auditory Neuropathy Spectrum Disorder
AOM	Acute Otitis Media
CHL	Conductive Hearing Loss
CSOM	Chronic Suppurative Otitis Media
dBHL	Decibel Hearing Level
ENT	Ear-Nose-Throat
HPCSA	Health Professions Council of South Africa
Hz	Hertz
kHz	Kilohertz
LMIC	Low- and Middle-income Countries
MOU	Maternal and Obstetric Unit
NICU	Neonatal Intensive Care Unit
OAE	Oto-acoustic Emissions
OM	Otitis Media
PEACH	Parents' Evaluation of Aural/Oral Performance of Children
RCWMCH	Red Cross War Memorial Children's Hospital
REAR	Real-ear Aided Response
RECD	Real-ear to Coupler Difference
SASL	South African Sign Language
SNHL	Sensorineural Hearing Loss
ТВ	Tuberculosis
USD	United States Dollar
VLBW	Very Low Birth Weight
WHO	World Health Organization
YLD	Years Lived with Disability

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CHAPTER 1

INTRODUCTION

1.1 Introduction

Globally, an estimated 466 million people, of which 34 million are children, suffer from disabling hearing loss (World Health Organization [WHO], 2021). Hearing loss is the second most prevalent developmental disability, affecting approximately 15.5 million children under the age of five years globally (Olusanya et al., 2018), and is the third largest cause of global Years Lived with Disability (YLD) (Haile, Orji, Briant, Adelson, Davis, & Vos, 2020). Approximately 95% of children with developmental disabilities reside in low- and middle-income countries (LMICs) (Olusanya et al., 2018). The WHO estimates the prevalence of hearing loss for children aged between five and 14 years at 1.9% in sub-Saharan Africa as opposed to 0.4% in high-income countries (WHO, 2013).

Sub-Saharan Africa has one of the highest prevalence rates of hearing loss, with an estimated 10.3 million children under the age of 10 years who suffer from permanent, disabling hearing loss (Olusanya et al., 2020). The estimated global cost within the education sector for providing support to children with hearing loss (aged 5 - 14 years) is 27 billion USD annually (WHO, 2021). Undetected and untreated hearing loss has a major negative impact on a child's speech, language, cognitive, educational, and socio-emotional development (Joint Committee on Infant Hearing [JCIH], 2019).

1.2 Childhood hearing loss

Most cases of disabling childhood hearing loss have preventable causes that are common in LMICs and make up nearly 60% of the aetiology of hearing loss in children (WHO, 2021). The WHO has categorised prevention in three tiers: primary prevention to avoid an adverse health condition; secondary prevention to detect a condition at an early stage and to treat it promptly; and tertiary prevention to reduce the impact of an

established condition and to restore function to the maximum extent possible (WHO, 2021). Children born into lower socioeconomic contexts have a higher incidence of middle ear pathology and subsequent preventable hearing loss, as well as considerably less access to non-emergency health resources (Epstein, Grant, Schiff, & Kasehagen, 2009).

Adverse pre-, peri-, and post-natal conditions are prominent risk factors for childhood hearing loss, especially in LMICs (Tharpe & Seewald, 2016). Higher rates of low birth weight and severe hyperbilirubinaemia, which are associated risk factors for childhood hearing loss, have been reported in LMICs (Olusanya, 2015). Childhood hearing loss may be associated with common infectious diseases found in LMICs, such as HIV/AIDS, malaria, and tuberculosis (TB) (Assuiti et al., 2013; Hrapcak et al., 2016; Zhao & Mackenzie, 2011). Vaccine-preventable infections which are associated with hearing loss, and occur commonly in LMICs, include rubella, measles, mumps, and meningitis (Caroça et al., 2017; WHO, 2018). Routine newborn and childhood immunization programmes are often rudimentary in many LMICs (Harris and Dodson, 2017; Van den Heever, 2016). In the African region, only 7% of African countries vaccinated against rubella in 2012 (WHO, 2013). The incidence of meningitis is the highest in sub-Saharan African countries and is often referred to as the sub-Saharan meningitis belt (Pitkäranta et al., 2007). Some meningitis strains are becoming resistant to penicillin-based antibiotics, which gives rise to a greater use of aminoglycosides as first line treatment (WHO, 2013). An effective vaccination became available in 2000, which means that meningitis is gradually being eliminated from more developed regions but continues to rage in LMICs (Vella & Pace, 2015).

Possible reasons for the difference in prevalence of hearing loss in high-income countries and LMICs include the absence of well-managed hearing screening programmes, the impact of poverty and malnutrition on hearing, lack of awareness of hearing loss and its devastating effects in children, and limited access to hearing healthcare in LMICs (WHO, 2021). Furthermore, the proportion of hearing loss attributed to post-natal causes, such as infectious diseases and middle ear disease, is typically higher in LMICs (Olusanya et al., 2020).

Due to the limited availability of hearing screening programmes, as well as poor data capturing and management within existing programmes (Meyer, Swanepoel, Le Roux, & van der Linde, 2012), the nature and associated risk profile of childhood hearing loss in South Africa is largely unknown. Apart from studies from nearly four decades ago conducted in schools for the deaf (Sellars & Beighton, 1983), only preliminary data on the nature and associated risk profile of childhood hearing loss in South Africa are available (Le Roux, Swanepoel, Louw, Vinck, & Tshifularo, 2015; Swanepoel, Johl, & Pienaar, 2013). At the time of these early aetiological reports, diagnostic categories of hearing loss did not include auditory neuropathy spectrum disorder (ANSD). In addition, with the advent of newborn hearing screening programmes, the risk profiles for permanent childhood hearing loss were extended and described more accurately (Olusanya, 2011). This was not accounted for in early South African reports (Sellars & Beighton, 1983; Sellars & Beighton, 1978; Sellars, Groeneveldt, & Beighton, 1976). The current risk profile of South African children with hearing loss includes admittance to the Neonatal Intensive Care Unit (NICU), hyperbilirubinaemia, family history of childhood hearing loss, prematurity, and meningitis (Le Roux, et al., 2015; Swanepoel et al., 2013). Within the South African context, it is not always possible to ascertain the exact aetiology of hearing loss in children, however, risk factors or aetiological factors can be documented instead (Lebeko, Bosch, Noubiap, Dandara, & Wonkam, 2015).

Information regarding unique risk factors for childhood hearing loss in LMICs is important so that infants may be referred for early hearing screening to identify hearing loss timeously (Olusanya, 2011). *Study I aimed to provide one of the first reports on the profile of childhood hearing loss* in the Western Cape Province of South Africa, by describing the nature, associated risk factors, and age of diagnosis for childhood hearing loss in a cohort from the Red Cross War Memorial Children's Hospital (RCWMCH).

1.3 Early detection of childhood hearing loss in LMICs

Early detection of hearing loss is imperative as the first step to accurate diagnosis and subsequent timeous intervention (Swanepoel, Storbeck, & Friedland, 2009; WHO, 2021). A major challenge in the appropriate and timeous management of childhood hearing loss in LMICs is delayed presentation and late diagnosis. A study conducted in Nigeria revealed that only 16.6% of hearing-impaired children presented to audiological services within the first year of onset while over 41% presented after five years (Adedeji, Tobih, Sogebi, & Daniel, 2015). A study conducted in South Africa reported that 47% of diagnoses of childhood hearing loss were made after 36 months of age despite initial parental suspicion of hearing loss before 12 months of age in about 40% of children (Swanepoel et al., 2013). A delay of up to 18 months between caregiver suspicion and diagnosis of childhood hearing loss has been reported in another study in South Africa (Van der Spuy & Pottas, 2008).

The delay in seeking help, late diagnosis of hearing loss, and poor access to intervention may be attributed to lack of education and misconceptions about the causes of hearing loss (Nikolopoulos, 2015; Swanepoel & Almec, 2008; Tucci, Merson, & Wilson, 2010). Where there are low levels of parental education as well as low socioeconomic status, the risk of poor follow-up rates for hearing assessments and subsequent intervention is higher in families who need to travel greater distances (Cavelcanti & Guerra, 2012). A systematic review including reports from various countries assessed barriers to follow-up after newborn hearing screening. The review postulated that the foremost reasons for poor follow-up rates were low levels of parental education, travel distance, employment responsibilities, stigmatic attitude towards hearing loss, and competing healthcare needs (Ravi et al., 2016). More than 90% of deaf children are born to hearing parents, therefore many parents do not have supportive peers who have experience in the complex process of hearing loss diagnoses and intervention (Mitchell & Karchmer, 2004).

Newborn hearing screening is often only available in certain parts of the country or at certain primary-level health clinics and hospitals in South Africa. It is estimated that

less than 10% of the approximate one million babies born annually in South Africa will have access to hearing screening services (Meyer et al., 2012). This estimate implies that most South African children with congenital or early-onset hearing loss will likely not receive early auditory rehabilitation which is required for the acquisition of speech and language milestones (Meyer et al., 2012; Theunissen & Swanepoel, 2008). A 2008 survey of hearing screening services in public hospitals in South Africa indicated that less than 7.5% of public hospitals offered any neonatal hearing screening services (Theunissen & Swanepoel, 2008). This small percentage is particularly concerning, as the public healthcare system services approximately 85% of the South African population (National Treasury Department, Republic of South Africa, 2007). In a study on the risk profiles of South African children with profound hearing loss, results indicated that approximately 73% of children did not receive newborn hearing screening (Le Roux et al., 2015), which impedes early detection, diagnosis, and enrolment in early intervention services for a large number of children in South Africa.

There are several barriers to the implementation of newborn hearing screening programmes in LMICs: screening equipment for oto-acoustic emissions (OAEs) or auditory brainstem response (ABR) is expensive to purchase, interpretation of the results requires training and expertise, and often audiologists and healthcare workers are unavailable in rural locations (Mupawose, 2009; Swanepoel et al., 2010). Early discharge of neonates from hospital (< 24 hrs), is common practice in many LMICs, which makes it challenging to complete newborn hearing screening timeously (van Dyk, Swanepoel, & Hall, 2015). Hearing screening through community-based midwife obstetric units following hospital discharge has been trialled in South Africa (De Kock, Swanepoel, & Hall, 2016), and training to enable nurses and other community-workers to assist with the burden of primary prevention would extend the reach of hearing healthcare professionals in LMICs (Crisp & Chen, 2014).

The Joint Committee on Infant Hearing (JCIH, 2019) has established guidelines to promote early detection, accurate diagnosis, and timeous intervention for children with hearing loss. These guidelines stipulate that all infants should be screened no later than one month after birth, diagnosis of hearing loss should occur before three months

of age, and intervention for hearing loss should commence before six months of age (JCIH, 2019). The large number of births occurring outside of hospitals in many LMICs, as well as the many barriers in terms of access to follow-up services, means that the JCIH guidelines are helpful but need to be supplemented by local investigations and reports (Olusanya, Okhakhu, & Somefun, 2011).

1.4 Service delivery for children with hearing loss in LMICs

The implications of hearing loss in LMICs are far-reaching. Poverty-stricken communities have greater exposure to health risks, and limited access to health services, which predispose them to disease and resultant complications such as sensory impairment (WHO, 2021). People with sensory impairment have greater healthcare costs, and decreased income, which pushes them further into poverty. This cycle affects individuals, families, communities, and even countries (WHO, 2013; WHO, 2021).

Hearing healthcare services in LMICs are not prioritised by health systems that are overwhelmed by life-threatening diseases (WHO, 2021; Swanepoel, et al., 2010). Poor hearing health infrastructure and resources (personnel and equipment), as well as geographical barriers such as distance, leads to limited accessibility of hearing healthcare services (Swanepoel et al., 2010; WHO, 2021). The risk of poor follow-up rates for hearing assessments and timely intervention is higher in families who need to travel greater distances (Cavalcanti & Guerra, 2012; Ravi, et al., 2016). Compared to high-income countries, LMICs have an unequal proportion of hearing loss burden, as well as a limited number of well-trained hearing healthcare professionals (Harris & Dodson, 2017). The number of audiologists and Ear-Nose-Throat (ENT) specialists is reported to be lowest in African countries, with an average estimate of one audiologist for every 0.8 million people and one ENT specialist for every 1.2 million people in sub-Saharan Africa (Mulwafu, Ensink, Kuper, & Fagan, 2017). Over a 10-year period, between 2005 and 2015, there has been no substantial improvement in these numbers (Mulwafu et al., 2017). Barriers to increasing and maintaining the supply include limited funding for education of these professionals, relocation of trained professionals

to high-income countries, low remuneration, and lack of a career path for hearing healthcare professionals (Wilson, Tucci, Merson, & O'Donoghue, 2017). Furthermore, hearing health infrastructure, rehabilitation services, and assistive resources are often underdeveloped in LMICs (Harris & Dodson, 2017).

In LMICs like South Africa, healthcare facilities are typically tiered into three levels of care: primary, such as community-based services and point-of-entry clinics; secondary, which include district and regional hospitals; and tertiary, which encompasses specialised services (National Health Act, 2003). Due to the limited number of primary-level hearing screening sites in these settings, children are often referred directly to a centralised tertiary-level hospital for initial hearing screening, when available. Referrals to central tertiary-level hospitals for primary care services like hearing screening add to growing waiting lists for specialised care like diagnostic hearing assessments and hearing aid fittings. Direct referrals to a central tertiary-level hospital often imply that parents and caregivers must travel further to access hearing healthcare infrastructure, which may in turn lead to poor follow-up rates, delayed diagnoses, and later access to hearing technology. Childhood hearing loss impedes speech, language, and academic development (JCIH, 2019), and early auditory stimulation is crucial to minimise the adverse effects of hearing loss in children (Wolfe & Smith, 2016).

Access to sustainable hearing healthcare services in LMICs is an important public health priority (Swanepoel & Clark, 2018). Innovative service delivery models, with an emphasis on decentralisation, are required to develop sustainable services in these settings (Swanepoel & Clark, 2018). Decentralisation is the transfer of responsibility for planning, management, and financing from central to peripheral levels of government and has been a key health sector reform in a wide range of LMICs over the past decade (McIntyre & Klugman, 2003). Despite implementing decentralisation as a strategy across many health systems, the impact on health equity is still unclear (Sumah, Baatiema, & Abimbola, 2016). However, in order to minimise such inequity, government, health sectors, and communities must address socioeconomic and

financial barriers and implement complementary mechanisms alongside decentralisation (Sumah et al., 2016).

The growing burden of hearing loss in LMICs (WHO, 2021) is disproportionate to the lack of hearing healthcare services available, and efforts to reach underserved communities are inadequate (Swanepoel et al., 2010; Swanepoel & Clark, 2018). If hearing healthcare services are not available at primary-level healthcare clinics, many communities in LMICs do not have access to these services at all (Tanser, Gijsbertsen, & Herbst, 2006), and tertiary-level services are being over-burdened with screening services that should be conducted at a lower level of care. Therefore, approaches that incorporate the delivery of community-based hearing care to decentralise services is a priority (Louw, Swanepoel, Eikelboom, & Myburgh, 2017; Suen, Bhatnagar, Emmett, Marrone, Kleindienst, Swanepoel, et al., 2019). *Study II investigated a decentralised model of hearing healthcare* through district hearing screening in the Western Cape Province, South Africa.

1.5 Management of hearing loss in children

Management of childhood hearing loss involves prevention where possible, early identification, accurate diagnosis, selection of appropriate hearing technology, and auditory rehabilitation. Children with hearing loss who reside in rural areas typically receive hearing technology (like hearing aids or cochlear implants) much later when compared to children who reside in urban areas (Bush, Kaufman, & McNulty, 2017). The WHO estimates that less than 10% of hearing aid needs are met in LMICs across the lifespan (WHO, 2021).

It is now universally agreed that to ensure optimal outcomes for children with hearing loss, the earliest possible access to appropriate intervention is required (WHO, 2021). A primary component of intervention for children with hearing loss is access to sound using hearing aids or other assistive technologies (Bagatto et al., 2011). The main aim of fitting hearing aids is to improve functional listening skills and to promote

participation in hearing-specific communication situations (Bagatto et al., 2011). Hearing aid outcomes are typically described by obtaining aided speech perception results, feedback from parent and teacher questionnaires, as well as documenting hearing aid use via data-logging tracker software in the device (American Academy of Audiology [AAA], 2013; Bagatto & Scollie, 2019). Accurate description of a child's auditory behaviour and outcomes with hearing aid use is important to make rehabilitative decisions, such as determining efficacy of hearing aids and rehabilitation programmes and evaluating the appropriateness of educational placement and academic performance (Tharpe & Seewald, 2016). Measuring hearing aid outcomes is a complex process because no single measurement exists to determine outcomes on the multidimensional aspects of auditory behaviour in children (Saunders, Chisholm, & Abrams, 2005). This process becomes even more complicated due to barriers (such as lack of standardised outcomes assessment tools in a multi-lingual and multi-cultural context), within a resource constrained LMIC context typical of most South African children.

Hearing aid use can be measured objectively by recording the data logging information that is automatically stored in a hearing aid and reported as average daily use in hours (Saunders, Bott, & Tietz, 2020). Data logging information is ideal for use in controlled observations (Laplante-Lévesque, Nielsen, Jensen, & Naylor, 2014; Saunders et al., 2020), and can be collected and recorded by audiologists whenever the hearing aids are connected to a computer with programming software. Consistent hearing aid use is critical for children to benefit from early intervention programmes and is the foundation for the development of spoken language (Marnane & Ching, 2015; Muñoz, Olson, Twohig, Preston, Blaiser, & White, 2015). Children with hearing loss who consistently use well-fitted hearing aids develop better vocabulary, grammar, and oral language (Tomblin, Harrison, Ambrose, Walker, Oleson, & Moeller, 2015; Walker, Holte, McCreery, Spratford, Page, & Moeller, 2015).

Hearing aid fitting does not necessarily mean full-time hearing aid use in children (McCreery & Walker, 2017). Previous studies suggest that children generally use their hearing aids between 5.5 and 8.5 hours a day (Gustafson, Davis, Hornsby, & Bess,

2017; Jones & Launer, 2010), and 40% of children in a large-scale multi-centre study in the United States used their hearing aids for less than 4 hours per day (Jones & Launer, 2010). These findings indicate that children with hearing loss do not have the same auditory exposure as their normal-hearing peers (Galland, Taylor, Elder, & Herbison, 2012; Paruthi et al., 2016), and more context-specific evidence is necessary regarding factors that influence hearing aid use to promote equivalent auditory-based outcomes for children with hearing loss.

Understanding parent-related challenges with hearing aid management and potential factors that are associated with hearing aid use can help audiologists to better support families of children with hearing loss (Muñoz et al., 2015; Wiseman & Warner-Czyz, 2018). Available evidence suggests that lower levels of maternal education, retention challenges, lack of caregiver-perceived benefit with hearing aid use, and limited access to hearing healthcare services all contribute to decreased hearing aid use in children (Marnane & Ching, 2015; Muñoz et al., 2015; Muñoz, Larsen, Nelson, Yoho, & Twohig, 2019; Wiseman & Warner-Czyz, 2018).

Limited evidence is available regarding typical outcomes for children with hearing loss in South Africa and potential contributing factors to hearing technology use (Booysen, Le Roux, Masenge, & Swanepoel, 2021). A recent South African study identified a range of intrinsic and extrinsic predictive factors for increased hearing technology use in a large diverse sample of children with hearing loss (Booysen et al., 2021). Intrinsic predictors included older chronological age, more severe degrees of hearing loss, and older age at diagnosis and hearing aid fitting (Booysen et al., 2021). Independent use of hearing technology, at least one cochlear implant as part of the hearing technology fitting, co-ordinated onsite audiological management, self-procured batteries, auditoryoral communication mode, and regular caregiver attendance at intervention sessions were identified as extrinsic predictive factors of increased hearing technology use (Booysen et al., 2021). This recent South African study utilised non-probability convenience sampling to select children from an auditory-based intervention programme, therefore limiting true generalisation of findings (Etikan, Musa, & Alkassim, 2016). The use of spoken language as mode of communication is not always an available option to all children with hearing loss growing up in LMICs. In

South Africa, unequal access to hearing technology, intervention, and support to promote auditory/oral communication remains a challenge for children with hearing loss (Khoza-Shangase & Kanji, 2021).

Describing hearing aid outcomes in children from low-resourced settings will contribute to the currently limited evidence base on hearing healthcare services for vulnerable populations. *Study III described hearing aid outcomes and potential factors associated with hearing aid use in South African children* with bilateral sensorineural hearing loss (SNHL) accessing the public health care system.

1.6 Conclusion

Limited data is available on the nature and aetiological factors associated with childhood hearing loss in South Africa. Knowledge regarding unique aetiological factors for childhood hearing loss in LMICs like South Africa will ensure that at-risk infants are identified and referred for early hearing screening and timeous intervention. Lack of access to hearing screening services for children in resource-constrained areas remain a challenge for early diagnosis of hearing loss and appropriate intervention. There is a need for policymakers to allocate resources for, and plan strategically to promote access to ear and hearing care (WHO, 2021). Describing the effect of a decentralised model of hearing health care will provide valuable information to inform strategic decision-making. Limited context-specific evidence-based outcomes have been described for children who use hearing aids in low-resourced contexts. Children with hearing loss require specialist interventions; therefore, describing their outcomes with hearing aids are important to plan for educational support and intervention services (Bagatto et al., 2011).

CHAPTER 2

METHODOLOGY

2.1 Research objectives

The aim of this study was to 1) describe the profile of childhood hearing loss; 2) to explore a decentralised model of hearing healthcare through district hearing screening; and 3) to describe the hearing aid outcomes of children with bilateral SNHL in the Western Cape public healthcare system, South Africa.

To achieve the main aim, this study was divided into three research objectives, each constituting a research project that was published or submitted as an article in ISI accredited peer-reviewed journals. These three studies are summarised in Table 2.1 according to titles and objectives.

Study	Study I	Study II	Study III
Title	Profile of childhood hearing loss in the Western Cape, South Africa	Decentralising paediatric hearing services through district healthcare screening, Western Cape, South Africa	Outcomes of children with sensorineural hearing loss fitted with binaural hearing aids at a paediatric public hospital in South Africa
Objectives	To describe the nature, associated risk factors and age of diagnosis for childhood hearing loss in a South African cohort from the Western Cape Province.	 To explore a decentralised model of hearing healthcare through district hearing screening in the Western Cape Province, South Africa as measured by: Attendance rates for initial hearing screening Patient travelling distance Number of referrals to a tertiary-level hospital and hearing outcomes 	 To describe hearing aid outcomes of children aged 0-13 years with bilateral sensorineural hearing loss by: Recording average daily hearing aid usage at one-month and three-months post-fitting Identifying factors that are associated with hearing aid use Describing oral/aural performance as measured by the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire
Publication status	Accepted and published: Kuschke, S., Swanepoel, D. W., Le Roux, T., & Strauss, S. (2020). Profile of childhood hearing loss in the Western Cape, South Africa. <i>International Journal of Pediatric</i> <i>Otorhinolaryngology, 137</i> , 110248. https://doi.org/10.1016/j.ijporl.2020.110248	Accepted and published: Kuschke, S., Le Roux, T., Scott, A. J., & Swanepoel, D. W. (2021). Decentralising paediatric hearing services through district healthcare screening in Western Cape Province, South Africa. <i>African Journal of</i> <i>Primary Health Care & Family Medicine,</i> <i>13</i> (1). https://doi.org/10.4102/phcfm.v13i1.2903	Accepted and in press: Kuschke, S., Swanepoel, D. W., & Le Roux, T. (2021). Outcomes of children with sensorineural hearing loss fitted with binaural hearing aids at a paediatric public hospital in South Africa. <i>International Journal of Pediatric</i> <i>Otorhinolaryngology. In press.</i> https://doi.org/10.1016/j.ijporl.2021.110977
Chapter in thesis	Chapter 3	Chapter 4	Chapter 5

Table 2.1. Summary of studies according to title and objectives

2.2 Research context

The Western Cape Province is one of nine provinces in South Africa. The Cape Town metropole has a population of 4 067 774 and is situated in the southern peninsula of the Western Cape Province (Statistics South Africa, 2014). The Metropole incorporates eight health sub-districts with eight district-level hospitals (of which only three have Audiology services). Victoria Hospital is a secondary hospital in the South Peninsula health district of the metropolitan region, and currently has no audiology services. The hospital has 159 beds and is situated in Wynberg, Western Cape. All patients aged 0-13 years who are from the Victoria catchment area and who are in need of audiology services are referred to RCWMCH.

The Western Cape has three tertiary academic hospitals. RCWMCH is the only dedicated paediatric tertiary-level academic hospital in sub-Saharan Africa and serves as a central referral hospital for patients (birth to 13 years of age) across the entire Western Cape Province. The Audiology Department assesses and provides hearing rehabilitation for approximately 300 children per month. Referrals are received from district hospitals, as well as from a number of primary level clinics and maternal and obstetric units (MOUs). RCWMCH is situated in a LMIC and serves mostly children who do not have access to private medical insurance from the public health care sector. RCWMCH is a referral hospital for specialised cases, and as such, a large number of children with syndromes and infectious diseases such as HIV, TB, cytomegalovirus, rubella, and meningitis are seen.

Patient data of all children seen at the RCWMCH Department of Audiology is captured daily on an electronic datasheet. The data sheet includes categorical and numerical data: chronological age; age of suspicion and diagnosis of hearing loss; geographic area of residence; referral source; type of assessment conducted, hearing outcomes (type and degree of hearing loss per ear); reason for referral; risk factors for hearing loss; management and referrals made. Only four audiologists have access and permission to enter data into this data sheet, which is stored on a password-protected computer, and backed up monthly. All four audiologists have been trained to enter data in a uniform manner for consistency and reliability.

A Departmental protocol for paediatric hearing aid fitting (based on the 2013 American Academy of Audiology Clinical Practice Guidelines on Paediatric Amplification and the 2019 Ontario Protocol for the Provision of Amplification) is used by all audiologists at RCWMCH. Behind-the-ear air-conduction hearing aids from the same company was fitted for all the participants in this research project. All hearing aid fittings are verified at the initial fitting by calculating the aided audibility of speech through the hearing aid as measured with probe microphone measures (AAA, 2013; Bagatto & Scollie, 2019). Real-ear aided response (REAR) probe microphone measurements are done where possible. In cases where REAR measurements cannot be obtained, simulated REAR measurements in a coupler using measured or age-appropriate real-ear to coupler difference (RECD) are obtained (AAA, 2013; Bagatto & Scollie, 2019).

The Aided Speech Intelligibility Index (SII) Normative Values v1.0, Revision 2 form is used for all patients who are fitted with hearing aids in order to determine whether the child's hearing aid fitting is electro-acoustically acceptable for his/her pure tone average (AAA, 2013; Bagatto & Scollie, 2019). Validation of hearing aid fitting is done in accordance with international protocols (Ontario Protocol for the Provision of Amplification, 2019) by issuing caregiver questionnaires like the PEACH questionnaire (Ching & Hill, 2005) (Appendix A). The PEACH questionnaire in the original English format is issued to caregivers (for children ≥24 months) in hard-copy format at the initial hearing aid fitting. Caregivers are encouraged to observe their children's behaviour in the month following initial hearing aid fitting, and to complete the PEACH questionnaire in the week prior to their one-month follow-up appointment. The managing audiologist scores and records the questionnaire at the one-month post-fitting follow-up appointment. In cases where caregivers are not proficient in reading and writing in English, the PEACH questionnaire is administered interview-style by the managing audiologist.

2.3 Ethical considerations

The research project was approved by the University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419; Appendix B), the University of Cape Town Human Research Ethics Committee (365/2019; Appendix C), the Red Cross War Memorial Children's Hospital Ethics Committee (RCC203; Appendix D), and the Western Cape Health Research sub-directorate (WC_201906_023; Appendix E).

A major theme throughout this research is equitable access to hearing healthcare. This implies that hearing healthcare should be available to everyone, irrespective of socioeconomic status, age, sex, religion, or geographical location (WHO, 2021). Medical and healthcare research is subject to ethical standards that promote respect for all human beings and protect their health and rights (South African National Health Act, 2013). The current study was conducted within the framework of general ethical concepts for medical and social research (Declaration of Helsinki, 2013) and the ethical guidelines set out in the South African National Health Act (2013). The ethical concepts applied in the research design, participant selection, and data collection and analysis procedures of this study, are summarised below in Table 2.2.

Table 2.2. Ethical concepts applied to formulation of research design, participant selection, and data collection and analysis

 procedures (South African National Health Act, 2013; Declaration of Helsinki, 2013).

Ethical concept	Application to study
The research protocol must be submitted for consideration, comment, guidance, and approval to the concerned research ethics committee before the study begins.	The study was approved by the University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419; Appendix B), the University of Cape Town Human Research Ethics Committee (365/2019; Appendix C), the Red Cross War Memorial Children's Hospital Ethics Committee (RCC203; Appendix D), and the Western Cape Health Research sub-directorate (WC_201906_023; Appendix E).
	All the participants in this largely retrospective cohort study were either children or their caregivers who were seen at the RCWMCH Department of Audiology, or who were seen at Victoria District Hospital as part of an outreach hearing screening project. It was therefore necessary for the researcher to obtain institution-specific ethical clearance from the Ethics Review Committee of RCWMCH as well as Victoria District Hospital prior to commencing the study.
Any form of health research conducted in South Africa	The rationale and research design of this study were carefully considered in order to contribute to
which involves human participation, must be relevant	the overall needs of young children with hearing loss. By collecting data from the Department of
both to the overall health and developmental needs of	Audiology at a tertiary paediatric academic hospital in the Western Cape, which serves children from
the people of the country, as well as the individual	various parts of the country, the individual requirements of this population would be addressed.
needs of those who are affected by the concerns being	
investigated in the study.	
Participants' rights to privacy and confidentiality	Participant confidentiality was be ensured as all identifying information was treated in a confidential
should be protected at all times. The confidentiality of	manner. When the data required for this study was retrieved from the Audiology Departmental
data that could identify participants should also be	database at RCWMCH, all identifying or personal information was excluded, and data were
protected.	presented anonymously. Although results from the district hearing screening intervention formed part
	of routine clinical investigation, participant confidentiality for Study II was maintained by excluding all identifying or personal information when the data was presented. Confidentiality and the right to
	privacy were confirmed in a signed routine research consent slip at the Department of Audiology (Appendix F).

Informed assent/consent should be obtained from	Data were retrieved retrospectively from the Audiology Departmental database at RCWMCH for
participants/their caregivers. For a potential research	
	Studies I and III. Caregivers of patients who are seen at the Department of Audiology at RCWMCH
subject who is incapable of giving informed consent,	sign a routine consent slip for permission to use patient data for research and educational purposes
the researcher must seek informed consent from the	(Appendix F). A letter of informed consent was issued to the caregivers of participants prior to data
legally authorised representative.	collection for the decentralised screening project in Study II (Appendix G). Informed assent was
	obtained from children over the age of seven years to participate in Study II (Appendix H). Informed
	consent letters were issued to parents of children who met the inclusion criteria for Study III and who
	completed a PEACH questionnaire (Appendix I).
Respect for persons should be maintained at all times	All research procedures were carefully explained to both caregivers and participants, and data
in terms of dignity and autonomy.	collection only commenced once informed consent and assent had been obtained. Caregivers
	(Appendix G and I) and participants (Appendix H) were informed that they had a right to choose to
	participate or withdraw from the research at any time, without negative consequences.
Researchers, authors, sponsors, editors and	All research information, including the methods, techniques of analysis and findings, have been
publishers all have ethical obligations with regard to	reported systematically and made available to other researchers. Research findings and results were
the publication and dissemination of the results of	reported in an accurate manner without misinterpreting, misrepresenting, intentionally misleading
research.	others or withholding findings or results (Leedy & Ormrod, 2015). Individuals who contributed to, as
	well as references that were consulted during the execution of this study, were acknowledged to
	ensure that plagiarism was avoided. Upon completion of the study, results and limitations were
	reported in the three articles, which were submitted to ISI accredited peer-reviewed journals for
	publication. Additionally, a thesis was compiled and will be available both online and in hard copy
	format at the University of Pretoria library.
The selection, recruitment and inclusion or exclusion	All participants who adhered to the inclusion criteria were included.
of research participants should be just and fair.	
Protection from harm: The rights, safety and wellbeing	There were no risks involved for the participants of this study. The benefit for the population in this
of participants are the most important considerations	study include: a description of the clinical profile and risk factors for childhood hearing loss; more
in research and should be placed above the interest of	equitable access to hearing healthcare through decentralisation of services; a description of the
science and society.	hearing aid outcomes for children with SNHL in a low-resourced context.
Plagiarism will not be tolerated in research.	This thesis is the researcher's own original work. Where secondary material was used, it was
	carefully acknowledged and referenced in accordance with university requirements.
Storage of data.	All raw data collected during this research will be stored electronically at the Department of Speech-
	Language Pathology and Audiology at the University of Pretoria for at least 15 years. Additionally,
	all research datasets will be uploaded onto the Research Data Repository at the University of
	Pretoria.

2.4 Study I: Nature, associated risk factors and age of diagnosis for childhood hearing loss, Western Cape, South Africa

2.4.1 Research design and participants

Study I followed a descriptive research design, as this type of research design involves identifying the characteristics of an observed phenomenon, which in this case was the clinical profile of young children aged 0 - 6 years with confirmed hearing loss, and not changing the condition under investigation (Leedy & Ormrod, 2020). A retrospective cohort design encompassing data from January 2019 to July 2019 was employed for Study I. Cohort studies follow a group of participants over time and are also known as observational studies since there is no manipulation of any variable (Haynes & Johnson, 2009). The primary advantages of a retrospective cohort design are lower costs and no need to follow participants over time (Haynes & Johnson, 2009). Data collected included demographic information, type and degree of hearing loss, documented risk factors associated with hearing loss, and age of suspicion and diagnosis of hearing loss. Purposive sampling (Etikan et al., 2016) was used to select participants. Inclusion criteria was set out as follows, and made up the sample of 240 children:

All children aged 0 - 6 years with a diagnosis of confirmed unilateral or bilateral hearing loss (irrespective of type or degree of hearing loss) of \geq 20 dBHL averaged across 0.5 kHz, 1 kHz, and 2 kHz (Northern and Downs, 2002) at RCWMCH between January 2018 and January 2019.

2.4.2 Data collection material and equipment

Quantitative data was collected for Study I, since quantitative research is structured with the purpose of explaining, predicting, or expanding, using various measuring or data-capturing instruments (Leedy & Ormrod, 2020). An electronic database which is updated daily in the Department of Audiology at RCWMCH to record patient data was used to retrospectively review clinical and patient data. Some data that are not

routinely included in the electronic database were captured from clinical records in patient hospital files. The data sheet included categorical and numerical data.

2.4.3 Data collection procedures

Prior to data collection for all three studies, the research project was approved by the University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419) (Appendix B), the University of Cape Town Human Research Ethics Committee (365/2019) (Appendix C), the Red Cross War Memorial Children's Hospital (RCWMCH) Ethics Committee (RCC203) (Appendix D), and the Western Cape Health Research sub-directorate (WC_201906_023) (Appendix E).

An electronic database, updated daily with patient data in the Department of Audiology at RCWMCH, was used to conduct a retrospective review of clinical data from children under the age of six years who were diagnosed with confirmed hearing loss between January 2019 and July 2019. Some data that were not routinely included in the electronic database were captured from clinical records in patient hospital files. Caregivers of patients who are seen at the Department of Audiology at RCWMCH sign a routine consent slip for permission to use patient data for research and educational purposes (Appendix F). Data collected included demographic information, type and degree of hearing loss, documented risk factors associated with hearing loss, and age of suspicion and diagnosis of hearing loss. Only children under six years of age with confirmed hearing loss were included in the sample, due to the paucity of information on the hearing profile in very young children who are not yet of school-going age in South Africa.

The audiological test battery typically included tympanometry, acoustic reflex-testing, OAEs, and frequency-specific air- and bone-conduction auditory brainstem response (ABR) testing where indicated. Behavioural audiometry (air- and bone-conduction pure tone testing) was used where age-appropriate, to determine the type and degree of hearing loss. Normal peripheral hearing was defined as air-conduction thresholds ≤15 dBHL (WHO, 2021). Hearing loss was indicated when the pure tone average was

>15 dBHL across three frequencies (500 Hz, 1 000 Hz and 2 000 Hz). The minimum diagnostic criteria for ANSD were the presence of OAEs or a clear cochlear microphonic response at 85 dBnHL and 95 dBnHL with absent or abnormal ABR waves (Berlin et al., 2010).

2.4.4 Data analysis

Data were captured on Microsoft Excel 2016 (Microsoft Corp, Redmond, WA), and analysed using SPSS 24 (Version 24.0.IBM Corp., Armonk, NY). Descriptive statistical methods were used to describe the clinical profile of participants in terms of percentages of occurrence as well as mean ages and standard deviations: demographics, type and degree of hearing loss, etiological factors associated with hearing loss, age of suspicion and age at diagnosis of hearing loss.

2.5 Study II: Decentralising paediatric hearing services through district healthcare screening, Western Cape, South Africa

2.5.1 Research design and participants

A pragmatic, quasi-experimental research design was used for Study II, with a sevenmonth control period of standard hearing service provision at a tertiary hospital (June 2018 to December 2018), and a seven-month intervention period where hearing screening was offered at a district hospital (June 2019 to December 2019). This design was implemented to determine the effects of a decentralised model of hearing healthcare in terms of attendance rates for initial hearing screening, patient travelling distance, number of referrals to a tertiary-level hospital, and hearing outcomes.

Prospective cohorts (for the intervention arm of Study II) recruit a group of participants, measure predictor variables, identify potential confounders, and by following the unit over time, measure specific outcome variables (Cummings, Newman & Hulley, 2001, in Haynes & Johnson, 2009). The general purpose of this design is to describe the incidence or analyse associations of risk factors (predictor variables) for a specific

outcome variable or condition (Cummings, Newman & Hulley, 2001, in Haynes & Johnson, 2009). The major advantage of using a prospective cohort is the ability to control the sampling, quality, and selection of predictor variables (Haynes & Johnson, 2009).

Consecutive purposive sampling was used to select participants for Study II. All patients who were referred to the tertiary hospital for initial hearing screening from the district hospital catchment area during the control period (June 2018 to December 2018), and who attended their hearing screening appointment at the tertiary hospital, were included in the tertiary group, regardless of the reason for referral. The tertiary hospital group comprised of 315 participants.

Referrals for initial hearing screening from the district hospital during the intervention period (June 2019 to December 2019) were selected based on specific inclusion criteria. High-risk patients (e.g., prematurity <34 weeks gestation, hyperbilirubinaemia, and syndromes associated with hearing loss), were excluded and booked at the tertiary hospital. Patients with known middle ear pathology such as otitis media or otorrhoea were also excluded from the district group, as they would have been better served at the tertiary hospital with a diagnostic hearing assessment. The district hospital group comprised of 158 participants.

2.5.2 Data collection material and equipment

An electronic patient database from the Department of Audiology at the tertiary hospital was used to retrospectively review data of the patients from the district hospital catchment area who were referred to the tertiary hospital for initial hearing screening during the control period (June 2018 to December 2018). Data included demographic information, reason for referral, initial hearing screening results, and number of children from the district hospital catchment area who were referred directly to the tertiary hospital. The same electronic patient database was used to review the number of children from the district hospital catchment area who were referred to the

tertiary hospital for initial hearing screening during the seven-month intervention period at the district hospital (June 2019 to December 2019).

A hearing screening data sheet for the seven-month intervention period at the district hospital (June to December 2019) was used to record patient data in terms of demographics, geographical area of residence, reason for referral, OAE screening results, and need for further diagnostic testing. The Maico Eroscan® OAE test system was used for initial hearing screening during both the control and intervention periods. The system incorporates a screening function with a four-frequency (2 000 - 5 000 Hz) low-to-high distortion-product OAE testing protocol, and conducts a fast, automatic test showing a Pass/Refer result. The signal-noise-ratio is set at 6dB, and a Pass result is obtained if three frequencies pass. Patients in the district group who referred the initial screening unilaterally or bilaterally underwent tympanometry (GSI39 AutoTymp®) to check their middle ear status.

2.5.3 Data collection procedures

An electronic patient database from the Department of Audiology at the tertiary hospital (RCWMCH) was used to retrospectively review data of the patients from the district hospital (Victoria Hospital) catchment area who were referred to the tertiary hospital for initial hearing screening during the control period (June to December 2018). Data included demographic information, reason for referral, initial hearing screening results, and number of children from the district hospital catchment area who were referred directly to the tertiary hospital. Only initial OAE hearing screening results were included for the tertiary group, as diagnostic testing was done on the same day at the tertiary hospital if a patient referred OAE screening unilaterally or bilaterally, instead of scheduling a re-screen two weeks later at the tertiary hospital. Diagnostic assessment results were also included for those children who referred initial OAE screening unilaterally or bilaterally in the tertiary group. The same electronic patient database was used to review the number of children from the district hospital for initial hearing and the tertiary hospital for the tertiary hospital in the tertiary group.

screening during the seven-month intervention period at the district hospital (June to December 2019).

A letter of informed consent was issued to the caregivers of participants prior to data collection for the decentralised screening project in Study II (Appendix G). Informed assent was obtained from children over the age of seven years to participate in Study II (Appendix H). A hearing screening data sheet for the seven-month intervention period at the district hospital (June to December 2019) was used to record patient data in terms of demographics, geographical area of residence, reason for referral, OAE screening results, and need for further diagnostic testing (Appendix J). The Maico Eroscan® OAE test system was used for initial hearing screening during both the control and intervention periods. Patients in the district group who referred the initial screening unilaterally or bilaterally underwent tympanometry to check their middle ear status and were referred to the paediatrician at the district hospital on the same day as the initial hearing screening in order to treat any middle ear pathology. These patients were re-screened at the district hospital after two weeks, and if another unilateral or bilateral Refer result was obtained on the re-screen, they were referred for diagnostic hearing assessment at the tertiary hospital.

2.5.4 Data analysis

Data were entered into Microsoft Excel 2016 (Microsoft Corp, Redmond, WA), and descriptive analysis was performed in terms of percentage of occurrence, means and standard deviations for demographic information and hearing outcomes. Data were imported into SPSS 26 (Version 26.0. Armonk, NY: IBM Corp.) for inferential analysis. Pearson Chi-square test was utilised for categorical data (gender; language), whereas Student's *t*-test was utilised for parametrical numerical data (attendance rates; travelling distances; number of referrals). A *p*-value of \leq 0.05 was considered significant.

2.6 Study III: Outcomes of children with sensorineural hearing loss fitted with binaural hearing aids at a paediatric public hospital in South Africa

2.6.1 Research design and participants

A retrospective review of clinical and caregiver-reported data was employed for Study III. Purposive sampling (Etikan et al., 2016) was used to include 68 children aged 0-13 years with a diagnosis of confirmed bilateral SNHL of >20dBHL averaged across 0.5 kHz, 1 kHz, and 2 kHz, with an air-bone gap <15 dBHL averaged over 0.5 kHz, 1 kHz, and 2 kHz (WHO 2021), who were fitted at RCWMCH with binaural air-conduction hearing aids between January 2017 and January 2019.

A purposive sample is one whose characteristics are defined for a specific, relevant purpose (Etikan et al., 2016; Leedy & Ormrod, 2020). The findings of a study based on purposive convenience sampling can be generalised to the sub-population from which the sample was drawn (Leedy & Ormrod, 2020). Convenience sampling has high internal validity, but limited external validity (Etikan et al., 2016). Purposive convenience sampling is an appropriate sampling technique to use when studying a minority sub-population with specific characteristics (Leedy & Ormrod, 2020).

2.6.2 Data collection material and equipment

An electronic database which is updated daily in the Department of Audiology at RCWMCH to record patient data was used to retrospectively review clinical and patient data. Some data that were not routinely included in the electronic database (such as validation from the PEACH questionnaire) were captured from clinical records in patient hospital files.

The PEACH rating scale (Ching & Hill, 2005) (Appendix A) is a questionnaire that assesses the listening performance of children in a range of communication situations in quiet and background noise and can be used for children ≥24 months (Bagatto & Scollie, 2019). The PEACH rating scale was developed as an abbreviated version of

the PEACH Diary and has been validated on normal hearing children (Bagatto & Scollie, 2013). The PEACH requires parents to rate their child's performance in different listening situations on a scale from 0 ("Never") to 4 ("Always"). The PEACH rating scale includes 13 questions, including one question about device use, one question about tolerance for loud sounds, six questions about quiet listening situations, and five questions about listening in background noise. A percentage score for Quiet, Noise, and Overall is calculated by adding the numerical values for the response to each question and dividing it by the total number of potential points for each subscale. The total percentage score for each subscale is then used to plot performance with hearing aids in Quiet, Noise, and Overall, to indicate whether performance is typical, whether possible review is indicated, or whether further review is indicated.

2.6.3 Data collection procedures

Informed consent letters were issued to parents of children who met the inclusion criteria for Study III and who completed a PEACH questionnaire (Appendix I). Participants were identified retrospectively via a departmental electronic database and their demographic information was recorded. Independent variables that could influence hearing aid use were identified via the same database. Behind-the-ear air-conduction hearing aids from the same company was fitted for all the participants. All hearing aids were verified at the initial fitting by calculating the aided audibility of speech through the hearing aid as measured with probe microphone measures (AAA, 2013). REAR probe microphone measurements were done where possible. In cases where REAR measurements could not be obtained, simulated REAR measurements in a coupler using measured or age appropriate RECD were obtained (AAA, 2013; Bagatto & Scollie, 2019).

The average daily hearing aid use (in hours) was documented by utilising data logging information stored in each hearing aid at one-month and three-months post-fitting time periods. The hospital files of children who attended their one-month hearing aid fitting follow-up appointment were then reviewed to obtain hearing aid validation information as measured by the PEACH questionnaire.

The PEACH questionnaire in the original English format was issued to caregivers in hard-copy format at the initial hearing aid fitting. The managing audiologist scored and recorded the questionnaire at the one-month post-fitting follow-up appointment. Caregivers were encouraged to observe their children's behaviour in the month following initial hearing aid fitting, and to complete the PEACH questionnaire in the week prior to their one-month follow-up appointment. In cases where caregivers were not proficient in reading and writing in English, the PEACH questionnaire was administered interview-style by the managing audiologist. There is a section for additional comments at the end of the PEACH questionnaire, therefore qualitative written parent-reported outcomes at the one-month post-fitting appointment were also obtained and recorded from returned PEACH questionnaires for qualitative thematic analysis.

2.6.4 Data analysis

Data were imported into Microsoft Excel 2016 (Microsoft Corp, Redmond, WA) and analysed using R statistical computing program (Version 4.1). Quantitative analysis of data included descriptive and inferential statistics. Student's *t*-test was used to compare average hearing aid use (h/day) at one-month and three-months post-fitting, average hearing aid use between subgroups of children with additional disabilities and neuro-typically developing children, as well as average hearing aid use between groups of children with *Typical Overall* PEACH scores and those who required review. Hearing aid fitting software automatically averages hearing aid use between the previous and current date every time the hearing aid is coupled to the programming software.

Categorical and continuous variables were identified from the departmental electronic database. Continuous variables (age at diagnosis and hearing aid fitting) were converted into categories (*Toddler* [0 - 2 years], *Pre-school* [3 - 6 years] and *School-going* [> 6 years]). Analyses of variance (ANOVA) (α level = 0.01) was used to determine whether there was a bivariate relationship between the outcome variable (average daily hearing aid use) and the independent variables. Subsequently

independent categorical variables that were significantly associated with hearing aid use (dependent variable Y) were included in two multiple linear regression models (one-month and three-months post-fitting). Binary indicators (1;0) were applied to use these categorical variables in the multiple linear regression models. Multiple linear regression was performed to examine the simultaneous effect of multiple predictors on Y. Hearing aid use for the right and left ears differed minimally for all participants, therefore the ear with the highest data logging was selected for statistical analyses. For all analyses, the level of significance was set at .05 (p < 0.05).

Qualitative thematic analysis was applied for caregiver reported outcomes written in the additional comments section of the PEACH questionnaire. The caregiver reported written text was reviewed by the first author and themes were extracted, which were subsequently checked by the co-authors to establish a final set. These themes with examples were grouped into advantages of and barriers to hearing aid use.

CHAPTER 3

PROFILE OF CHILDHOOD HEARING LOSS IN THE WESTERN CAPE, SOUTH AFRICA

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3.1 Abstract

Objectives: To describe the nature, associated risk factors and age of diagnosis for childhood hearing loss in a South African cohort from the Western Cape Province.

Methods: A retrospective review of clinical data from children under six years of age with confirmed hearing loss at RCWMCH was conducted between 1 January 2019 and 31 July 2019. Data collected included demographic information, type and degree of hearing loss, documented risk factors associated with hearing loss, and age of suspicion and diagnosis of hearing loss.

Results: The study sample included 240 children with hearing loss, with a mean age of 42 months (21.8 SD; range 2-72). More than two thirds (68.3%) of the children presented with bilateral hearing loss. The majority presented with conductive hearing loss (64.6%), followed by sensorineural (28.7%) and mixed hearing loss (3.3%) or auditory neuropathy spectrum disorder (3.3%). More than half (51.8%) of the bilateral sensorineural hearing loss were of a profound degree. The most prominent risk factor for conductive hearing loss was otitis media, for sensorineural hearing loss it was a family history of childhood hearing loss, and for auditory neuropathy spectrum disorder it was hyperbilirubinaemia. Approximately one third of patients (27.1%) with

sensorineural hearing loss did not have any associated risk factors. The mean age of diagnosis of permanent congenital or early-onset hearing loss was 31.4 months (22.8 SD; range 2 - 72), with a mean delay of nine months (13.2 SD; range 0 - 60) between age of suspicion and diagnosis of hearing loss (n=93).

Conclusion: The large proportion of preventable hearing losses in this sample highlights the importance of maximising primary health care efforts to treat preventable causes timeously. Age of diagnosis of permanent congenital or early-onset hearing loss was severely delayed undermining prospects of positive outcomes through early intervention. Infant hearing screening services in the public health sector of South Africa should be prioritised alongside primary health care efforts to reduce preventable risks for hearing loss.

Keywords: childhood hearing loss; risk factors; age of diagnosis

3.2 Introduction

An estimated 466 million people globally suffer from disabling hearing loss of more than 30dBHL in the better hearing ear, which equates to nearly 6% of the world's population (WHO, 2021). Of these, 34 million are children (WHO, 2021). Hearing loss is the second most prevalent developmental disability, affecting approximately 15.5 million children under the age of 5 years world-wide (Olusanya et al., 2018). Sub-Saharan Africa is one of the regions where the prevalence of disabling hearing loss in children under the age of 14 years is greatest (Olusanya et al., 2018). The World Health Organisation (WHO) estimates the prevalence of hearing loss for children aged between 5 and 14 years at 1.9% in sub-Saharan Africa as opposed to 0.4% in high-income countries (WHO, 2018).

Most cases of disabling childhood hearing loss have preventable causes that are common in low-to-middle-income countries (LMICs) and make up nearly 60% of the aetiology of hearing loss in children (WHO, 2021). Children born into lower socioeconomic contexts have a higher incidence of middle ear pathology and subsequent preventable hearing loss, as well as considerably less access to non-emergency health resources (Epstein et al., 2009). Adverse pre-, peri- and post-natal conditions are prominent risk factors for childhood hearing loss, especially in LMICs (Olusanya et al, 2018). Higher rates of low birth weight and severe hyperbilirubinaemia, which are associated risk factors for childhood hearing loss, have been reported in LMICs (Olusanya, 2015). Additionally, vaccine-preventable infections like rubella and meningitis, which are associated with sensorineural hearing loss in children, occur more commonly in LMICs (Caroča et al., 2017).

Possible reasons for the difference in prevalence of hearing loss in high-income countries and LMICs include the absence of well-managed hearing screening programmes, the impact of poverty and malnutrition on hearing, lack of awareness of hearing loss and its devastating effects in children, and limited access to hearing healthcare in LMICs (WHO, 2021). Furthermore, the proportion of hearing loss

attributed to post-natal causes such as infectious diseases and middle ear disease is typically higher in LMICs (WHO, 2021).

Due to the limited availability of hearing screening programmes, as well as poor data capturing and management within existing programmes (Meyer et al, 2012), the nature and associated risk profile of childhood hearing loss in South Africa is largely unknown. Apart from studies from nearly four decades ago conducted in schools for the deaf (Sellars & Beighton, 1983), only preliminary data on the nature and associated risk profile of childhood hearing loss in South Africa are available (Le Roux et al., 2015; Swanepoel et al., 2013). This study aimed to provide one of the first reports on the profile of childhood hearing loss in the Western Cape Province of South Africa, by describing the nature, associated risk factors and age of diagnosis for childhood hearing loss in a cohort from Red Cross War Memorial Children's Hospital (RCWMCH).

3.3 Methods

The study was approved by the University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419), the University of Cape Town Human Research Ethics Committee (365/2019), RCWMCH Ethics Committee (RCC203) and the Western Cape Health Research sub-directorate (WC_201906_023).

3.3.1 Study population

RCWMCH is one of only two dedicated paediatric academic hospitals in sub-Saharan Africa. It provides specialist diagnostic audiology and intervention services to children from birth to 13 years from the public health care sector. Only children under the age of six were included in this study due to the paucity of information on the hearing profile in very young children who are not yet of school-going age in South Africa.

3.3.2 Procedures

An electronic database, updated daily with patient data in the Department of Audiology at RCWMCH, was used to conduct a retrospective review of clinical data from children under the age of six years who were diagnosed with confirmed hearing loss between January 2019 and July 2019. Some data that were not routinely included in the electronic database were captured from clinical records in patient hospital files.

Data collected included demographic information, type and degree of hearing loss, documented risk factors associated with hearing loss, and age of suspicion and diagnosis of hearing loss. Only children under six years of age with confirmed hearing loss were included in the sample. The audiological test battery typically included tympanometry, acoustic reflex-testing, OAEs, and frequency-specific air- and bone-conduction auditory brainstem response (ABR) testing where indicated. Behavioural audiometry (air- and bone conduction pure tone testing) was used where age-appropriate, to determine the type and degree of hearing loss. Normal peripheral hearing was defined as air-conduction thresholds \leq 15 dBHL (WHO, 2021). Hearing loss was indicated when the pure tone average was > 15dBHL across three frequencies (500, 1 000, and 2 000 Hz). The minimum diagnostic criteria for ANSD were the presence of OAEs or a clear cochlear microphonic response at 85 dBnHL and 95 dBnHL with absent or abnormal ABR waves (Berlin et al., 2010).

3.3.3 Data analysis

Data were captured on Microsoft Excel 2016 (Microsoft Corp, Redmond, WA), and analysed using SPSS 24 (Version 24.0.IBM Corp., Armonk, NY). Descriptive statistical methods were used.

3.4 Results

A total of 1 154 paediatric patients under the age of six years were seen at RCWMCH Department of Audiology during the study period (January 2019 - July 2019). Approximately one in five (20.8%) of these patients were diagnosed with hearing loss.

3.4.1 Demographics

The mean age of the 240 patients younger than six years of age who were diagnosed with hearing loss was 42 months (21.8 SD; range 2 - 72) with slightly more males (55.0%). The majority of patients were of coloured background (53.7%). Foreign patients from neighbouring sub-Saharan African countries constituted 11.7% of the sample. English was recorded as home language by the majority of persons (40.4%), followed by isiXhosa (27.5%) and Afrikaans (20.4%). Most referrals (53.8%) were received from Ear-Nose-Throat (ENT) specialists, followed by medical out-patients (22.9%), district referrals (10%), genetics (5.8%), Cerebral Palsy Clinic (2.9%) and others (3.5%).

3.4.2 Type and degree of hearing loss

The majority of patients presented with bilateral hearing loss (68.3%) (Table 3.1). Conductive hearing loss (CHL) was the most prevalent type of hearing loss (65.0%). The degree of CHL for the worse ear was predominantly mild (64.7%). Approximately one third of CHLs were of a moderate degree (31.4%). Bilateral permanent hearing losses (SNHL, ANSD and mixed) made up 27.9% of hearing losses. Bilateral hearing losses made up 22.5% of SNHL and 40.4% of CHL. SNHL (including mixed hearing loss) constituted 91.7%, and ANSD constituted 8.3% of permanent hearing losses. SNHL was predominantly profound in nature. Figure 3.1 presents a profile of the degree of bilateral SNHL for the worse ear (n = 54) and unilateral SNHL (n = 16).

Types of hearing loss	Bilateral	Unilateral	Combined
	% (n)	% (n)	% (n)
CHL	40.4 (97)	24.6 (59)	65.0 (156)
SNHL	22.5 (54)	6.6 (16)	29.1 (70)
Mixed hearing loss	2.5 (6)	0.4 (1)	2.9 (7)
ANSD	2.9 (7)	-	2.9 (7)
Total	68.3 (164)	31.6 (76)	100 (240)

Table 3.1. Profile of hearing losses type and laterality (n = 240)

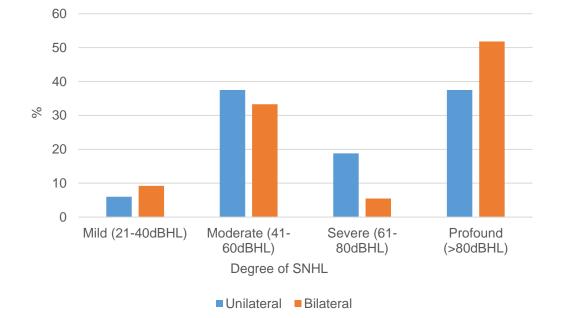


Figure 3.1: Degrees of SNHL for pure tone average threshold across 500, 1 000 and, 2 000 Hz (n = 70)

3.4.3 Risk factors associated with hearing loss

The most prominent risk factor for conductive hearing loss was middle ear pathology (73.1%), for SNHL was a family history of childhood hearing loss (18.6%), and for ANSD it was hyperbilirubinaemia (85.7%). Middle ear pathology included otitis media (OM), acute otitis media (AOM), and chronic suppurative otitis media (CSOM).

Syndromes included in this sample were Goldenhar, Trisomy 21, CHARGE, Pierre Robin and KID syndrome (Table 3.2). Approximately 70% of children with ANSD had two or more risk factors. Nearly one third (27.1%) of children with SNHL had no risk factors (Table 3.3).

	CHL (n = 156)	SNHL (n = 70)	ANSD (n = 7)	Mixed HL (n = 7)
	% (n)	% (n)	% (n)	% (n)
Middle ear pathology	73.1 (114)	-	-	28.5 (2)
Tympanic membrane perforations	12.2 (19)	-	-	-
Syndromic	3.2 (5)	11.4 (8)	-	71.4 (5)
Hyperbilirubinaemia	1.9 (3)	7.1 (5)	85.7 (6)	-
Family history of childhood hearing loss	1.2 (2)	18.6 (13)	14.2 (1)	42.8 (3)
Microtia	5.7 (9)	-	-	28.5 (2)
Нурохіа	-	10.0 (7)	28.5 (2)	-
Cytomegalovirus	0.6 (1)	10.0 (7)	-	-
Bacterial meningitis	-	7.1 (5)	-	-
VLBW* < 1 500g	-	1.4 (1)	57.1 (4)	-
Ototoxicity	-	4.2 (3)	-	-
TB** Mastoiditis	1.2 (2)	-	-	14.2 (1)
Rubella	-	2.8 (2)	-	-

Table 3.2. Documented risk factors for childhood hearing loss (n = 240)

*VLBW – Very low birthweight

**TB - Tuberculosis

Table 3.3. Number of risk factors	s for childhood he	earing loss (n = 240)
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	1 Risk % (n)	2 Risks % (n)	3 Risks % (n)	None % (n)
CHL (n = 156)	85.2 (133)	13.4 (21)	0.6 (1)	0.6 (1)
SNHL (n = 70)	65.7 (46)	7.1 (5)	-	27.1 (19)
Mixed $(n = 7)$	71.4 (5)	2.8 (2)	-	-
ANSD (n = 7)	-	71.4 (5)	2.8 (2)	-

3.4.4 Age of hearing loss suspicion and diagnosis

Age of hearing loss suspicion and diagnosis were recorded for 93 patients with permanent congenital or early-onset hearing loss, and included SNHL (n = 70), ANSD (n = 7) and permanent congenital CHL secondary to structural and genetic aetiologies (n = 13) (Table 4). Half of the participants in this sample (50%) were diagnosed with permanent hearing loss only after 36 months of age. Approximately only one third (29%) of children were diagnosed with permanent congenital or early-onset hearing loss before 12 months of age. On average, there is a delay of nine months (13.2 SD; range 0-60) between age of suspicion and age at diagnosis of hearing loss.

Table 3.4. Age of bilateral congenital or early-onset hearing loss suspicion and diagnosis (n = 93)

	Age at suspicion (months)	Age at diagnosis (months)	Suspicion-to- diagnosis delay
Mean (SD)	22.4 (20.6)	31.4 (22.8)	9.1 (13.2)
Range	1-69	2-72	0-60

3.5 Discussion

Approximately two thirds of children diagnosed with hearing loss in this South African sample from the Western Cape Province presented with CHL. This is in line with recent reports from the WHO, which postulates that the leading causes of childhood hearing loss in LMICs are conductive and treatable (WHO, 2018; WHO, 2021). The large number of children diagnosed with conductive hearing loss secondary to middle ear pathology could be attributed to the fact that the main referral source was from ENT specialists, and that RCWMCH is a tertiary referral facility with a combined ENT and Audiology service. It is evident that awareness and training for primary-level healthcare doctors and nurses in LMICs is important to provide effective first-line treatment for middle ear pathology such as AOM, OM, and CSOM, so that hearing loss and subsequent adverse effects on hearing, speech – and language development can be minimised.

More than half (51.8%) of bilateral SNHL cases were of a profound degree. A previous South African study also indicated a profound degree of hearing loss in 50% of all SNHL cases (Swanepoel et al., 2013). Estimates from high-income countries suggest that profound hearing loss make up 20-30% of permanent childhood hearing loss (WHO, 2021). The higher incidence of profound SNHL in this sample could be attributed to the fact that children with profound hearing losses tend to be identified sooner than children with less severe hearing losses, since the signs of profound hearing loss are more readily identified and may prompt parents to seek audiological assessment earlier (Durieux-Smith, Fitzpatrick, & Whittingham, 2008). Milder and even moderate losses, especially in the absence of newborn hearing screening programmes, may remain undetected until school failures or other behavioural patterns arise in school (Durieux-Smith et al., 2008). The profound nature in nearly half of SNHL cases implies that these children will not necessarily benefit optimally from hearing aids and highlights the importance of early diagnosis of hearing loss, in order to refer timeously for cochlear implant assessment. Early auditory stimulation is essential for optimal speech- and language outcomes in children with severe-profound hearing loss (Wolfe & Smith, 2018). Within the South African context, limited funding for cochlear implants within the public sector is available (Bhamjee, Le Roux, Schlemmer, Perold, Cass, Schroeder, et al., 2019). The financial implications associated with cochlear implantation has been identified by parents as the most prominent challenge regarding the paediatric cochlear implantation process in South Africa (Bhamjee et al., 2019). The high proportion of profound SNHL indicates that these children are audiological candidates for cochlear implantation. However, children in South Africa do not have equal access to cochlear implants, especially in the public healthcare sector, and therefore cochlear implantation is considered as a privileged intervention (Bhamjee et al., 2019; Le Roux et al., 2015).

ANSD as a proportion of all SNHL (including mixed hearing losses) constituted 8.3%, which is in line with previous reports of 5-17% (Bielecki, Horbulewicz, & Wolan, 2011; Le Roux et al., 2015). All of the children with ANSD diagnosis in this sample had hyperbilirubinaemia, which required phototherapy and blood transfusion. Hyperbilirubinaemia is more prevalent in African countries due to a higher incidence of G6PDD and limited treatment facilities (Cappellini & Fiorelli, 2008; Olusanya, 2015).

More than half of the children diagnosed with ANSD in this sample were also born prematurely (< 34 weeks gestation) with a very low birth weight of < 1 500g. Higher rates of very low birth weight have been reported in LMICs (Caroča et al., 2017; Olusanya, 2015). A previous South African study on the risk profile of children with profound hearing loss also included prematurity (< 34 weeks gestation) as a risk factor in 15.1% of all cases, and 40% of ANSD cases (Le Roux et al., 2015).

More than two thirds (65.7%) of children with SNHL presented with at least one risk factor for hearing loss. The most prominent risk factor for SNHL was a family history of childhood hearing loss present in 18.6% of cases. This finding is in line with two previous South African studies on the risk profiles of children with SNHL (Le Roux et al., 2015; Swanepoel et al., 2013). A multi-centre study across cochlear implant programmes in South Africa reported on family history of permanent childhood hearing loss as a risk factor for SNHL (19.6%) (Le Roux et al., 2015). A study conducted at a paediatric referral centre in Pretoria reported on any family history of childhood hearing loss as a risk factor for SNHL (27%) (Swanepoel et al., 2013). The higher incidence of family history reported in the Pretoria study could have been due to the fact that parents were able to report on any family history of childhood hearing loss, including transient episodes of childhood hearing loss due to middle ear pathology (Swanepoel et al., 2013). The high incidence of syndromic risks (11.4%) for children with SNHL in this study could be attributed to the specialised tertiary institution where the data in the current study was collected. Nearly one third (27.1%) of patients with SNHL did not present with any risk factors for hearing loss, highlighting the need for universal newborn hearing screening, and not only targeted high-risk screening in South Africa.

The average age of hearing loss diagnosis for children with permanent congenital or early-onset hearing loss (including SNHL, ANSD, and permanent congenital CHL) was 31.4 months (n = 93), surpassing two and a half years of age. This finding highlights the consequences of the lack of newborn hearing screening programmes and appropriate follow-up in the public sector of South Africa (Meyer et al., 2012; Swanepoel et al., 2009). Delayed diagnosis of hearing loss results in delayed initiation of intervention and predisposes this population to poorer speech- language and

academic outcomes (Le Roux et al., 2015). RCWMCH is a referral facility for many foreign patients from sub-Saharan Africa, including Zimbabwe, the Democratic Republic of the Congo, and Malawi. Twenty-eight children in this sample were foreign patients (11.7%), and more than two thirds (64.8%) of them were diagnosed with severe-profound hearing loss for the first time at RCWMCH at ages well beyond recommended guidelines due to a lack of audiology services in their native countries.

The mean delay of nine months between age of suspicion and diagnosis of hearing loss in this study is less than the 22 months mean delay reported in a previous South African study (Swanepoel et al., 2013). The shorter time between suspicion and diagnosis of hearing loss in this study could be due to the high incidence of a family history of childhood hearing loss as a risk factor for SNHL in this sample, or due to many children (12.1%) in the sample having complex co-morbidities, resulting in more timeous referral to audiology at a tertiary institution.

3.6 Conclusion

The nature of childhood hearing loss at the RCWMCH tertiary health care facility was predominantly bilateral and conductive. The burden of preventable hearing loss in this sample was high, supporting the case for primary level healthcare facilities to treat preventable causes of hearing loss timeously. Age of diagnosis for permanent congenital or early-onset hearing loss was significantly delayed beyond recommended ages for optimal early intervention outcomes. Universal newborn hearing screening services in the public health sector of South Africa should be prioritised along with identification and early treatment of preventable risks for hearing loss.

CHAPTER 4

DECENTRALISING PAEDIATRIC HEARING SERVICES THROUGH DISTRICT HEALTHCARE SCREENING, WESTERN CAPE, SOUTH AFRICA

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4.1 Abstract

Background: Childhood hearing loss is a global epidemic most prevalent in low- and middle-income countries where hearing healthcare services are often inaccessible. Referrals for primary care services to central hospitals add to growing lists and delays the time-sensitive treatment of childhood hearing loss.

Aim: To compare a centralised tertiary model of hearing healthcare with a decentralised model through district hearing screening for children in the Western Cape province, South Africa.

Setting: A central paediatric tertiary hospital in Cape Town and a district hospital in the South Peninsula region.

Methods: A pragmatic quasi-experimental study design was used with a 7-month control period at a tertiary hospital (June 2019 to December 2019). Decentralising was measured by attendance rates, travelling distance, number of referrals to the tertiary hospital and hearing outcomes. There were 315 children in the tertiary group and 158

in the district group. Data were collected from patient records and an electronic database at the tertiary hospital.

Results: Attendance rate at the district hospital was significantly higher (p < 0.001). Travel distance to the district hospital was significantly shorter (p < 0.001). Number of referrals to the tertiary hospital decreased significantly during the intervention period (p < 0.001). Most children in both the tertiary and district groups (78.7% and 80.4%, respectively) passed initial hearing screening bilaterally.

Conclusion: Hearing screening should be conducted at the appropriate level of care to increase access, reduce patient travelling distances and associated costs and reduce the burden on tertiary-level hospitals.

Keywords: childhood hearing loss; decentralisation; hearing healthcare; low- and middle-income countries; otoacoustic emissions.

4.2 Introduction

Hearing loss is the second most prevalent developmental disability, affecting approximately 15.5 million children under the age of 5 years globally (Olusanya et al., 2018). Approximately 95% of children with developmental disabilities reside in low-and middle-income countries (LMICs) (Olusanya et al., 2018). Sub-Saharan Africa has one of the highest prevalence rates of hearing loss (WHO, 2021), with an estimated 10.3 million children under the age of 10 years who suffer from permanent disabling hearing loss (Olusanya et al., 2020). Undetected and untreated hearing loss has a major negative impact on a child's speech, language, cognitive, educational, and socio-emotional development (JCIH, 2019).

Hearing healthcare services in LMICs are not prioritised by health systems overwhelmed by life-threatening diseases (Swanepoel et al., 2010). Identification of hearing loss in children is often impeded in LMICs because of the absence of well-managed hearing screening programmes, the impact of poverty and malnutrition on hearing and the lack of public and professional awareness of hearing loss and its devastating effects in children (WHO, 2021). In addition, poor hearing health infrastructure and resources (personnel and equipment) and geographical barriers such as distance, lead to limited accessibility of hearing healthcare services (Swanepoel et al., 2010; WHO, 2021). Children born into a lower socioeconomic status have considerably less access to non-emergency health resources (Olusanya et al., 2018; Tharpe & Seewald, 2016). Furthermore, the risk of poor follow-up rates for hearing assessments and timely intervention is higher in families who need to travel greater distances (Cavalcanti & Guerra, 2012; Ravi et al., 2016).

Compared with high-income countries, LMICs have an unequal proportion of hearing loss burden and a limited number of well-trained hearing healthcare professionals (Harris & Dodson, 2017). The number of audiologists and Ear–Nose–Throat (ENT) specialists are reported to be lowest in African countries, with an average estimate of one audiologist for every 0.8 million people and one ENT specialist for every 1.2 million people in sub-Saharan Africa (Mulwafu et al., 2017). Over a 10-year period, between

2005 and 2015, there has been no substantial improvement in these numbers (Mulwafu et al., 2017).

In LMICs such as South Africa, healthcare facilities are typically tiered into three main levels of care: primary such as point-of-entry clinics, secondary that includes district and regional hospitals and tertiary, which encompasses specialised services (National Health Act, 2003). Because of the limited number of primary-level hearing screening sites in these settings, children are often referred directly to a centralised tertiary-level hospital for initial hearing screening when available. Referrals for primary care services such as hearing screening at central tertiary-level hospitals add to growing waiting lists for specialised care such as diagnostic hearing assessments and hearing aid fittings. Direct referrals to a central tertiary hospital often imply that parents and caregivers must travel further to access hearing healthcare infrastructure, which may in turn lead to poor follow-up rates, late diagnoses, and late access to hearing technology. Childhood hearing loss impedes speech, language, and academic development (WHO, 2021) and early auditory stimulation is crucial to minimise the adverse effects of hearing loss in children (Wolfe & Smith, 2016).

Access to sustainable hearing healthcare services in LMICs is an important public health priority (Swanepoel & Clark, 2018). Innovative service delivery models, with an emphasis on decentralisation, are required to develop sustainable services in these settings (Swanepoel & Clark, 2018). Decentralisation is the transfer of responsibility for planning, management, and financing from central to peripheral levels of government and has been a key health sector reform in a wide range of LMICs over the past decade (McIntyre & Klugman, 2003). Despite being implemented as a strategy across many health systems, the impact of decentralisation on health equity is still unclear (Sumah et al., 2017). In order to minimise such inequity, government, health sectors and communities must address socio-economic and financial barriers and implement complementary mechanisms alongside decentralisation (Sumah et al. 2017).

The growing burden of hearing loss in LMICs (WHO, 2021) is disproportionate to the lack of hearing healthcare services available and efforts to reach underserved communities are inadequate (Swanepoel et al., 2010). If hearing healthcare services are not available at primary-level healthcare clinics, many communities in LMICs do not have access to these services at all (Tanser et al., 2006) and tertiary-level services are being overburdened with screening services that should be conducted at a lower level of care. Therefore, approaches that incorporate the delivery of community-based hearing care in order to decentralise hearing healthcare services is a priority (Louw et al., 2017; Suen et al., 2019).

This study aimed to compare a centralised tertiary model of hearing healthcare to a decentralised model through district hearing screening for children in the Western Cape Province, South Africa. The effects of a decentralised model of hearing healthcare were measured in terms of attendance rates for initial hearing screening, patient travelling distance, number of referrals to a tertiary-level hospital and hearing outcomes.

4.3 Methods

4.3.1 Study design

A pragmatic quasi-experimental study design was implemented, with a seven-month control group receiving standard hearing service provision at a tertiary hospital (from June 2018 to December 2018), compared with a seven-month intervention group where hearing screening was offered at a district hospital (from June 2019 to December 2019).

4.3.2 Setting

The Cape Town metropole has a population of 4 067 774 and is situated in the Southern Peninsula of the Western Cape Province, South Africa. The metropole incorporates eight health sub-districts with eight district-level hospitals of which only

three have audiology services. Victoria Hospital is a district hospital with 159 beds in the South Peninsula health district of the metropolitan region and currently has no audiology services. No audiological services are available at any of the primary healthcare clinics or maternity and obstetric units (MOUs) in this area, which result in referrals for initial hearing screening of older children based on risk factors or concerns for hearing loss. All patients aged 0 - 13 years who are from the district hospital catchment area and who need audiology services are referred directly to Red Cross War Memorial Children's Hospital, which is a central tertiary-level hospital in Cape Town.

The Western Cape has three tertiary academic hospitals. Red Cross War Memorial Children's Hospital is one of two dedicated paediatric tertiary-level academic hospitals in sub-Saharan Africa and serves as a central referral hospital for paediatric patients across the entire Western Cape who require specialised healthcare services. The Department of Audiology at this tertiary facility assesses and provides hearing rehabilitation for approximately 300 children per month. Referrals are received from district hospitals, primary level clinics and MOUs. Both the district and tertiary hospitals in this study are situated in a LMIC and serve mostly children from the public healthcare sector who do not have access to private medical insurance.

4.3.3 Study population and sampling group strategy

Consecutive sampling was used to select participants for both the tertiary and district groups.

Tertiary group sampling

All patients who were referred to the tertiary hospital via email for initial hearing screening from the district hospital catchment area during the control period (June 2018 to December 2018), and who attended their hearing screening appointment at the tertiary hospital, were included in the tertiary group, regardless of the reason for referral. These patients were retrospectively selected from the audiology departmental

electronic database at the tertiary hospital to form the tertiary group of 315 paediatric patients.

District group sampling

All consecutive referrals for initial hearing screening from facilities that fell within the district hospital catchment area were sent via email to the tertiary hospital during the intervention period (from June 2019 to December 2019). These referrals were selected for the decentralised hearing screening project at the district hospital. Only referrals who met the specified inclusion criteria for the district hearing screening project were included in the district group. The primary method of hearing screening for the district group utilised OAEs, which assesses cochlear function, therefore, referrals for initial screening of high-risk patients who presented with risk factors for retro-cochlear pathology or auditory neuropathy spectrum disorder (e.g. prematurity < 34 weeks gestation, low birthweight, hyperbilirubinaemia, and congenital syndromes associated with hearing loss) were excluded and booked at the tertiary hospital. Patients with known middle ear pathology such as otitis media or otorrhoea were also excluded from the district group, as they were likely to fail screening because of middle ear abnormality and would have been better served at the tertiary hospital with a diagnostic hearing assessment.

As a result of limited time and space available at the district hospital, only 10 - 15 paediatric patients were booked per afternoon twice per month for the seven-month intervention period, which equated to a sample size of 190 referred patients. Parents of referred children were contacted telephonically by the tertiary hospital's audiology clerk to arrange an appointment for a hearing screening at the district hospital during the intervention period (from June 2019 to December 2019). Children who attended their initial hearing screening appointment at the district hospital were included and formed the district group of 158 patients. The hearing screening at the district hospital was conducted by two audiologists from the tertiary hospital. Most of the hearing screening appointments coincided with routine follow-up paediatrician visits at the district hospital.

4.3.4 Data collection

An electronic patient database from the Department of Audiology at the tertiary hospital was used to retrospectively review data of the patients from the district hospital catchment area who were referred to the tertiary hospital for initial hearing screening during the control period (from June 2018 to December 2018). Data included demographic information, reason for referral, initial hearing screening results, and number of children from the district hospital catchment area who were referred directly to the tertiary hospital. Only initial OAE hearing screening results were included for the tertiary group, as diagnostic testing was carried out on the same day at the tertiary hospital if a patient referred OAE screening unilaterally or bilaterally, instead of scheduling a rescreen two weeks later at the tertiary hospital. Diagnostic assessment results were also included for those children who referred initial OAE screening unilaterally or bilaterally in the tertiary group. The same electronic patient database was used to review the number of children from the district hospital catchment area who were referred to the tertiary hospital for initial hearing screening during the seven-month intervention period at the district hospital (from June 2019 to December 2019).

A hearing screening data sheet for the seven-month intervention period at the district hospital (from June to December 2019) was used to record patient data in terms of demographics, geographical area of residence, reason for referral, OAE screening results, and need for further diagnostic testing. Patients in the district group who referred the initial screening unilaterally or bilaterally underwent tympanometry to check their middle ear status and were referred to the paediatrician at the district hospital on the same day as the initial hearing screening in order to treat any middle ear pathology. These patients were rescreened at the district hospital after two weeks, and if another unilateral or bilateral refer result was obtained on the rescreen, they were referred for diagnostic hearing assessment at the tertiary hospital.

Equipment

The Maico Eroscan® OAE test system was used for initial hearing screening during both the control and intervention periods. The system incorporates a screening function with a four-frequency (2 000 Hz – 5 000 Hz) low-to-high distortion-product OAE testing protocol and conducts a fast, automatic test showing a pass or refer result. The signal-to-noise ratio is set at 6 dB, and a pass result is obtained if three frequencies pass. The reliability and validity of OAEs for use in a screening setting are well-established (Ravi et al., 2016).

4.3.5 Data analysis

Data were entered into Microsoft Excel 2016 (Microsoft Corp, Washington) and descriptive analysis was performed. Data were imported into the Statistical Package for the Social Sciences (SPSS) (version 26.0. New York, IBM Corp.) for inferential analysis. Pearson's Chi-square test was utilised for categorical data, whereas Student's *t*-test was utilised for parametrical numerical data. A *p*-value of \leq 0.05 was considered significant.

4.3.6 Ethical considerations

The study was approved by the University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419), the University of Cape Town Human Research Ethics Committee (365/2019), Red Cross War Memorial Children's Hospital Ethics Committee (RCC203) and the Western Cape Health Research sub-directorate (WC_201906_023). The tertiary hospital in this study has an Outreach Policy Agreement with all Western Cape Health Facilities, which was used in conjunction with a letter requesting institutional permission from the district hospital to conduct an outreach OAE-screening service there twice per month for seven months. A letter of informed consent was issued to the caregivers of participants prior to data collection. Informed assent was obtained from children over the age of seven years.

4.4 Results

4.4.1 Demographics

The mean age of patients at the time of initial hearing screening was 48.4 months (39.0 SD; range: 1 - 156) and 52.3 months (35.1 SD.; range: 1 - 144) in the tertiary and district groups, respectively. The tertiary and district groups were similar in terms of age, gender, and language distribution (Table 4.1).

Table 4.1. Demographic characteristics of paediatric patients in the control and intervention groups

	Tertiary group (n=315)	District group (n=158)	
	n (%)	n (%)	<i>p</i> - value
Mean age in months (SD; range)	48.4 (39.0; 1-156)	52.3 (35.1; 1-144)	0.287
Gender			
Female	121 (38.4)	60 (38.0)	
Male	194 (61.6)	98 (62.0)	0.801
Home language			
English	176 (55.9)	76 (48.1)	
Afrikaans	64 (20.3)	39 (24.7)	
isiXhosa	50 (15.9)	34 (21.5)	
Other	25 (7.9)	9 (5.7)	
Mean travel distance from home			
in km (SD; range)			
To district hospital		12.6 (7.7; 1.2-36.8)	- 0.001
To tertiary hospital		19.1 (9.1; 5.1-37.6)	< 0.001

4.4.2 Attendance rates

An attendance rate of 83.2% (158/190) was found during the seven-month intervention period for patients attending the district hearing screening project, which was significantly higher than the attendance rate of 70.2% (315/449) for patients from the district hospital catchment area who were seen for initial hearing screening at the tertiary hospital during the control period (p < 0.001).

4.4.3 Travel distance

The mean travel distance for patients in the district group commuting from home to the district hospital was 12.6 km (7.7 SD.; range: 1.2 - 36.8). This distance was significantly shorter than the travel distance of 19.1 km (9.1 SD.; range: 5.1 - 37.6), which patients would have had to travel from home to the tertiary hospital (p < 0.001).

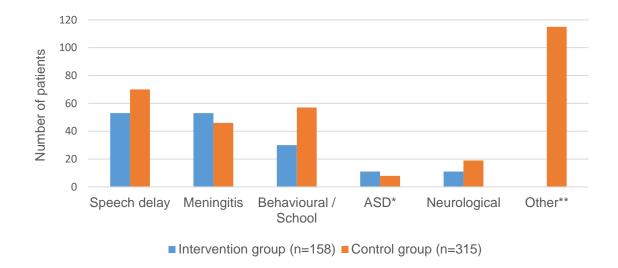
4.4.4 Number of initial hearing screening referrals to the tertiary hospital

A total of 1 729 patients were referred from facilities across the Western Cape to the tertiary hospital during the control period (from June 2018 to December 2018), of which 449 (26.0%) referrals were for initial hearing screening from the district hospital catchment area. Throughout the intervention period (from June 2019 to December 2019), during which the district screening project was being conducted, the tertiary hospital received a total of 1601 referrals from facilities across the Western Cape Province, with a significant decrease to 114 (7.1%) referrals for initial hearing screening from the district hospital catchment area (p < 0.001).

4.4.5 Reasons for referral

The reasons for referral for initial hearing screening are depicted (Figure 4.1). During the control period (n = 315), 115 referrals (36.5%) were received for reasons that were excluded from the intervention period analysis. When excluding these 115 referrals, the most common reasons for referral in the tertiary group were speech delay (35.0%) and behavioural or school-related concerns (28.5%) (n = 200). In the district group,

speech delay (33.5%) and meningitis (33.5%) were the most common reasons for referral (n = 158).



*Autism Spectrum Disorder

** Referrals not included during intervention period analysis, including; ENT referrals, risk factors for retro-cochlear pathology, genetic syndromes, head trauma, and ototoxicity

Figure 4.1: Reason for referral for initial hearing screening

4.4.6 Hearing screening outcomes for the control and intervention period

Outcomes of the initial OAE hearing screenings for the tertiary group and diagnostic assessment results for patients who referred initial OAE screening unilaterally or bilaterally from June 2018 to December 2018 are presented (Table 4.2). For the tertiary group, most patients (n = 248/315, 78.7%) passed the initial OAE screening bilaterally. The number of patients who required diagnostic assessment in the tertiary group were 67 (21.3%). Of the 67 patients who required diagnostic assessment, 54 (80.6%) attended their appointments. Half of the patients (n = 27/54, 50%) were diagnosed with mild conductive hearing loss.

Outcomes of the initial OAE screenings from the intervention period at the district hospital and the diagnostic assessment results for patients referred to the tertiary

hospital after a unilateral or bilateral refer result on rescreening at the district hospital, are also presented (Table 4.2). For the district group, most patients (n = 127/158, 80.4%) passed OAE screening bilaterally, whilst less than 10% referred OAE screening in both ears. The follow-up attendance rate for rescreening at the district hospital 2 weeks after the initial screening was 80.8% (n = 21/26). The total number of patients in the district group that needed referral to the tertiary hospital for specialised diagnostic assessment were 15 (n = 15/158, 9.5%), of which 11 (n = 11/15, 73.3%) attended the diagnostic hearing assessment appointment. Of these 11 patients, nearly half (n = 5/11, 45.5%) presented with mild conductive hearing loss.

4.5 Discussion

This study explored the effect of decentralising hearing healthcare services from a tertiary-level hospital to a district-level hospital in the Western Cape Province, South Africa. Decentralised hearing screening resulted in increased attendance rates for initial hearing screening, shorter travelling distances for patients and decreased referral rates to a tertiary-level hospital.

Attendance rates were significantly higher for initial hearing screening at the district hospital when compared with initial screening at the tertiary hospital. Non-attendance can result in underutilisation of healthcare provider time and can lead to longer appointment waiting time for patients (Downer, Meara, & Da Costa, 2005). Furthermore, especially in severely resource-constrained settings typical of LMICs, non-attendance delays the identification, diagnosis, and timeous intervention of healthcare conditions (Boksmati, Butler-Henderson, Anderson, & Sahama, 2016). The Health Professions Council of South Africa Early Hearing Detection and Intervention Guidelines suggest that a 70% and higher follow-up return rate for hearing screening is considered ideal, but that the feasibility of attaining a high follow-up rate is influenced by various factors such as access to healthcare facilities and personal constraints such as poverty (Health Professions Council of South Africa [HPCSA], 2018).

Table 4.2. Hearing screening outcomes and diagnostic assessment results for the tertiary and district groups

Hearing screening outcomes	Tertiary group	District group
	n (%)	n (%)
Initial OAE screen	(n = 315)	(n = 158)
Bilateral pass	248 (78.7)	127 (80.4)
Bilateral refer	41 (13.0)	15 (9.5)
Unilateral refer	15 (4.8)	11 (7.0)
Bilateral could not elicit	11 (3.5)	5 (3.2)
OAE re-screen		(n = 21/26)
Bilateral pass		11 (52.4)
Bilateral refer		6 (28.6)
Unilateral refer		4 (19.0)
	Diagnostic assess	ment results at the tertiary
		hospital
	(n = 54/67)	(n = 11/15)
Normal hearing	17 (31.4)	3 (27.3)
Degrees of hearing loss*		
Mild (21 – 40 dBHL)		
Conductive hearing loss	27 (50.0)	5 (45.5)
Sensorineural hearing loss	0 (0.0)	0 (0.0)
Moderate (41 – 60 dBHL)		
Conductive hearing loss	4 (7.4)	1 (9.1)
Sensorineural hearing loss	3 (5.6)	1 (9.1)
Profound (> 80 dBHL)		
Conductive hearing loss	0 (0.0)	0 (0.0)
Sensorineural hearing loss	3 (5.6)	1 (9.1)

* Pure tone average threshold for worst ear across 500, 1 000 and 2 000 Hz

The follow-up attendance rate for rescreening at the district hospital two weeks after the initial screening was high (80.8%). This could be attributed to the fact that the second screening was also conducted at a community level and coincided with a paediatrician visit to follow up on middle ear pathology for the majority of patients who referred OAE screening bilaterally. A high follow-up attendance rate (89.4%) for hearing screening was also found in a recent South African community-based study when the rescreening was conducted at a community-level as opposed to a public healthcare institution (Eksteen, Launer, Kuper, Eikelboom, Bastawrous, & Swanepoel, 2019). Patients who needed referral to the tertiary hospital for specialised diagnostic assessment had an attendance rate of 73.3%, which is in line with a previous South African community-based hearing screening study that found an attendance rate for diagnostic assessments of 75.8% (Eksteen et al., 2019).

Patient travelling distance was significantly shorter to the district hospital as opposed to the tertiary hospital. Access to services is one of the leading barriers to hearing healthcare in underserved communities (Harris & Dodson, 2017). The costs involved in attending healthcare appointments, both in terms of time taken off from work and travel costs for patients with limited resources, remain a further challenge in accessing healthcare in LMICs (Dookie & Sing, 2012). Therefore, primary healthcare is an important strategy employed in South Africa, in order to provide more accessible patient-centred services closer to home (Dookie & Singh, 2012). Community delivered hearing healthcare models have been identified as an important strategy to increase the accessibility and affordability of hearing healthcare in underserved communities (Suen et al., 2018; Wilson, Tucci, Merson, & O'Donoghue, 2017).

The inaccessibility of hearing healthcare services at a primary- or district-level, which adds severe strain on tertiary-level specialised services, may be alleviated by decentralising services. The results of this study corroborate this. The number of direct referrals for initial hearing screening from the district hospital catchment area to the tertiary hospital significantly decreased after implementation of the decentralised hearing screening project at the district hospital. The decreased number of referrals to the tertiary hospital for initial hearing screening support decreased waiting times and improved capacity to provide specialised diagnostic hearing assessments and intervention to patients requiring tertiary-level care.

More than 80% of children who attended the initial hearing screening during the intervention period at the district hospital passed initial OAEs bilaterally. This high pass rate is a positive outcome for the premise of decentralising hearing screening services to a more appropriate level of care. The majority of patients (78.7%) in the tertiary group also passed initial OAE screening, which supports the premise that hearing outcomes are similar for initial hearing screening regardless of the level of care where hearing screening is conducted. Telehealth applications are available for hearing assessment of older children (Suen et al., 2019), however, utilising OAEs in a screening setting is advantageous in terms of time taken to conduct and minimal training that is required.

The referral rate for diagnostic hearing assessment at the tertiary hospital for the children who attended hearing screening during the intervention period at the district hospital was 9.5%. This percentage is higher than the reported referral rate of a South African community-based hearing and vision screening study of 5.4%, which utilised smartphone-based pure tone audiometry screening (Eksteen et al., 2019). A possible reason for the higher referral rate is the method of screening. OAE screening is sensitive to middle ear pathology, and it is more likely to fail in the presence of abnormal middle ear function (Narayan, Kooknoor, & Rajalakshmi, 2016).

Referral for diagnostic testing in the tertiary group (21.3%) was twice as high in the district group (9.5%). The higher number of diagnostic assessments in the tertiary group were because no opportunity for rescreening after two weeks was provided, as all patients who referred initial screening unilaterally or bilaterally or those for whom OAE screening results could not be elicited, underwent diagnostic assessment on the same day in order to minimise follow-up appointments at the tertiary hospital.

Providing hearing screening at a district level increased access to medical treatment for all children who presented with middle ear pathology as evidenced by abnormal tympanometry results on the day of initial OAE screening. These children were assessed and treated by the paediatrician on the same day, instead of waiting for months to get an ENT appointment at the tertiary hospital. Thus, middle ear pathology was treated timeously and effectively at a more appropriate level of care, decreasing the added burden to long tertiary waiting lists. Early identification of middle ear pathology is a primary-level healthcare service, and it would be more appropriate to refer children even closer to home to their nearest community healthcare centres for treatment (Wilson et al., 2017). This would in turn minimise the burden on district level staff and address the problem of preventative hearing loss in children at grassroots level (Wilson et al., 2017).

A limitation of this study was that tertiary-level audiologists conducted the hearing screening at the district hospital during the intervention period. In addition, no sample size calculation was conducted, and group size was pragmatically determined by number of patients over the specified time periods. Future studies should assess the training needs of community healthcare workers and nurses to conduct hearing screening at district hospital facilities. The premise of task-shifting through community-based hearing screening programmes has been proposed as a way to improve access to hearing healthcare (Suen, 2019; Yousuf Hussein, Swanepoel, Biagio de Jager, Myburgh, Eikelboom, & Hugo, 2016). Community healthcare workers and nurses can be trained to screen for hearing loss using mobile health technology via home-based visits to reach vulnerable communities in LMICs (Yousuf Hussein et al., 2016), thereby improving access to hearing healthcare professionals in South Africa.

4.6 Conclusion

Decentralised hearing screening programmes conducted at the appropriate level of care can increase access to hearing healthcare, reduce patient travelling distances and associated costs and reduce the burden on tertiary-level hospitals. Accessible hearing screening yields higher attendance rates, leading to more effective and timeous treatment of the adverse effects of childhood hearing loss.

CHAPTER 5

OUTCOMES OF CHILDREN WITH SENSORINEURAL HEARING LOSS FITTED WITH BINAURAL HEARING AIDS AT A PAEDIATRIC PUBLIC HOSPITAL IN SOUTH AFRICA

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5.1 Abstract

Objectives: To describe hearing aid outcomes of children aged 0 - 13 years with bilateral sensorineural hearing loss in terms of average daily hearing aid use at one-month and three-months post-fitting, to identify factors that predict hearing aid use, and to describe oral/aural performance as measured by the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire.

Methods: Retrospective review of clinical data and caregiver reported outcomes. Quantitative data analysis included descriptive and inferential statistics. Linear regression models were built to determine predictive factors for hearing aid use. For all analyses, the level of significance was set at .05 (p < 0.05). Thematic analysis was done for qualitative caregiver-reported outcomes.

Study sample: Sixty-eight children aged 0 - 13 years with a diagnosis of confirmed bilateral sensorineural hearing loss, and who were fitted with binaural air-conduction hearing aids at Red Cross War Memorial Children's Hospital in Cape Town, South Africa, between January 2017 and December 2019.

Results: Average daily hearing aid use increased significantly (p < 0.05) from onemonth (5.0; 3.0 SD; range 0.3 - 14.0) to three-months post-fitting (5.9; 3.4 SD; range 1.1 – 16.8). Average PEACH scores were higher in Quiet (73.4%) than in Noise (69.6%). More than half (52.2%) of children required review based on their overall percentage PEACH scores. Higher average daily hearing aid use was significantly associated with higher overall PEACH scores (p < 0.05). Neuro-typically developing children had significantly higher hearing aid use than children with additional disabilities (p < 0.001). Qualitative caregiver feedback revealed themes pertaining to advantages and barriers to hearing aid use.

Conclusion: Outcomes of children with SNHL fitted with binaural hearing aids at a pediatric public hospital in South Africa demonstrated increased average daily hearing aid use from one-month to three-months post-fitting. Aural/oral performance was typical for one in two children. Children with additional disabilities had significantly poorer hearing aid use and aural/oral performance requiring more support for this vulnerable group to realize sufficient benefit from hearing aid use.

Keywords: Hearing aid outcomes; sensorineural hearing loss; data logging; PEACH questionnaire

5.2 Introduction

Childhood hearing loss is a global challenge and the second most prevalent developmental disability (Olusanya et al., 2018). Hearing loss affects approximately 15.5 million children under the age of five years worldwide (Olusanya et al., 2018) and is the third largest cause of global YLD (Haile et al., 2020). The estimated global cost within the education sector for providing support to children with hearing loss (aged 5 - 14 years) is 27 billion USD annually (WHO, 2021). Approximately 95% of children with developmental disabilities reside in low- and middle-income countries (LMICs) (Olusanya et al., 2018). In sub-Saharan Africa alone, an estimated 10.3 million children under the age of 10 years suffer from permanent disabiling hearing loss (Olusanya et al., 2020).

Due to poor hearing healthcare infrastructure and limited well-managed neonatal hearing screening programmes, less than 10% of the more than one million babies born annually in South Africa have access to hearing screening services (Meyer et al., 2012; Theunissen & Swanepoel, 2008). The initiation of early intervention services for children with hearing loss is often delayed in resource-constrained settings where widespread poverty is rife (Abdalla & Omar, 2011). Childhood hearing loss without intervention impedes the normal acquisition of spoken language (Lederberg, Schick, & Spencer, 2013), placing children at increased risk of poor academic performance, social- and emotional developmental delays, and behavioural disorders (Marschark, Shaver, Nagle, & Newman, 2015). Children with more severe hearing losses demonstrate poorer literacy than their normal-hearing peers with educational levels that are generally lower (Lederberg et al., 2013). Without appropriate and timeous intervention, the negative consequences of childhood hearing loss continue into adulthood with significant lifetime costs in loss of productivity (WHO, 2021).

Management of childhood hearing loss involves prevention where possible, early identification, accurate diagnosis, selection of appropriate hearing technology, and auditory rehabilitation. Children with hearing loss who reside in rural areas typically receive hearing technology (such as hearing aids or cochlear implants) much later when compared to children who reside in urban areas (Bush et al., 2017). In sub-Saharan Africa there is typically less than one hearing health professional to every million people, which severely limits capacity to deliver services for timely detection and intervention (Mulwafu et al., 2017). Along with other challenges including poor awareness and lacking infrastructure the result is that more than 80% of hearing aid needs are not met in LMICs (Clark, 2021; WHO, 2018).

It is now universally agreed that to ensure optimal outcomes for children with hearing loss, the earliest possible access to appropriate intervention is required (WHO, 2021). A primary component of intervention for children with hearing loss is access to sound using hearing aids or other assistive technologies (Bagatto et al., 2011). The main aim of fitting hearing aids is to improve functional listening skills and to promote participation in hearing-specific communication situations (Bagatto et al., 2011). Hearing aid outcomes are typically described by obtaining aided speech perception results, feedback from parent- and teacher questionnaires, as well as documenting hearing aid use via data-logging tracker software in the device (AAA, 2013; Tharpe & Seewald, 2016). Hearing aid outcomes assessment is an important part of evidencebased clinical practice (Bagatto, 2011; AAA, 2013). Accurate description of a child's auditory behaviour and outcomes with hearing aid use is important to make rehabilitative decisions, such as identifying areas that require auditory training, determining the effectiveness of the hearing aids and rehabilitation programmes, and evaluating the appropriateness of educational placement and academic performance (Tharpe & Seewald, 2016). Measuring hearing aid outcomes is a complex process because no single measurement exists to determine outcomes on the multidimensional aspects of auditory behaviour in children (Saunders et al., 2005). This process becomes even more complicated due to barriers including a lack of standardised outcomes assessment tools in a multi-lingual and multi-cultural context within a resource constrained LMIC typical of most South African children.

Consistent hearing aid use is crucial for children to benefit from early intervention programmes and is the foundation for the development of spoken language (Marnane & Ching, 2015; Muñoz et al., 2015). Children with hearing loss who consistently use

optimally fitted hearing aids develop better vocabulary, grammar, and oral language (Tomblin et al., 2015; Walker et al., 2015). Understanding caregiver-related challenges with hearing aid management and potential factors that are associated with hearing aid use can help audiologists to better support families of hearing-impaired children so that they may reach equivalent auditory-based outcomes as their hearing peers (Muñoz et al., 2015; Wiseman & Warner-Czyz, 2018). Limited evidence is available regarding typical outcomes for children with hearing loss in South Africa and potential contributing factors (Swanepoel et al., 2009). Children with hearing aids will help with planning for audiology services, educational support, amplification and intervention services (Tharpe & Seewald, 2016). The aim of this study was to describe hearing aid outcomes and potential predictors of hearing aid use in South Africa children with bilateral sensorineural hearing loss (SNHL) accessing the public health care system.

5.3 Methods

5.3.1 Setting

RCWMCH is the only dedicated paediatric tertiary-level academic hospital in sub-Saharan Africa and serves as a central referral hospital for patients across the entire Western Cape who require specialised healthcare services. Due to the hospital's central geographical area, many patients travel long distances to access healthcare services. Caregivers pay hospital fees according to their household income classification level (H0 - H3). Families with no or minimal income are served free of charge. The Audiology Department at RCWMCH provides specialist diagnostic audiology and intervention services to children from birth to 13 years from the public health care sector.

5.3.2 Study design

A retrospective review of clinical and caregiver-reported data from children aged 0 -13 years with bilateral SNHL who were fitted with binaural hearing aids between January 2017 and December 2019 at RCWMCH.

5.3.3 Study population and sampling strategy

Purposive sampling was used to identify all children aged 0 - 13 years with a diagnosis of confirmed symmetrical bilateral SNHL of >20 dB HL averaged across 0.5 kHz, 1 kHz, and 2 kHz, with an air-bone gap < 15 dB HL averaged over 0.5 kHz, 1 kHz, and 2 kHz, and who were fitted at RCWMCH with binaural air-conduction hearing aids between January 2017 and December 2019.

5.3.4 Data collection procedures

Participants were identified retrospectively via a departmental electronic database and their demographic information was recorded. Independent categorical variables that could influence hearing aid use were identified via the same database. Behind-the-ear air-conduction hearing aids from the same company was fitted for all the participants. All hearing aids were verified at the initial fitting by calculating the aided audibility of speech through the hearing aid as measured with probe microphone measures (AAA, 2013). REAR probe microphone measurements were done where possible. In cases where REAR measurements could not be obtained, simulated REAR measurements in a coupler using measured or age appropriate RECD were obtained (AAA, 2013).

The average daily hearing aid use (h/day) was documented by utilising data logging information stored in each hearing aid at one-month and three-months post-fitting intervals. The hospital files of children who attended their one-month hearing aid fitting follow-up appointment were reviewed to obtain hearing aid validation information as measured by the PEACH questionnaire.

The PEACH rating scale is a questionnaire that assesses the listening performance of children in a range of communication situations in quiet and background noise (Ching & Hill, 2005). The PEACH rating scale was developed as an abbreviated version of the PEACH Diary (Ching & Hill, 2005) and has been validated on normal hearing children (Bagatto & Scollie, 2013). The PEACH rating scale requires parents to rate their child's performance in different listening situations on a scale from 0 ("Never") to 4 ("Always"). The PEACH rating scale includes 13 questions, including one question about device use, one question about tolerance for loud sounds, six questions about quiet listening situations, and five questions about listening in background noise. A percentage score for Quiet, Noise, and Overall is calculated by adding the numerical values for the response to each question and dividing it by the total number of potential points for each subscale (Ching & Hill, 2005). The total percentage score for each guestion and dividing it by the total number of potential points for each subscale (Ching & Hill, 2005). The total percentage score for each subscale is then used to plot performance with hearing aids in Quiet, Noise, and Overall, to indicate whether performance is typical, whether possible review is indicated, or whether further review is indicated.

The PEACH questionnaire in the original English format was issued to caregivers in hard-copy format at the initial hearing aid fitting. The managing audiologist scored and recorded the questionnaire at the one-month post-fitting follow-up appointment. Caregivers were encouraged to observe their children's behaviour in the month following initial hearing aid fitting, and to complete the PEACH questionnaire in the week prior to their one-month follow-up appointment. In cases where caregivers were not proficient in reading and writing in English, the PEACH questionnaire was administered interview-style by the managing audiologist. There is a section for additional comments at the end of the PEACH questionnaire, therefore qualitative written caregiver-reported outcomes at the one-month post-fitting appointment were also obtained and recorded from returned PEACH questionnaires for qualitative thematic analysis.

5.3.5 Data analysis

Data were imported into Microsoft Excel 2016 (Microsoft Corp, Redmond, WA) and analysed using R statistical computing program (Version 4.1). Quantitative analysis of data included descriptive and inferential statistics. Student's *t*-test was used to compare average hearing aid use (h/day) at one-month and three-months post-fitting, average hearing aid use between subgroups of children with additional disabilities and neuro-typically developing children, as well as average hearing aid use between groups of children with *Typical Overall* PEACH scores and those who required review. Hearing aid fitting software automatically averages hearing aid use between the previous and current date every time the hearing aid is coupled to the programming software.

Categorical and continuous variables were identified from the departmental electronic database. Continuous variables (age at diagnosis and hearing aid fitting) were converted into categories (*Toddler* [0 - 2 years], *Pre-school* [3 - 6 years] and *School-going* [> 6 years]). Analyses of variance (ANOVA) (α level = 0.01) was used to determine whether there was a bivariate relationship between the outcome variable (average daily hearing aid use) and the independent variables. Subsequently independent categorical variables that were significantly associated with hearing aid use (dependent variables Y) were included in two multiple linear regression models (one-month and three-months post-fitting). Binary indicators (1;0) were applied to use these categorical variables in the multiple linear regression models. Multiple linear regression was performed to examine the simultaneous effect of multiple predictors on *Y*. Hearing aid use for the right and left ears differed minimally for all participants, therefore the ear with the highest data logging was selected for statistical analyses. For all analyses, the level of significance was set at .05 (p < 0.05).

Qualitative thematic analysis was applied for caregiver reported outcomes written in the additional comments section of the PEACH questionnaire. The caregiver reported written text was reviewed by the first author and themes were extracted, which were subsequently checked by the co-authors to establish a final set. These themes with examples were grouped into advantages of and barriers to hearing aid use.

5.4 Results

Sixty-eight children with bilateral SNHL who were fitted with binaural hearing aids between January 2017 and December 2019 were included in the study sample. Characteristics of the study population is presented in Table 1. More than half of the participants (52.9%) had congenital/early onset hearing loss, while most participants (38.2%) had a moderate degree of hearing loss (n = 68). The mean age of suspicion of hearing loss for participants with congenital/early onset SNHL was 23.9 months (16.3 SD; range 1 - 72), the mean age of diagnosis was 31.6 months (22.7 SD; range 2 - 72) and the mean age at hearing aid fitting was 32.5 months (23.9 SD; range 3 - 74) for these children (n = 36). There was approximately one-month delay between hearing loss diagnosis and hearing aid fitting for the congenital/early onset group. More than a quarter (26.5%) of children in this sample had additional disabilities (n = 68).

5.4.1 Hearing aid use

Data logging information was obtained for 61 participants at the one-month follow-up interval, and for 51 participants at the three-month follow-up interval. Missing data was accounted for by children not attending their one- or three-month follow-up appointments, or audiologists not recording data logging information at the follow-up sessions. Mean hearing aid use (h/day) at one- and three-month post-fitting is depicted in Table 5.2 for the right and left ears respectively. There was a significant increase in mean hearing aid use at three-months post-fitting (p = 0.030). Average daily hearing aid use was calculated for the subgroup of children with additional disabilities (n = 18) and compared to the neuro-typically developing children in this sample (n = 33) at the three-month follow-up interval. Neuro-typically developing children had significantly higher (p < 0.001) hearing aid use of 6.5 h/day (3.1 SD; range 1.2 – 14.2) than children with additional disabilities with 2.8 h/day (1.4 SD; range 0.3 – 5.2).

	% (n)
Gender	
Male	45.6 (31)
Female	54.4 (37)
Household income	
H0 (Formally unemployed)	8.8 (6)
H1 (0 USD – 400.62 USD per month*)	70.6 (48)
H2 (400.62 USD – 1430.84 USD per month*)	13.2 (9)
H3 (> 1430.84 USD per month*)	7.4 (5)
Home language	
English	50.0 (34)
Afrikaans	11.8 (8)
Xhosa	32.3 (22)
Other	5.9 (4)
Language of instruction	
English	55.9 (38)
Afrikaans	4.4 (3)
Xhosa	3.0 (2)
South African Sign Language	27.9 (19)
Augmentative and alternative communication	8.8 (6)
Educational setting	
Mainstream school	23.5 (16)
Inclusive mainstream school	4.4 (3)
Signing school	20.6 (14)
Hearing impaired skills school	13.2 (9)
Special needs school	17.7 (12)
Not of school-going age	20.6 (14)
Age at diagnosis of hearing loss in months	
Total sample (n = 68)	
Mean (SD)	54.9 (34.3)
Range	2-156

Table 5.1. Characteristics of study population (n = 68)

	Congenital/early onset (n = 36)	
	Mean (SD)	31.6 (22.7)
	Range	2-72
A	ge at hearing aid fitting in months	
	Total sample (n = 68)	
	Mean (SD)	57.0 (34.2)
	Range	3-157
	Congenital/early onset (n = 36)	
	Mean (SD)	32.5 (23.9)
	Range	3-74
C	Dnset of hearing loss	
	Congenital/early onset	52.9 (36)
	Acquired	30.9 (21)
	Unknown	16.2 (11)
A	Additional disabilities**	
	One or more additional disability	26.5 (18)
	No additional disabilities	73.5 (50)
۵	Degree of hearing loss***	
	Mild (16 – 40 dBHL)	20.6 (14)
	Moderate (41 – 60 dBHL)	38.2 (26)
	Severe (61 – 80 dBHL)	14.7 (10)
	Profound (> 80 dBHL)	26.3 (18)

dBHL – decibels hearing level

*Exchange rate of 1 USD = R14.56 (South African rand/ZAR)

**Additional disabilities included cerebral palsy, syndromes, neuro-developmental delay

***WHO classification of degree of HL based on the better ear 4FPTA (WHO 2016, 2020)

Average daily hearing aid use was also calculated for an additional two subgroups of children at the three-month follow-up interval: those whose language of instruction was South African Sign Language (SASL) (n = 19) and those with returned PEACH scores (n = 23). Average daily hearing aid use for the SASL sub-group was 4.0 h/day (2.2 SD; range 1.1 - 9.1), and for the PEACH subgroup 6.6 h/day (3.1 SD; range 1.5 - 14).

Hearing aid use	Right ear (h/day)	Left ear (h/day)	Average right and left ear (h/day)	p - value
1-month post-fitting				
Mean (SD)	5.0 (3.0)	4.9 (2.9)	5.0 (3.0)	
Range	0.3 -14.0	0.3 - 12.3	0.3 - 14.0	0.030
3-month post-fitting				0.030
Mean (SD)	5.9 (3.4)	5.8 (3.3)	5.9 (3.4)	
Range	1.1 -16.8	1.1 -16.3	1.1 - 16.8	

Table 5.2. Data logging at one-month (n = 61) and three-months (n = 51) post-hearing aid fitting

5.4.2 Factors associated with hearing aid use

Eight categorical variables (gender, aetiology of hearing loss, onset of hearing loss, additional disabilities, household income, home language, language of instruction, degree of hearing loss) and two continuous variables (age at diagnosis, age at hearing aid fitting) were identified from the departmental electronic database. After continuous variables were converted into categories, ANOVA (α level = 0.01) significantly associated six of the ten potential independent categorical variables with hearing aid use (dependent variable Y), namely gender, onset of hearing loss, additional disabilities, household income, language of instruction and degree of hearing loss (Table 5.3). These six independent categorical variables were included in two multiple linear regression models (one-month and three-months post-fitting). Based on the *p*-value of all the independent variable's coefficients, multiple linear regression models were not able to significantly predict factors that influence hearing aid use at the one-month and three-month post-fitting follow-up interval respectively (*p* - value = 0.34 and 0.51).

Table 5.3. Factors associated with hearing aid use

	Parameter		Hearing	Hearing aid use (h/day)		
Independent variable		n	Mean	SD (range)	(<i>α</i> = 0.01)	Coefficient
Gender	Male	31	5.1	3.1 (0.2 – 12.1)	< 0.01	-0.379
	Female	37	5.9	3.3 (0.3 – 14.0)		0
Aetiology of hearing loss	Syndromic	24	2.6	1.2 (0.7 – 4.9)	0.5	N/A
	Infectious	15	3.1	1.4 (1.1 – 5.2)		
Onset of hearing loss	Congenital	36	4.2	2.0 (1.8 – 11.6)	< 0.01	-0.092
	Acquired	21	5.8	3.1 (1.1 – 12.1)		0
dditional disabilities	No additional disabilities	50	6.5	3.1 (1.2 – 14.2)	< 0.001	0
	Additional disabilities	18	2.8	1.4 (0.3 – 5.2)		-2.335
lousehold income	Low	54	4.5	3.3 (0.8 – 10.2)	< 0.01	0
	(< 400.62 USD per month)					
	High	14	7.1	2.1 (3.7 – 12.4)		2.435
	(> 400.62 USD per month)					
lome language	English	34	4.8	2.2 (1.2 – 11.2)	0.2	N/A
	Other	34	5.1	3.1 (0.3 – 12.4)		
anguage of instruction	Auditory-oral	43	5.2	3.0 (1.2 – 16.8)	< 0.01	0
	Visual	25	3.8	2.1 (1.1 – 8.4)		-2.385

Degree of hearing loss	Mild-moderate	40	4.8	3.2 (0.2 – 11.2)	< 0.01	0
	(16 - 60 dBHL)					
	Severe-profound	28	5.9	3.4 (1.2 – 14.2)		0.667
	(61 - > 90 dBHL)					
Age at diagnosis of hearing loss	Toddler (0 – 2 years)	12	5.1	4.2 (1.2 – 16.8)	0.6	N/A
	Pre-schooler (3 – 6 years)	32	5.5	3.5 (0.8 – 12.4)		
	School-going (> 6 years)	15	5.0	2.1 (1.5 – 9.4)		
Age at hearing aid fitting	Toddler (0 – 2 years)	12	5.1	4.2 (1.2 – 16.8)	0.5	N/A
	Pre-schooler (3 – 6 years)	36	5.6	4.3 (1.0 – 12.4)		
	School-going (> 6 years)	17	5.2	2.2 (1.6 – 9.4)		

5.4.3 Caregiver reported outcomes

Caregivers observed their children's behaviour in the month following initial hearing aid fitting and completed the PEACH questionnaire in the week prior to their one-month follow-up appointment. PEACH questionnaires were returned by caregivers for 23 participants at the one-month follow-up appointment. Most children (78.3%; n = 18/23) reportedly wore their hearing aids either always or often. Loudness discomfort ratings indicated that most children (87%; n = 20/23) were never or seldom upset by loud sounds. Figure 5.1 depicts the frequency distribution of caregiver-reported hearing aid use and loudness discomfort ratings for 23 participants. Mean PEACH scores were higher in Quiet (73.4%) than in Noise (69.6%) (Table 5.4). Approximately half of the participants (47.8%; n = 11) showed typical overall performance based on their PEACH percentage scores (Figure 5.2). Significantly higher hearing aid use (p < 0.05) of 7.0 h/day (2.1 SD; range 3.9 – 11.2) was recorded for the *Typical Overall Performance* group (n = 11) when compared to the groups who required review (6.1 h/day; 3.9 SD; range 1.5 – 12.4) (n = 12).

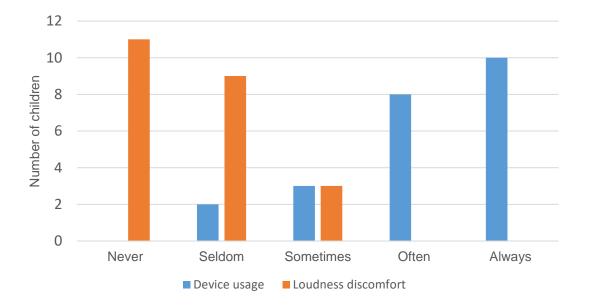


Figure 5.1: Parent-reported device use and loudness discomfort (n = 23)

Percentage score in Quiet (mean (SD); range)	73.4 (23.0); 25 - 100
Percentage score in Noise (mean (SD); range)	69.6 (23.0); 15 - 100
Percentage score Overall (mean (SD); range)	71.7 (29.0); 5 - 100

 Table 5.4.
 Mean PEACH scores for Quiet, Noise, and Overall (n = 23)

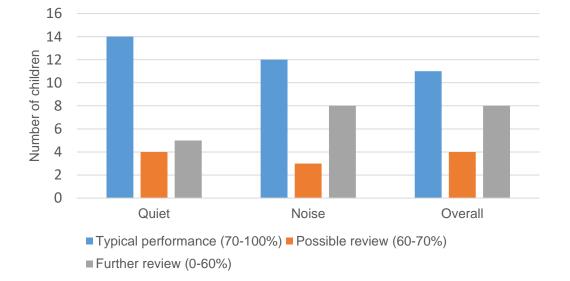


Figure 5.2: PEACH indication for Quiet, Noise, and Overall scores (n = 23)

Written feedback in the *additional comments* section of the PEACH questionnaire was obtained from 12 caregivers. All written comments were included in the analysis of the qualitative data and emerging themes were grouped into advantages and barriers to hearing aid use (Table 5.5). Caregiver-reported barriers to hearing aid use like retention challenges and bullying could be linked to sub-optimal hearing aid use in this sample.

	Theme	Examples
	Listening	"She used to put volume of TV loud before, now she playing television in soft volume"
		"The sound of water coming from a tap can be heard clearer"
		"It help him to concentrate more than he used to"
nse		"I can see a huge improvement with her and her response when one calls her or to follow instructions"
aid	Confidence and enjoyment	"She report it to teacher"
of hearing		"He has noticed the difference in story-time himself (first week already), and is quite proudly wearing the aids with no embarrassment factor at all"
of h		"She asks to wear the hearing aids without me reminding her"
		"Liking is being evidenced by his smiling when wearing them"
ntaç		"She clearly enjoys the sound of her own voice. She won't stop making sounds!"
Advantages		"He is very happy with his hearing aids and loves them"
A	Speech production	"I see an improvement with her speech as well, certain sounds that she couldn't pronounce before is now way clearer"
		"When she speaks her tone will be softer when the hearing aids are in and her tone will be louder when she removes it"
	Social interaction	"I can see a difference in my son's communication, with myself and others around him"
		"He now involves himself in the community with his friends"
e	Bullying	"A classmate threw a book against her ear"
d use		"Children grab it out of his ear and laugh"
g aid	Hearing aid retention	"During the first days of using the hearing aids, he used to dislike them. He would any and by all means have them removed"
arinç		"Due to 'rough and tumble' circumstances at after-school care, we allow him to leave them out"
hea		"The left hearing aid is getting loose often"
's to		"We not succeeding with it and it is difficult to put in his ears"
Barriers to hearing	Medical aspects	"He has complained of itchy ears often. Hearing aids are removed to itch and then replaced"
Ва		"Sometimes his ears leak and he can't wear them"

Table 5.5. Thematic analysis of additional written feedback from caregivers on the PEACH questionnaire

5.5 Discussion

This study aimed to describe hearing aid outcomes for children aged 0 - 13 years with bilateral SNHL in a low-resourced context. Daily average hearing aid use of 5.9 hours/day at the three-month follow-up interval for this study sample only just compared with average estimates for children of 5-8 hours per day (Marnane & Ching, 2015; Muñoz et al., 2015), and fell short of the recommended 10 hours per day for optimal language development (Tomblin et al., 2015). There was a significant increase in hearing aid use at the three-month follow-up interval when compared to the initial one-month follow-up. Findings from a previous study on predictors of change in hearing technology use in Australia showed consistent use was established for 62% of children within the first year of amplification, and 71% of children at three years postfitting (Marnane & Ching, 2015). A recent South African study on predictors for hearing technology use found a higher average of hearing device use (9.4 hours/day) over an eight-year period (Booysen et al., 2021).

The average age of hearing loss diagnosis for children with permanent congenital or early-onset hearing loss was 31.6 months (n = 36), which highlights the consequences of limited new-born hearing screening programmes in the public sector of South Africa (Meyer et al., 2012; Swanepoel et al., 2009). Delayed diagnosis of childhood hearing loss results in delayed initiation of intervention, which leads to poorer speech-language and academic outcomes (Marschark et al., 2015).

Lower household income was associated with decreased hearing aid use in this sample. Nearly 80% of children in this study sample came from low-income households (< 400.62 USD per month). In a recent South African study on hearing technology use in children, it was reported that children who required subsidized batteries due to poor socio-economic circumstances had reduced hearing technology use compared to those who were able to self-procure (Booysen et al., 2021). The setting for the current study was a centralised tertiary paediatric hospital, where the majority of patients had to travel long distances to access audiological services. Lack of access to follow-up audiological services (such as collecting hearing aid batteries

and hearing aid repairs) could have contributed to the poorer average daily hearing aid use in this sample. In a 2015 longitudinal study looking at hearing technology use for children at age three years, higher socio-economic status was associated with higher device use (Marnane & Ching, 2015). Low-income households often have less access to resources in terms of support-structures and experience more pressing needs (such as food-security) than hearing aid maintenance (Wiseman & Warner-Czyz, 2018).

In the current study, only 17.6% of children were placed at an education facility where audiology services were available onsite (inclusive mainstream and hearing-impaired skills schools in the Western Cape). Education settings where onsite audiologists are available could increase access to technology-related support and rehabilitation, which could in turn contribute to hearing aid use (Booysen et al., 2021). One in five (20.6%) children in this study were not old enough to attend formal schooling and were either looked after by family members at home or attending a crèche. Challenges around hearing aid use have been reported for children in daycare settings, with consistent use reported for 50%, 40%, and 70% of 6, 12, and 24-month old's respectively (Walker et al., 2015). An auditory-oral mode of communication was reported as a significant predictor of increased hearing technology use in children in a recent sample in the Western Cape Province of South Africa (Booysen et al., 2021). Nearly one third (27.95%) of children in the current study used SASL as primary mode of communication. Average daily hearing aid use in this subgroup was four hours, suggesting reduced necessity of auditory access through hearing aids for learning. Decreased hearing aid use is associated with limited access to healthcare services (Wiseman & Warner-Czyz, 2018) lack of perceived benefit for learning through audition (Muñoz et al., 2015) and contexts where the use of hearing technology is not enforced (Walker et al, 2015).

Most caregivers (78.3%) for the PEACH subgroup (n = 23) reported that their children wore hearing aids either always or often, however, the average data logging in this subgroup was 6.6 h/day. Parents frequently over-estimate hearing aid use when compared with data logging information stored in the hearing aid (Walker et al., 2015).

Nearly half of the children (47.8%) showed typical overall performance based on their percentage PEACH scores. Significantly higher hearing aid use of 7.0 h/day was recorded for the *Typical Overall Performance* group when compared to the group who required review. In a 2015 study on hearing aid and cochlear implant use in young children, higher PEACH scores were associated with higher device use scores (Marnane & Ching, 2015). More than half (52.2%) of children in this study whose caregivers completed the PEACH required review based on their overall PEACH percentage scores. PEACH scores in Quiet (73.4%) were higher than in Noise (69.6%). Noisy environments have a negative impact on the listening and learning opportunities for children with hearing loss, both at home and in educational settings (Benítez-Barrera, Grantham, & Hornsby, 2020). Improving the signal-to-noise-ratio for children with hearing loss should be an important goal to mitigate the negative effect of noisy environments (Benítez-Barrera et al., 2020). One in four (26.5%) children in the current study presented with additional disabilities, which was associated with significantly lower hearing aid use, and likely contributed to poorer functional listening performance (Walker et al., 2015). Audiologists who provide intervention for children with additional disabilities should work collaboratively within a multi-disciplinary team to find innovative solutions for increased hearing aid use and functional listening outcomes.

Qualitative caregiver reported feedback revealed themes of perceived advantages of hearing aid use, and barriers to hearing aid use. Hearing healthcare professionals play an important role in helping caregivers to address challenges relating to the ongoing management of their child's hearing loss (Muñoz et al., 2015) so that consistent hearing aid use can be achieved. In a previous study regarding pediatric hearing aid use, caregiver challenges regarding navigating daily hearing aid management were associated with hearing aid use (Muñoz et al., 2015) and should be addressed continuously by the managing audiologist. Caregivers noticed and reported benefits such as improved confidence and enjoyment of hearing aid use, better speech production, and increased social interaction within one month after fitting. Caregiver perception of hearing aid benefit is an important indicator for hearing aid use (Muñoz et al., 2015). Caregiver-reported barriers included difficulty with keeping the hearing aids in their children's ears. Solutions such as retention caps for younger children in

situations like traveling in car seats could alleviate some of the difficulty caregivers experience with facilitating hearing aid use (Booysen et al., 2021).

Involving older children to become active agents and advocates in their management plan could address barriers to technology use such as bullying. Tools like *My Hearing Explained* from the Ida institute can be used to elicit a conversation about hearing, speech understanding and listening energy (Ida Institute, 2021). Parents and children can engage with the audiologist and draw on their personal experiences and challenges to create a better communication situation for the entire family. Children with hearing loss and their families should be empowered and given a voice to participate in their intervention, so that barriers like bullying can potentially be replaced with skills like assertiveness and self-advocacy (Ida Institute, 2021).

The sample size in the current study was limited and therefore regression models were likely underpowered to identify relationships between independent variables and hearing aid use. Bigger sample sizes could contribute to the knowledge base on predictors of hearing aid use in children within a low-resourced context (Booysen et al., 2021). The wide age distribution in this sample had an impact on generalising findings. Future studies on hearing aid outcomes in LMICs should consider age-group-specific distribution pockets. Although there was a significant improvement in average hours of hearing aid use at the three-month follow-up interval, outcomes were only recorded at one- and three-months post-fitting. Future longitudinal data on hearing aid use in the LMIC context will be valuable to determine whether hearing aid use increases over a longer period, so that predictors and barriers to hearing aid use can be described more comprehensively.

5.6 Conclusion

Outcomes of children with SNHL fitted with binaural hearing aids at a paediatric public hospital in South Africa demonstrated sub-optimal average daily hearing aid use of 5.9 hours, which increased from month one to three. At-risk groups like children from low-income households and those with additional disabilities require more support to ensure optimal hearing aid use. Aural/oral performance was typical for nearly half of the children in this sample, and higher hearing aid use resulted in better functional listening performance. Caregivers report hearing aid benefit within one month of fitting. Hearing healthcare practitioners should empower caregivers and children to participate actively in their intervention to identify potential address barriers to hearing aid use early on.

CHAPTER 6

DISCUSSION AND CONCLUSION

The aim of this chapter is to contextualize the results obtained in Studies I-III, discuss clinical implications, and critically evaluate the research conducted within public healthcare facilities in the Western Cape Province of South Africa in terms of strengths and limitations. Additionally, future research recommendations are proposed.

6.1 Summary of findings

The aim of this thesis was to 1) describe the profile and aetiological factors associated with childhood hearing loss; 2) to explore the effects of a decentralised model of hearing healthcare through district hearing screening; and 3) to describe the hearing aid outcomes of children with bilateral SNHL in the Western Cape public healthcare system, South Africa.

This research project comprised three studies. Study I described the nature, associated risk factors and age of diagnosis for childhood hearing loss in a South African cohort of 240 children from the RCWMCH between 1 January 2019 – 31 July 2019. The predominant type of hearing loss was conductive and treatable (64.6%). More than half (51.8%) of bilateral sensorineural hearing losses were of a profound degree. The most prominent risk factor for conductive hearing loss was OM, for SNHL it was a family history of childhood hearing loss, and for ANSD it was hyperbilirubinaemia. Approximately one third of patients (27.1%) with SNHL did not have any associated risk factors. The mean age of diagnosis of permanent congenital or early-onset hearing loss was 31.4 months (22.8 SD; range 2 - 72), with a mean delay of nine months (13.2 SD; range 0 - 60) between age of suspicion and diagnosis of hearing loss (n = 93).

Study II compared a centralised tertiary model of hearing healthcare with a decentralised model through district hearing screening for children in the Western

Cape Province, South Africa. At the district hospital, attendance rates were significantly higher (p < 0.001) and travel distance was significantly shorter (p < 0.001). The number of referrals to the tertiary hospital decreased significantly during the intervention period (p < 0.001). Most children in both the tertiary and district hospital groups (78.7% and 80.4%, respectively) passed initial hearing screening bilaterally. The results of Study II highlight that hearing screening should be conducted at the appropriate level of care to increase access, reduce patient travelling distances and associated costs, and reduce the burden on tertiary-level hospitals.

Study III described hearing aid outcomes for 68 children with bilateral SNHL at a paediatric public hospital in South Africa in terms of average daily use and oral/aural performance as measured by the PEACH questionnaire. Average daily hearing aid use increased significantly (p < 0.05) from one-month (5.0; 3.0 SD; range 0.3 - 14.0) to three-months post-fitting (5.9; 3.4 SD; range 1.1 – 16.8). Average PEACH scores were higher in Quiet (73.4%) than in Noise (69.6%). More than half (52.2%) of children required review based on their overall percentage PEACH scores. Higher average daily hearing aid use was significantly associated with higher overall PEACH scores (p < 0.05). Neuro-typically developing children had significantly higher hearing aid use than children with additional disabilities (p < 0.001). Qualitative caregiver feedback revealed themes pertaining to advantages (improved listening skills, speech production and social interaction skills, confidence, and enjoyment of using hearing technology) and barriers (bullying and retention challenges) to hearing aid use.

6.2 Clinical implications

A number of clinical implications emerged from the research. These clinical implications are discussed according to three themes: Unique risk factors for childhood hearing loss in low-resourced contexts; equitable access to hearing healthcare services in LMICs; and context-specific hearing aid outcomes for children accessing the public healthcare system in South Africa.

6.2.1 Risk factors for childhood hearing loss in low-resourced contexts

The large proportion of preventable hearing losses (64.6%) in the Study I sample highlights the importance of maximising primary healthcare efforts to treat preventable causes timeously. Annually, acute middle ear infection affects over 700 million people globally, which comprises mostly children under five years (Monasta et al., 2012). Sub-Saharan Africa has one of the highest incidence rates of middle ear infection at an estimated 43% compared to 3.64% in central Europe (Monasta et al., 2012). The variation can be ascribed to factors like malnutrition and poor socioeconomic circumstances (WHO, 2021). Medical and surgical management for conditions like wax and OM can be cost–effective in the long term by reducing resultant hearing loss and morbidity due to complications (Venekamp, Mick, Schilder, & Nunez, 2018; Shaikh et al., 2017).

Other preventable causes of childhood hearing loss, such as vaccine preventable causes including meningitis and rubella, made up nearly 6% of all hearing losses in the Study I sample. Furthermore, pre-, peri-, and post-natal complications including hyperbilirubinaemia, hypoxia, and very low birth weight (≤ 1 500g) constituted more than 10% of the risk factors for hearing loss in the Study I sample. Nearly 60% of childhood hearing loss causes can be prevented through public health strategies such as early identification of ear disease, improved pre-, peri-, and post-natal care, and comprehensive vaccine rollout (WHO, 2021). The WHO estimates that more than 19% of childhood hearing loss can be prevented by immunization against rubella and meningitis (WHO, 2017). Countries should consider these factors when planning for immunization coverage and should ensure that effective immunization policies are implemented (WHO, 2021).

Study I demonstrated that in a sample of 240 children, nearly one-third (27.1%) of those with SNHL had no risk factors for childhood hearing loss, highlighting the importance of universal versus risk-based screening coverage. An estimated 50–60% of infants with permanent congenital or early-onset hearing loss have risk factors (Hyde, 2005); therefore, an unacceptably high percentage of babies with hearing loss

will be missed by using a risk-based newborn hearing screening approach (Bamford, Fortnum, Bristow, Smith, Vamvakas, & Davies, 2007). Hearing screening in newborns provides significant advantages in terms of preventing delays in diagnosis and initiating intervention early, thereby ensuring improved language and cognitive development (Neumann, Tavartkiladze, Bu, & White, 2019; Yoshinaga-Itano, 2004). The cost–effectiveness of newborn hearing screening is well documented in studies from high-income countries, as well as middle-income countries like China, India, Nigeria, and the Philippines (Sharma, Gu, Ching, Marnane, & Parkinson, 2019). In India, a cost analysis revealed life-time savings of over 500 000 International Dollars per identified case of hearing loss (Burke, Shenton, & Taylor, 2012).

The age of diagnosis of permanent congenital or early-onset hearing loss was severely delayed in Study I (mean age 31.4 months) and Study III (mean age 31.6 months), undermining prospects of positive outcomes through early intervention. Parents are often unaware of the need for newborn hearing screening, and educating parents to identify risk factors for hearing loss, as well as seeking hearing healthcare services for their child timeously, is important to mitigate the adverse effects associated with childhood hearing loss (Olusanya, Emokpae, Renner, & Wirz, 2009). There is a lack of evidence regarding the status of newborn hearing screening programmes in the private and public healthcare sectors in South Africa (Bezuidenhout, Khoza-Shangase, De Maayer, & Strehlau, 2021). Available evidence suggests that limited success has been achieved within existing programmes (Maluleke, Khoza-Shangase, & Kanji, 2018; Swanepoel et al., 2009), with only 27% of hospitals in the public sector offering some form of newborn hearing screening (Theunissen and Swanepoel, 2008). Even in the private healthcare sector of South Africa, where resource-constraints are less severe, there is a significant delay in the diagnosis and intervention of childhood hearing loss (Meyer, Swanepoel, & Le Roux, 2014). Hearing healthcare providers should advocate for the mandated rollout of universal newborn hearing screening coverage in both the public and private healthcare sectors of South Africa.

The high proportion of preventable causes for hearing loss in the Study I sample implies that awareness and training for primary-level hearing healthcare professionals,

including doctors and nurses in LMICs, is essential. Effective first-line treatment for vaccine preventable illnesses, as well as middle ear pathology such as AOM, OM, and CSOM, can minimise the adverse effects of hearing loss on speech – and language development in children. A great need remains for training hearing healthcare practitioners to guide and impart accurate and relevant information to parents, and to ensure timely diagnosis and appropriate interventions and support for families of children with hearing loss (Olusanya, 2015; Ravi, Gunjawate, Yerraguntla, & Rajashekhar, 2018).

6.2.2 Equitable access to hearing healthcare services in LMICs

Unaddressed hearing loss poses an annual global cost (including healthcare, education, and societal costs) of over 980 billion USD (WHO, 2021). Many of these costs can be reduced through strategies such as identifying and treating preventable causes of hearing loss timeously, making hearing healthcare accessible to all people (especially those in underserved communities), implementing context-specific newborn hearing screening programmes, and embarking on advocacy and awareness campaigns (WHO, 2021).

Decentralised hearing services within communities and through primary healthcare services are strategies aimed at increasing access and decreasing costs (Suen et al., 2019). In Study II, the number of direct referrals for initial hearing screening from the district hospital catchment area to the tertiary hospital significantly decreased after implementation of the decentralised hearing screening project at the district hospital. The decreased number of referrals to the tertiary hospital for initial hearing screening support decreased waiting times and improved capacity to provide specialised diagnostic hearing assessments and intervention to patients requiring tertiary-level care. In addition, patients receiving hearing screening services at the district hospital had to travel a significantly shorter distance, and attendance rates for initial- and follow-up hearing screening was significantly higher when conducted at a district-level as opposed to a central, tertiary-level.

Providing hearing screening at a district level in Study II increased access to medical treatment for all children who presented with middle ear pathology as evidenced by abnormal tympanometry results on the day of initial OAE screening. These children were assessed and treated by the paediatrician on the same day, instead of waiting for months to get an ENT appointment at the tertiary hospital. Thus, middle ear pathology was treated timeously and effectively at a more appropriate level of care, decreasing the added burden to long tertiary waiting lists. Primary healthcare is an important strategy employed in South Africa to provide more accessible patient-centred services closer to home (Dookie & Singh, 2012). Community delivered hearing healthcare models have been identified as an important strategy to increase the accessibility and affordability of hearing healthcare in underserved communities (Suen et al., 2019; Wilson et al., 2017). Hearing healthcare must be accessible at all levels of care and should be integrated with national health services (WHO, 2021).

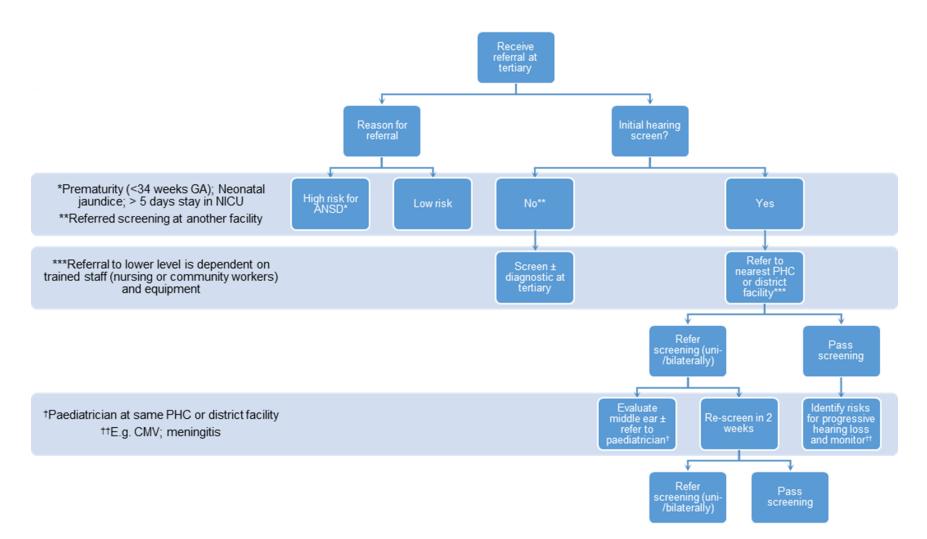
The rising global prevalence of hearing loss is estimated at 2.5 billion by 2050 (1 in every 4 people) (WHO, 2021). Urgent public health action is vital to minimise the burden of this projected growth on healthcare, education, and economic systems (WHO, 2021). There is a great demand for hearing healthcare services, yet they are often unavailable at primary-level facilities due to the lack of equipment and infrastructure in low-resourced settings (Fagan & Jacobs, 2009; Mulwafu et al., 2017). Providing ear and hearing care is further impeded by the long distances that people from rural communities need to travel to gain access (Bright et al., 2017; Taber, Leyva, & Persoskie, 2015). The inaccessibility of hearing healthcare services at a primary- or district-level, which adds severe strain on tertiary-level specialised services, may be alleviated by decentralising services. The advent of tele-audiology and portable, boothless technology-based solutions, enable hearing healthcare screening to be conducted in more remote settings (such as schools and communities), with limited training and resources required (WHO, 2021; Yousuf-Hussein et al., 2016).

The results of Study II highlight that hearing screening should be conducted at the appropriate level of care to increase access, reduce patient travelling distances and associated costs and reduce the burden on tertiary-level hospitals. Based on the

results of Study II, a service delivery model for decentralised hearing screening in the Western Cape Province of South Africa is proposed in Figure 6.1. In the Western Cape Province of South Africa, limited primary- and secondary-level hearing healthcare services are available. Therefore, referrals for primary-level services, like hearing screening, is often received at central, tertiary-level facilities. Direct referrals to a central tertiary hospital imply that parents and caregivers must travel further to access hearing healthcare infrastructure, which may in turn lead to poor follow-up rates, late diagnoses, and late access to hearing technology. The proposed decentralised service delivery model for the Western Cape Province (Figure 6.1) delineates recommended steps to follow from direct referral at tertiary (often due to limited availability of primary-and secondary-level services). The model is dependent on the availability of trained staff (nurses or community healthcare workers) to conduct hearing screening, as well as appropriate equipment at the site.

6.2.3 Hearing aid outcomes for children using the public healthcare system, South Africa

The results of Study III suggests that decreased hearing aid use in children who live in low-resourced contexts is associated with limited access to healthcare services, lack of perceived benefit for learning through audition, and educational contexts where the use of hearing technology is not enforced. Study III also indicated that at-risk groups like children from low-income households and those with additional disabilities require more support to ensure optimal hearing aid use, as higher hearing aid use resulted in better functional listening performance in the Study III sample.



ANSD – Auditory Neuropathy Spectrum Disorder; CMV – Cytomegalovirus; GA – Gestational Age; NICU – Neonatal Intensive Care Unit; PHC – Primary Healthcare Clinic

Figure 6.1: Proposed decentralised service delivery model for hearing screening in the Western Cape Province, South Africa

In the WHO African Region, 90% of people who need hearing technology do not have access to it (WHO, 2021). Appropriate use of hearing aids could reduce the YLD associated with hearing loss by 59% (WHO, 2021). It is therefore important to provide support to caregivers of children with hearing loss, so that consistent hearing aid use is established early on. Reporting on hearing aid outcomes and factors that are associated with increased or decreased hearing aid use is an important aspect of paediatric hearing loss management, as hearing aid fitting is the most common recommendation following diagnosis of childhood hearing loss (HPCSA, 2018; JCIH, 2019). Access to sound via hearing technology is considered the most critical factor to maximise a child's potential to reach age-appropriate auditory-based outcomes through listening (Flexer & Wolfe, 2020).

Although multiple linear regression models in Study III could not significantly predict hearing aid use in children, likely due to a small sample size, several factors were significantly associated with hearing aid use. These factors, along with caregiver-reported barriers to hearing aid use with recommended clinical strategies to improve hearing aid use, are discussed in Table 6.1. Results from Study III serves as a reminder that comprehensive hearing healthcare for children is an ongoing process, and optimal hearing outcomes is dependent on continuous follow-up and rehabilitation after hearing aid fitting (Erbasi, Scarinci, Hickson, & Ching, 2018; Tomblin, Oleson, Ambrose, Walker, & Moeller, 2020).

Factors associated with	Clinical strategy to improve hearing aid use
reduced hearing aid use	
Additional disabilities	Parents of children with hearing loss report that behavioural challenges are difficult to manage, especially if the child has additional developmental conditions (Whicker, Muñoz, & Nelson, 2019). One in four (26.5%) children in Study III presented with additional disabilities, which was associated with significantly lower hearing aid use, and likely contributed to poorer functional listening performance (Walker et al., 2015). Audiologists who provide intervention for children with additional disabilities should work collaboratively within a multi-disciplinary team to find innovative solutions for increased hearing aid use and functional listening outcomes. Audiologists should also recognize that full-time hearing aid use may be an unrealistic goal (McCreery & Walker, 2017), and should tailor their recommendations to promoting quality hearing and listening experiences rather than focusing on quantity of hearing hours (Booysen et al., 2021), especially in children with additional disabilities.
Lower household income	Nearly 80% of children in Study III came from low-income households (< 400.62 USD per month). In a recent South African study on hearing technology use in children, it was reported that children who required subsidized batteries due to poor socioeconomic circumstances had reduced hearing technology use compared to those who were able to self-procure (Booysen et al., 2021). Low-income households often have less access to resources in terms of support-structures and experience more pressing needs (such as food-security) than hearing aid maintenance (Wiseman & Warner-Czyz, 2018). Audiologists should identify families with limited resources and should put supportive strategies in place such as issuing bulk batteries to limit hospital visits, and scheduling audiology visits on the same day as other appointments where possible. Recent eHealth solutions offer novel opportunities for practical problem-solving to support parents with hearing aid management (Muñoz et al., 2017, 2020; Whicker et al., 2020), and can help to overcome challenges with access to hearing healthcare services.
Visual language of instruction	An auditory-oral mode of communication was reported as a significant predictor of increased hearing technology use in children in a recent sample from the Western Cape Province of South Africa (Booysen, 2021). Nearly one-third (27.95%) of children in Study III used SASL as primary mode of communication. Average daily hearing aid use in this subgroup was only four hours, suggesting

Table 6.1. Factors associated with reduced hearing aid use and clinical strategies to improve hearing aid use in children

	reduced necessity of auditory access through hearing aids for learning. Audiologists and teacher of the Deaf should consider the added burden of responsibility that hearing technology places on families and should evaluate whether children who use SASL as mode of communication benefit from hearing aid use through caregiver and teacher validation questionnaires.
Less severe degrees of hearing loss	Frequent monitoring of hearing aid use is often required for children with milder degrees of hearing loss (Ambrose et al., 2020; Muñoz et al., 2019). The concept of listening in noise and resultant listening fatigue (Hoffman et al., 2019) should be included in
	the counselling of children with hearing loss and their caregivers. Greater awareness of the negative impact of inconsistent hearing aid use in challenging listening environments might lead to increased use.
Retention challenges	Audiologists should liaise with hearing aid companies and offer standard retention solutions such as retention caps to young children to increase hearing aid use (Wiseman & Warner-Czyz, 2018). Retention solutions should be part of the standard hearing aid fitting package for the paediatric population.
Bullying	Hearing loss is frequently associated with internal behavioural problems like low self-esteem (Vas, 2017). Even when hearing loss is addressed and managed, the associated stigma may prevent consistent use of hearing technology (Mousavi, Movallali, & Nare, 2017).
	Involving older children to become active agents and advocates in their management plan could address barriers to technology use such as bullying. Tools like <i>My Hearing Explained</i> from the Ida institute can be used to elicit a conversation about hearing, speech understanding and listening energy (Ida Institute, 2021). Parents and children can engage with the audiologist and draw on their personal experiences and challenges to create a better communication situation for the entire family. Children with hearing loss and their families should be empowered and given a voice to participate in their intervention, so that barriers like bullying can potentially be replaced with skills like assertiveness and self-advocacy (Ida Institute, 2021). The stigma associated with hearing loss, the use of hearing technology and sign language can be alleviated through increased awareness within communities, by empowering people with hearing loss, and by including people with hearing loss in policy discussions (WHO, 2021).

6.3 Research strengths and limitations

Appropriate interpretation, as well as a comprehensive evaluation of research findings within the framework of its strengths and limitations, is critical to maintain academic integrity (Leedy & Ormrod, 2020).

6.3.1 Research strengths

Several strengths were identified throughout this research project.

Study I provided one of the first reports on the profile of childhood hearing loss in the Western Cape Province of South Africa, by describing the nature, associated risk factors, and age of diagnosis for childhood hearing loss in a cohort from RCWMCH. Different countries and local communities will have different presentation of childhood hearing loss, and therefore different priorities (WHO, 2021). It is important for local hearing healthcare professionals to agree on service specifications, and to build on national frameworks to match the relevant epidemiology to the services needed in order to reduce the burden of hearing loss for childhood hearing loss in Charpe & Seewald, 2016; WHO, 2021). Study I contributed to the limited available data on the profile of childhood hearing loss in South Africa, and the results could be applicable to other similar low-resourced contexts.

Ecological validity was attained during Study II, as the pragmatic study design enabled an actual evaluation of existing services without any experimental aspects. In Study II, there were two distinct periods (control and intervention) with different clinical services (routine audiological assessment at the tertiary-level hospital during the control period, and decentralised OAE-screening at a district-level during the intervention period), allowing for comparison of outcomes due to the difference in service provision. The first South African report on a range of predictor variables for hearing technology use was only recently published (Booysen et al., 2021). Study III contributed to the limited available South African data regarding hearing technology use in children (Booysen et al., 2021). The results involved children from a low-resourced context, where delayed hearing loss diagnosis and intervention is typical (Le Roux et al., 2015; Swanepoel et al., 2013). Available reports about hearing technology use in children primarily originate from high-income countries like the United States of America (Tomblin, Walker, et al., 2015) and Australia (Ching et al., 2018). Considering that the greatest burden of childhood hearing loss exists in LMICs like South Africa (WHO, 2021), study findings from high-income contexts are not always applicable to lowresourced contexts. The results of Study III contribute to the limited existing South African data and could apply to similar contexts where information on hearing aid outcomes in children with hearing loss is needed most. Furthermore, the dependent variable in Study III (hearing aid use) was measured objectively in terms of data logging, instead of relying on subjective reports from parents, which contributed to the validity of data (Booysen, 2021).

Although predictors of hearing aid use could not be calculated as statistically significant in Study III (likely due to the small sample size), several factors that were associated with hearing aid use were identified (gender, onset of hearing loss, additional disabilities, household income, language of instruction, and degree of hearing loss). These factors are important to consider in the management of vulnerable children from low-income households, so that hearing healthcare professionals can put strategies in place to improve consistent hearing aid use in children to ensure optimal auditory exposure necessary for spoken language development (Tomblin et al., 2015). The qualitative branch of Study III enabled data collection on important caregiver perceptions. Caregivers noticed and reported benefits such as improved confidence and enjoyment of hearing aid use, better speech production, and increased social interaction within one month after fitting. Caregiver perception of hearing aid benefit is an important indicator for hearing aid use (Muñoz et al., 2015), and hearing healthcare professionals should provide opportunities for parents to communicate advantages and barriers to hearing aid use to plan for effective management.

This research project comprised of different research methods, with elements of quantitative and qualitative data. The quasi-experimental pragmatic research design in Study II enabled clinical, context-specific evaluation of decentralized hearing screening services. Collecting quantitative and qualitative data for Study III allowed for triangulation, which contributed to increased credibility and validity of the research findings. Triangulation ensures that fundamental bias arising from using a single method are overcome (Leedy & Ormrod, 2020). The qualitative data gathered from caregivers of hearing aid users facilitated a deeper understanding of real-world barriers to hearing aid use, to ultimately achieve better functional listening performance in children.

6.3.2 Research limitations

Several limitations of this research project are also acknowledged below.

Studies I and III relied on retrospective data review, which could have contributed to incomplete or missing data. For example, capturing data-logging information at oneor three-months post-hearing aid fitting could have been impacted either by patient non-attendance at the scheduled appointment or the audiologist omitting to record the data in the patient's hospital folder. Retrospective cohort studies are generally classified as level IV evidence and are typically inferior to randomized control studies (Leedy & Ormrod, 2020).

Study II measured outcomes during two distinct periods: control at the tertiary-level hospital, and intervention at the district-level hospital. While this pragmatic quasi-experimental design allowed for comparison of outcomes due to a difference in service provision, it is important to recognize that the outcomes for the control period were obtained through retrospective review, which subjects the data to the same limitations described above in terms of missing data.

Another limitation of Study II was that tertiary-level audiologists conducted the hearing screening project at the district hospital during the intervention period, which implies that test facilitators were used that were not representative of a minimally trained healthcare workforce cadre (for example community healthcare workers).

The sample size in Study III was limited and therefore regression models were likely underpowered to identify relationships between independent variables and hearing aid use. Furthermore, the wide age distribution in the Study III sample had an impact on generalizing findings related to hearing aid outcomes.

Although there was a significant improvement in average hours of hearing aid use at the three-month follow-up interval in Study III, data logging was only measured at two points in time (one-month and three-months post-fitting). Multiple measurements of data logging information over a longer period improves the measured accuracy of daily hearing aid use (Leedy & Ormrod, 2020).

The Study III sample encompassed a relatively small group of children, which could have contributed to potential underrepresentation of certain groups of children. Outcomes were only measured for children with bilateral SNHL (as opposed to children with other types of hearing losses using a range of hearing technologies). Significant and non-significant findings are not always generalizable and robust when small sample sizes are used (Leedy & Ormrod, 2020). Furthermore, non-responder bias could have occurred during Study III, as only 23 caregivers completed and returned the PEACH questionnaire at the one-month follow-up appointment.

6.4 Recommendations for future research

Research projects give rise to additional questions that should be addressed in future studies (Leedy & Ormrod, 2020). The nature and associated aetiological factors for childhood hearing loss were only described at one site in the Western Cape Province of South Africa. Only preliminary data on the profile of childhood hearing loss is

available from the Gauteng Province (Swanepoel, 2013; Le Roux et al., 2015). Future research should consider multi-site studies to gain more insight into the profile of childhood hearing loss from a more representative sample across different provinces in South Africa. Information regarding unique risk factors for childhood hearing loss in LMICs is important so that these infants can be referred for early hearing screening to identify hearing loss timeously (Olusanya, 2011).

In a study conducted in two South African provinces, findings revealed a lack of standardised newborn hearing screening implementation at primary, secondary, and tertiary levels of public healthcare (Khoza-Shangase, Kanji, Petrocchi-Bartal, & Farr, 2017). Some of the reasons for the sporadic implementation included limited access to equipment and human resources, financial constraints, and a lack of clear political direction by the South African government to mandate newborn hearing screening. These findings, as well as the results of Study I in terms of delayed diagnosis and intervention, have highlighted the need to ensure that context-specific studies regarding newborn hearing screening are conducted to ensure that contextually relevant strategies are put in place to promote early detection, identification, and intervention for hearing loss in children (Khoza-Shangase et al., 2017).

Due to the feasibility pilot-project nature of Study II, tertiary hospital audiologists conducted the hearing screening at the district hospital. Mobile technologies are evolving rapidly, and remote solutions are increasingly used to overcome accessibility issues (Muñoz et al., 2020; Whicker et al., 2020). Future studies should assess the training needs of community healthcare workers and nurses to conduct hearing screening at district hospital facilities, so that sustainable and accessible hearing screening services become the norm. The provision of ear and hearing healthcare should be based on a model that empowers communities, strengthens governance and accountability, prioritises care at primary and community levels, and is coordinated across sectors (WHO, 2021). The premise of task-sharing through community-based hearing screening programmes has been proposed as a way to improve access to hearing healthcare (Suen et al., 2019; Yousuf Hussein et al., 2016). Task-sharing involves the redistribution of clinical tasks among different healthcare

teams. Task-sharing aims to re-allocate tasks appropriately (through community healthcare workers and nurses at a primary level of care) to make more efficient use of available human resources (Bright et al., 2019; Suen et al., 2019). When implementing task-sharing, patients may have more access to services such as identification and management of common ear conditions like wax and acute or chronic OM, as well as identification of hearing loss through hearing screening (WHO, 2021). Community healthcare workers and nurses can be trained to screen for hearing loss using mobile health technology via home-based visits to reach vulnerable communities in LMICs, thereby improving access to hearing healthcare professionals in South Africa (Yousuf Hussein et al., 2016).

Larger sample sizes could contribute to the knowledge base on predictors of hearing aid use in children within a low-resourced context (Booysen et al., 2021), and future studies on hearing aid outcomes in South African children should consider age-group specific distribution pockets to accurately describe outcomes according to developmental level. Average daily hours of hearing aid use were only recorded at one- and three-months post-fitting in Study III. Future longitudinal data on hearing aid use in low-resourced contexts will be valuable to determine whether hearing aid use increases over a longer period, so that predictors and barriers to hearing aid use can be described more comprehensively.

Study III investigated outcomes of children using behind-the-ear hearing aids. A recent South African study found limited hearing technology use in children with bone anchored devices compared to those with behind-the-ear hearing aids or cochlear implants (Booysen, 2021). Measuring outcomes of children using bone conduction devices would be valuable, especially in light of the high prevalence of middle ear disease in LMICs (WHO, 2021).

6.5 Conclusion

This research project highlighted that hearing screening services in the public healthcare sector of South Africa should be prioritised alongside primary health care efforts to reduce preventable risks for hearing loss, and to minimise the adverse impact of unaddressed childhood hearing loss. Hearing screening services should be decentralized to an appropriate level of care to increase access to hearing healthcare, reduce patient travelling distances and associated costs, and reduce the burden on tertiary-level hospitals. Outcomes of South African children with SNHL fitted with binaural hearing aids demonstrated increased average daily hearing aid use from onemonth to three-months post-fitting. Hearing healthcare practitioners should empower caregivers to participate actively in the intervention process to identify and address potential barriers to hearing aid use early on, especially for vulnerable groups from low-income households and children with additional disabilities. Contextual knowledge regarding unique risk factors for hearing loss, barriers, and strategies to overcome inaccessibility of hearing healthcare services, and factors associated with hearing aid use in children, can guide hearing healthcare professionals to deliver effective, evidence-based paediatric hearing healthcare services in low-resourced countries.

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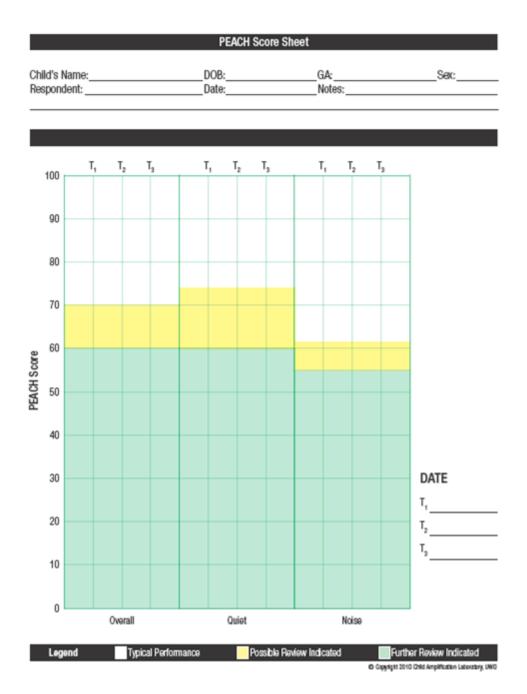
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Appendix A:

Parents' Evaluation of Aural/Oral Performance of Children





Notes: Enter the total scores by marking the percentage score on the graph corresponding to each subscale. Children with scores in the green shaded region warrant further review. A possible review is indicated when children have scores in the yellow shaded region. Children with scores in the unshaded region are performing as expected. Use the vertical lines (T1, T2, T3) within each subscale to indicate scores on different dates and note the date in the legend provided.

Pe	s' Evaluation of Au rformance of Child (P.E.A.C.H.) oped by Teresa Ching & Mar	ren
Child's Name:	Your Name:	
D.O.B:	Interviewer:	



Parents' Evaluation of Aural/Oral Performance of Children

(P.E.A.C.H.)

Developed by Teresa Ching & Mandy Hill

What is the PEACH?

 The PEACH (<u>P</u>arents' <u>E</u>valuation of <u>A</u>ural/oral performance of <u>C</u>hildren) is a questionnaire designed to record how your child is hearing and communicating with others when using his/her hearing aids and/or cochlear implant. We ask you to observe your child's listening behaviour in everyday life and give a rating in relation to a range of hearing and communication scenarios.

The PEACH is not a test. Remember even normal hearing people have some difficulty hearing in some situations. Children's listening skills improve as they grow and develop and as they get more listening practice.

Why use the PEACH?

 The PEACH is used to evaluate the effectiveness of your child's hearing aids and/or cochlear implant. Your PEACH ratings will be used to build a picture of your child's functional performance in everyday life situations. The results can be used by your child's audiologists to tailor audiological intervention to address the specific difficulties experienced by your child. The PEACH scores collected at several intervals over time can also be used to monitor your child's progress with intervention.

How do I do it?

- Think about your child's behaviour over the past week in relation to each question.
- Give a rating, based on the estimated percentage of time that your child displays the described behaviour.

What happens next?

 After you return a completed PEACH, a researcher may contact you to talk through your ratings. The researcher may ask you further questions to make sure they have a thorough understanding of the abilities and needs of your child.

Results from the PEACH will be used to monitor your child's progress. The information will also be passed onto your child's audiologist to guide intervention.

Pre-Rating Checklist

	Yes	No
Has the child been wearing his/her hearing aids and/or cochlear implant?		
Has the child been well/healthy?		
Have the child's hearing aids and/or cochlear implant been working properly?		
If the PEACH is used to assess performance when aided, it should only be completed when	en the answer to a	all of the

If the PEACH is used to assess performance when aided, it should only be completed when the answer to all of the above items is YES.

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PEACH - 2 -

	Question	Never 0%	Seldom 1 - 25%	Sometimes 26 - 50%	Often 51 - 75%	Always 75-100%
1.	How often has your child worn his/her hearing aids and/or cochlear implant?	0	1	2	3	4
2.	How often has your child complained or been upset by loud sounds?	4	3	2	1	0
3.	When you call, does your child respond to his/her name in a quiet situation?	0	1	2	3	4
4.	When asked, does your child follow simple instructions or do a simple task in a quiet situation?	0	1	2	3	4
5.	When you call does your child respond to his/her name in a noisy situation when he/she can't see your face? (examples of responses include looks up, turns, answers verbally)	0	1	2	3	4
6.	When asked, does your child follow simple instructions or do a simple task in a noisy situation?	0	1	2	3	4
7.	When you are in a quiet place reading with your child, how often does he/she pay close attention to what you are saying? OR if your child is listening to stories/songs on the TV or CD when there is no other background noise how often can he/she follow what is being said?	0	1	2	3	4
8.	How often does your child initiate/ participate in conversation in a quiet situation?	0	1	2	3	4
9.	How often does your child initiate/ participate in conversation in a noisy situation?	0	1	2	3	4
10.	How often does your child understand what you say in the car/bus/train?	0	1	2	3	4
11.	How often does your child recognise peoples' voices without seeing who was talking?	0	1	2	3	4
12.	How often does your child successfully use a phone?	0	1	2	3	4
13.	How often does your child respond to sounds other than voices?	0	1	2	3	4

Please reflect on your child's listening behaviour over the past week and circle the appropriate number

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PEACH - 3 -

Please provide comments regarding any of the above items:



	F	RAW Score	% Score
QUIET	(Q's 3+4+7+8+11+12) A	(A/24) x 100	
NOISE	(Q's 5+6+9+10+13) B	(B/20) x 100	
OVERALL	(A + B) C	(C/44) x 100	

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PEACH - 4 -

Appendix B:

University of Pretoria Research Ethics Committee of the Faculty of Humanities (HUM024/0419)



Research Ethics Committee

23 August 2019

Dear Ms S Kuschke

Project Title: Researcher: Supervisor: Department: Reference number: Degree: Childhood hearing loss in the Western Cape, South Africa Ms S Kuschke Prof DCDW Swanepoel Speech Language Path and Aud 04382463 (HUM024/0419) Doctoral

I have pleasure in informing you that the above application was **approved** by the Research Ethics Committee on 25 July 2019. 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 the actual research depart significantly from the proposed research, it will be necessary to apply for a new research approval and ethical clearance.

We wish you success with the project.

Sincerely

MMUShum

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

> Fakulteit Geesteswetenskappe Lefapha la Bomotho

Research Ethics Committee Members: Prof MME Schoeman (Deputy Dean); Prof KL Harris; Mr A Bizos; Dr L Blokland; Dr K Boovens; Dr A-M de Beer; Ms A dos Santos; Dr R Fasselt; Ms KT Govinder Andrew; Dr E Johnson; Dr W Kelleher; Mr A Mohamed; Dr C Puttergill: Dr D Revburg; Dr M Soer: Prof E Taliard: Prof V Thebe: Ms B Tsebe: Ms D Mokalaoa

Appendix C:

University of Cape Town Human Research Ethics Committee (365/2019)



UNIVERSITY OF CAPE TOWN Faculty of Health Sciences Human Research Ethics Committee



Room E53-46 Old Main Building Groote Schuur Hospital Observatory 7925 Telephone (021) 406 6626 Email: shuretta.thomas@uct.ac.za Website: www.health.uct.ac.za/fis/research/humanethics/forms

02 August 2019

HREC REF: 365/2019

Prof De Wet Swanepoel c/o Ms S Kuschke Communication Sciences and Disorders Audiology F-floor, OMB

Dear Prof Swanepoel

PROJECT TITLE: ASSESSMENT AND MANAGEMENT OF CHILDHOOD HEARING LOSS IN THE WESTERN CAPE, SOUTH AFRICA (PHD CANDIDATE - MS S KUSCHKE)

Thank you for submitting your response to the Faculty of Health Sciences Human Research Ethics Committee.

It is a pleasure to inform you that the HREC has formally approved the above-mentioned study.

Approval is granted for one year until the 30 August 2020.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period.

(Forms can be found on our website: www.health.uct.ac.za/fhs/research/humanethics/forms)

Please quote the HREC REF in all your correspondence.

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

Please note that for all studies approved by the HREC, the principal investigator **must** obtain appropriate institutional approval, where necessary, before the research may occur.

The HREC acknowledge that the student, Silva Kuschke will also be involved int this study.

Yours sincerely

/W

PROFESSOR M BLOCKMAN CHAIRPERSON, FHS HUMAN RESEARCH ETHICS COMMITTEE Federal Wide Assurance Number: FWA00001637. Institutional Review Board (IRB) number: IRB00001938

Appendix D:

Red Cross War Memorial Children's Hospital Ethics Committee (RCC203)



DR AN PARBHOO Manager: Medical Services Red Cross War Memorial Children's Hospital Email: Anita.Parbhoo@westerncape.gov.za Tel: +27 21 658 5430 Fax: +27 21 658 5006/5166

21 August 2019

Ms S Kuschke Audiology

Dear Ms Kuschke,

RESEARCH: RXH: RCC 203

PROJECT TITLE: Childhood hearing loss in the Western Cape, South Africa

It is a pleasure to inform you that the hospital Research Review Committee has approved your application to conduct above-mentioned study in the Audiology Department at Red Cross War Memorial Children's Hospital.

Yours sincerely,

apape

DR AN PARBHOO MANAGER: MEDICAL SERVICES

www.westerncape.gov.za

Appendix E:

Western Cape Health Research sub-directorate (WC_201906_023)



Health Impact Assessment Health Research sub-directorate Health.Research@westerncape.gov.za tel: +27 21 483 0866: fax: +27 21 483 9895 5th Floor, Norton Rose House, 8 Riebeek Street, Cape Town, 8001

www.capegateway.gov.za)

REFERENCE: WC_201906_023 ENQUIRIES: Dr Sabela Petros

University of Pretoria

Cnr Lynnwood Road and Roper Street

Hatfield

Pretoria

0028

For attention: Ms Silva Kuschke, Prof De Wet Swanepoel

Re: Childhood hearing loss in the Western Cape, South Africa

Thank you for submitting your proposal to undertake the above-mentioned study. We are pleased to inform you that the department has granted you approval for your research.

Please contact the following person to assist you with any further enquiries in accessing the following sites:

Victoria Hospital Dr Grae	ne Dunbar 021 799 1121
---------------------------	------------------------

Kindly ensure that the following are adhered to:

- 1. Arrangements can be made with managers, providing that normal activities at requested facilities are not interrupted.
- By being granted access to provincial health facilities, you are expressing consent to provide the department with an electronic copy of the final feedback (annexure 9) within six months of completion of your project. This can be submitted to the provincial Research Co-ordinator (Health.Research@westerncape.gov.za).
- 3. In the event where the research project goes beyond the estimated completion date which was submitted, researchers are expected to complete and submit a progress report

(Annexure 8) to the provincial Research Co-ordinator

(Health.Research@westerncape.gov.za).

4. The reference number above should be quoted in all future correspondence.

Yours sincerely

Dr Melvin Moodley Director: Health Impact Assessment 13 AUG 2019

DR M MOODLEY

DIRECTOR: HEALTH IMPACT ASSESSMENT

Appendix F:

Caregiver informed consent – Study II



Faculty of Humanities Department of Speech-Language Pathology and Audiology

Study title: Decentralising paediatric hearing services through district healthcare screening, Western Cape, South Africa

Principal Investigator: Silva Kuschke

Supervisor: Professor De Wet Swanepoel

Institution: University of Pretoria, Department of Speech-Language Pathology and Audiology

DAYTIME AND AFTERHOURS TELEPHONE NUMBER(S):

Daytime number/s: (021) 658 5406 After-hours number: 079 347 6087

DATE AND TIME OF FIRST INFORMED CONSENT DISCUSSION:

Day	Month	Year

	:
Time	

Dear Parent or Legal Guardian

Dear Mr. /Mrs.

1) INTRODUCTION

We invite your child to participate in a research study. This information document will help you to decide if your child may want to participate. Before you agree that your child may take part, you should fully understand what is involved. If you have any questions that this document does not fully explain, please do not hesitate to ask the researcher.

2) THE NATURE AND PURPOSE OF THIS STUDY

The aim of this study is to determine the effect of providing hearing healthcare services at a lower level hospital to reduce the number of patients who are referred to a central tertiary hospital for their first hearing test. I will be using information about your child's hearing test at Victoria District Hospital to achieve this aim.

3) EXPLANATION OF PROCEDURES AND WHAT WILL BE EXPEXTED FROM PARTICIPANTS

When we receive a referral from your child's doctor to do a hearing test on your child for the first time, we will come and see your child at Victoria District Hospital, instead of you having to travel to Red Cross War Memorial Children's Hospital (RCWMCH). We will put a soft probe in your child's ear, and four different sounds will be played in both of his/her ears. Your child will not have to respond to the sound behaviourally. The test takes about two minutes to complete, and the test is not painful. If your child passes the test, we will discharge him/her from audiology services; and if your child does not pass the test, we will ask you to come to RCWMCH for a more in-depth hearing test.

4) POSSIBLE RISK AND DISCOMFORT INVOLVED

There are only minimal risks involved in participating in the study. The hearing screening process (inserting the soft probe into your child's ear) may cause minimal discomfort. The hearing screening will take about two minutes of your child's time.

5) POSSIBLE BENEFITS OF THIS STUDY

Although your child will not benefit directly from the study, the results of the study may enable us to make hearing screening services available at more facilities closer to patients in future.

Apart from getting the results from your child's tests, there will be no other direct benefit for you. However, the results of the study will help to promote the availability of hearing screening services at more appropriate and accessible facilities in future.

6) YOUR CHILD'S RIGHTS AS A PARTICIPANT

Your child's participation in this study is entirely voluntary. Your child can refuse to participate or stop at any time during the study without giving any reason. Your child's withdrawal will not affect his/her to hearing screening services.

7) ETHICS APPROVAL

This Protocol was submitted to the Faculty of Health Sciences Research Ethics Committee, University of Pretoria, Medical Campus, Tswelopele Building, Level 4-59, Telephone numbers 012 356 3084 / 012 356 3085 and written approval has been granted by that committee. The study has been structured in accordance with the Declaration of Helsinki (last update: October 2013), which deals with the recommendations guiding doctors in biomedical research involving humans. A copy of the Declaration may be obtained from the investigator should you wish to review it.

8) INFORMATION AND CONTACT PERSON

The contact person for the study is Silva Kuschke. If you or your child have any questions about the study, please contact her at the following telephone numbers: 021 658 5406/079 347 6087. Alternatively, you may contact my supervisor at 012 420 2816.

9) COMPENSATION

Your child will not be paid to take part in the study. There are no costs involved for your child to be part of the study.

10) CONFIDENTIALITY

All information about your child will be kept strictly confidential. Once we have analysed the information no one will be able to identify your child. Research reports and articles in scientific journals will not include any information that may identify your child.

11) CONSENT TO PARTICIPATE IN THIS STUDY

- I confirm that the person requesting my consent for my child to take part in this study has told me about the nature and process, any risks or discomforts, and the benefits of the study.
- I have also received, read and understood the above written information about the study.
- I have had adequate time to ask questions and I have no objections for my child to participate in this study.
- I am aware that the information obtained in the study, including personal details, will be anonymously processed and presented in the reporting of results.
- I understand that my child will not be penalised in any way should my child wish to discontinue with the study and that withdrawal will not affect my child's
- My child is participating willingly.
- I have received a signed copy of this informed consent agreement.

Parent/Legal Guardian's name (Please print)	Date
Parent/Legal Guardian's signature	Date
Researcher's name (Please print)	Date
Researcher's signature	Date

AFFIRMATION OF INFORMED CONSENT BY AN ILLITERATE PARTICIPANT

I, the undersigned,, have read and have explained fully to the participant, named, the participant informed consent document, which describes the nature and purpose of the study in which I have asked the child's parent/legal guardian to participate. The explanation I have given has mentioned both the possible risks and benefits of the study and the alternative treatments available for the child's illness. The participant indicated that he/she understands that he/she will be free to withdraw from the study at any time for any reason and without jeopardizing the child's standard care.

I hereby certify that the patient has agreed to participate in this study.

Parent/Legal Guardian's name (Please print)	Date
Parent/Legal Guardian's signature	Date
Investigator's Name (Please print)	Date
Investigator's Signature	Date
Name of the person who witnessed the informed consent (Please print)	Date
Signature of the Witness	Date

Appendix G:

Caregiver informed consent – Study III



Study title: Outcomes of children with sensorineural hearing loss fitted with binaural hearing aids at a paediatric public hospital in South Africa

Principal Investigator: Silva Kuschke

Supervisor: Professor De Wet Swanepoel

Institution: University of Pretoria, Department of Speech-Language Pathology and Audiology

DAYTIME AND AFTER HOURS TELEPHONE NUMBER(S):

Daytime number/s: (021) 658 5406 After-hours number: 079 347 6087

DATE AND TIME OF FIRST INFORMED CONSENT DISCUSSION:

Day	Month	Year

	:
Time	

Dear Parent/Caregiver

Dear Mr. / Mrs.

1) INTRODUCTION

You are invited to volunteer for a research study. I am doing research for a doctoral degree purpose at the University of Pretoria. This information in this document is to help you to decide if you would like to participate. Before you agree to take part in this study you should fully understand what is involved. If you have any questions, which are not fully explained in this document, do not hesitate to ask the researcher. You should not agree to take part unless you are completely happy about all the procedures involved.

2) THE NATURE AND PURPOSE OF THIS STUDY

Data of children aged 0-13 years that are fitted with two hearing aids at the Department of Audiology at Red Cross War Memorial Children's Hospital will be used in this study. For this study, validated hearing questionnaires will be reviewed to determine the hearing aid outcomes of children who wear two hearing aids.

3) EXPLANATION OF PROCEDURES AND WHAT WILL BE EXPEXTED FROM PARTICIPANTS.

This study involves answering 13 questions on a scale of 0-4 with regard to your child's behaviour while wearing his/her hearing aids. If you are unable to read, the questionnaire will be administered interview-style by an audiologist when your child comes for his/her next visit for a follow-up audiology appointment. Your answers will be scored and interpreted by an audiologist.

4) POSSIBLE RISKS AND DISCOMFORTS INVOLVED

There are no medical risks associated with the study. The only possible risk and discomfort involved is taking the time to complete the questionnaire. This should not take longer than 10 minutes.

5) POSSIBLE BENEFITS OF THIS STUDY

Although you may not benefit directly, the study results may help us to improve the way we manage children with hearing loss who wear two hearing aids.

6) COMPENSATION

You will not be paid to take part in the study. There are no costs involved for you to be part of the study.

7) YOUR RIGHTS AS A RESEARCH PARTICIPANT

Your participation in this study is entirely voluntary and you can refuse to participate or stop at any time without stating any reason. Your withdrawal will not affect your child's access to audiology services or other medical care.

8) ETHICS APPROVAL

This Protocol was submitted to the Faculty of Health Sciences Research Ethics Committee, University of Pretoria, telephone numbers 012 356 3084 / 012 356 3085 and written approval has been granted by that committee. The study has been structured in accordance with the Declaration of Helsinki (last update: October 2013), which deals with the recommendations guiding doctors in biomedical research involving human/subjects. A copy of the Declaration may be obtained from the investigator should you wish to review it.

9) INFORMATION

If I have any questions concerning this study, I should contact:

Silva Kuschke Tel : 021 658 5406 Cell: 079 347 6087

10) CONFIDENTIALITY

All information obtained during this study will be regarded as confidential. Only the researcher will be able to identify you as participant. Results will be published or presented in such a fashion that patients remain unidentifiable. The hard copies of all your records will be kept in a locked facility at Red Cross War Memorial Children's Hospital, Western Cape.

11) CONSENT TO PARTICIPATE IN THIS STUDY

- I confirm that the person requesting my consent for my child to take part in this study has told me about the nature and process, any risks or discomforts, and the benefits of the study.
- I have also received, read and understood the above written information about the study.
- I have had adequate time to ask questions and I have no objections to participate in this study.
- I am aware that the information obtained in the study, including personal details, will be anonymously processed and presented in the reporting of results.
- I understand that I will not be penalised in any way should I wish to discontinue with the study and that withdrawal will not affect my further treatments.
- I am participating willingly.
- I have received a signed copy of this informed consent agreement.

Participant's name (Please print)	Date
Participant's signature	Date
Researcher's name (Please print)	Date
Researcher's signature	Date

AFFIRMATION OF INFORMED CONSENT BY AN ILLITERATE PARTICIPANT

(if suitable)

I, the undersigned,, have read and have explained fully to the participant, named, the informed consent document, which describes the nature and purpose of the study in which I have asked the him/her to participate. The explanation I have given has mentioned both the possible risks and benefits of the study. The participant indicated that he/she understands that he/she will be free to withdraw from the study at any time for any reason and without jeopardizing his/her standard care.

I hereby certify that the patient has agreed to participate in this study.

Participant's name (Please print)

Date

Participant's signature	Date			
Investigator's Name (Please print)	Date			
Investigator's Signature	Date			
Name of the person who witnessed the informed consent (Please print)	Date			
Signature of the Witness	Date			

Appendix H:

Informed assent for children 0-6 years – Study II

ICD 1C INFORMATION AND ASSENT DOCUMENT

Study title: Decentralising paediatric hearing services through district healthcare screening, Western Cape, South Africa **Principal Investigator:** Silva Kuschke

Supervisor: Professor De Wet Swanepoel

Institution: University of Pretoria, Department of Speech-Language Pathology and Audiology

Daytime telephone number/s: (021) 658 5406 / 079 347 6087

Date and time of informed consent discussion:

1) INTRODUCTION

My name Silva Kuschke, and my job is to do research on children with hearing loss. We want to know how well children who are referred to Red Cross Children's Hospital can hear. I am going to explain this research to you and invite you to be part of this research study. You can choose whether or not you want to participate in this study. We have discussed this research study with your mom/dad/legal guardian and they know that we are also asking for your permission. If you are going to be part in this research, your mom/dad/legal guardian must also agree. But if you do not want to participate, you do not have to. You may discuss anything on this form with your mom/dad/legal guardian or friends. You can decide whether to participate or not after you have talked it over. You do not have to decide immediately. There may be some words you don't understand or things that you want me to explain to you. Please ask me to stop at any time and I will explain.

2) WHAT IS RESEARCH?

Research is what we do to find new knowledge about subjects (and people). We use research studies to help us find more information about disease or illness. Research also helps us to find better ways of treating children who are sick.

3) WHAT IS THIS RESEARCH PROJECT ALL ABOUT AND WHAT IS EXPECTED OF ME?

Children with concerns about their hearing are sent to Red Cross Children's Hospital to have their ears tested. We want to bring this hearing test to Victoria Hospital, so that it is closer for you and your mom/dad/legal guardian to travel. We want to find out how many children can be tested at Victoria Hospital, how well their ears work, and how many need to go to Red Cross Children's Hospital for further hearing tests. To do this, we need information about how well your ears work.

4) WHY HAVE I BEEN INVITED TO TAKE PART IN THIS RESEARCH PROJECT?

You were referred to Red Cross Children's Hospital by your doctor to have a hearing test.

5) WHO IS DOING THE RESEARCH?

Myself and my three colleagues, who are audiologists at Red Cross Children's Hospital, will be performing the hearing tests.

6) WHAT WILL HAPPEN TO ME IN THIS STUDY?

When we receive a referral from your doctor to do a hearing test on you for the first time, we will come and see you at Victoria Hospital, instead of you having to travel to Red Cross Children's Hospital. We will put a soft probe in your ear, and four different sounds will be played in both of your ears. You will not have to respond to the sound. The test takes about two minutes to complete, and the test is not painful. If you pass the test, we will discharge you from audiology services; and if you do not pass the test, we will ask you to come to Red Cross Children's Hospital for a more in-depth hearing test.

7) CAN ANYTHING BAD HAPPEN TO ME?

Nothing bad can happen to you because of this research study.

8) CAN ANYTHING GOOD HAPPEN TO ME?

Apart from getting the results from your hearing test, there will be no other direct benefit for you. However, the results of the study will help us to test children's hearing closer to their homes in the future.

9) ETHICS APPROVAL

This Protocol was submitted to the Faculty of Health Sciences Research Ethics Committee, University of Pretoria, Medical Campus, Tswelopele Building, Level 4-59, Telephone numbers 012 356 3084 / 012 356 3085 and written approval has been granted by that committee.

10) WILL ANYONE KNOW I AM IN THE STUDY?

Only myself and my three audiology colleagues will know that you are participating in the study.

11) WHO CAN I TALK TO ABOUT THE STUDY?

Silva Kuschke - 0216585406

12) WHAT IF I DO NOT WANT TO DO THIS?

You do not have to participate in the study, even if your mom/dad/legal guardians have signed consent that you can participate. You can also withdraw from the study at any time without getting in trouble.

13) CONSENT TO PARTICIPATE IN THIS STUDY

Do you understand this research study and are you willing to participate in it?

Yes No

Do you understand that we will put a soft probe in your ear, and four different sounds will be played in both of your ears? You will not have to respond to the sound. The test takes about two minutes to complete, and the test is not painful.

Yes No

Has the researcher answered all your questions?

Yes No

Do you understand that you can pull out of the study at any time without any one consequence?

Yes No

You don't have to give us your answer now, take your time to think about it before you decide. If you sign at the bottom it will mean that somebody has explained this paper to you, and that you would like to be in this study.

Your Name: Person obtaining consent: Parent / Guardian / Audiologist as Witness:

Appendix I:

RCWMCH caregiver consent slips

RED CROSS WAR MEMORIAL CHILDREN'S HOSPITAL

Department of Audiology Tel: (021) 658 5406

Consent to use my child's audiological data for educational and research purposes

I, ______ (caregiver) hereby give consent that my child's audiological data may be used for educational and research purposes. Confidentiality will be maintained at all times.

Patient sticker

Signature: Caregiver

Signature: Audiologist

Date:

RED CROSS WAR MEMORIAL CHILDREN'S HOSPITAL

Department of Audiology Tel: (021) 658 5406

Consent to use my child's audiological data for educational and research purposes

I, ______ (caregiver) hereby give consent that my child's audiological data may be used for educational and research purposes. Confidentiality will be maintained at all times.

Patient sticker

Signature: Caregiver

Signature: Audiologist

Date:

RED CROSS WAR MEMORIAL CHILDREN'S HOSPITAL				
Department of Audiology Tel: (021) 658 5406				
Consent to use my child's audiological data for educational and research purposes				
I, (caregiver) hereby give consent that my child's audiological data may be used for educational and research purposes. Confidentiality will be maintained at all times.				
	Patient sticker			
Signature: Caregiver	Signature: Audiologist			
Date:				

Appendix J:

Hearing screening data sheet for the district-level intervention period

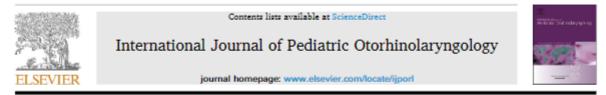
Image: series of the series	Date YY/MM/DD	File Number	Area indicated on sticker	Referral source	<1years (Age in Months)	>1years (Age in Years)	OAE Pass (R, L, BIL)	OAE Refer (R, L, BIL)	Refer to RCWMCH	Reason for referral (see tab for key)	Etiological factors related to hearing loss
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Appendix K: Proof of article publications

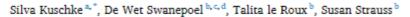
Study I –

Profile of childhood hearing loss, Western Cape Province, South Africa

International Journal of Pediatric Otorhinolaryngology 137 (2020) 110248



Profile of childhood hearing loss in the Western Cape, South Africa



Department of Audiology, Red Cross War Memorial Children's Hospital, Cape Town, South Africa
 Department of Speech-Language Pathology and Audiology, University of Pretoria, South Africa
 Ear Sciences Centre, School of Surgery, The University of Western Australia, Nedlands, Australia
 Ear Science Institute Australia, Subiaco, Australia

ARTICLEINFO

Keywordz Childhood hearing loss **Risk factors** Age of diagnosis

ABSTRACT

Objectives: To describe the nature, associated risk factors and are of diagnosis for childhood hearing loss in a South African cohort from the Western Cane Province. Methods: A retrospective review of clinical data from children under six years of age with confirmed hearing loss at Red Cross War Memorial Children's Hospital (RCWMCH) was conducted between 1 January 2019 and 31 July 2019. Data collected included demographic information, type and degree of hearing loss, documented risk factors associated with hearing loss, and age of suspicion and diagnosis of hearing loss Results: The study sample included 240 children with hearing loss, with a mean age of 42 months (21.8 SD; range 2-72). More than two thirds (68.3%) of the children presented with bilateral hearing loss. The majority presented with conductive hearing loss (64.6%), followed by sensorineural (28.7%) and mixed hearing loss (3.3%) or auditory neuropathy spectrum disorder (3.3%). More than half (51.8%) of the bilateral sensorineural hearing losses were of a profound degree. The most prominent risk factor for conductive hearing loss was otitis media, for sensorineural hearing loss it was a family history of childhood hearing loss, and for auditory neuropathy spectrum disorder it was hyperbilirubinaemia. Approximately one third of patients (27.1%) with sensorineural hearing loss did not have any associated risk factors. The mean age of diagnosis of permanent congenital or earlyonset hearing loss was 31.4 months (22.8 SD; range 2-72), with a mean delay of nine months (13.2 SD; range 0-60) between age of suspicion and diagnosis of hearing loss (n = 93). Conclusions: The large proportion of preventable hearing losses in this sample highlights the importance of maximising primary health care efforts to treat preventable causes timeously. Age of diagnosis of permanent congenital or early-onset hearing loss was severely delayed undermining prospects of positive outcomes through early intervention. Infant hearing screening services in the public health sector of South Africa should be prioritised alongside primary health care efforts to reduce preventable risks for hearing loss.

Chack for

Study II –

Decentralising paediatric hearing services through district healthcare screening, Western Cape, South Africa

African Journal of Primary Health Care & Family Medicine ISSN: (Online) 2071-2936, (Print) 2071-2928



CrossMark

Decentralising paediatric hearing services through district healthcare screening in Western Cape province, South Africa

- Page 1 of 7 Original Research -

Authors: Silva Kuschke¹ 🕲 Talita le Roux³ 🙂 Alex J. Scott³ Daniel C. d.W. Swaneppel^{3,4,1}

Affiliations: Department of Audiology, Faculty of Allied Health Communication Sciences. Red Cross War Memorial Children's Hospital, Cape Town, South Africa

²Department Speech Language Pathology and Audiology, Faculty of Humanities, University of Pretoria, Pretoria, South Africa

¹Department of Medicine, Faculty of Health Science, University of Cape Town, Cape Town, South Africa

⁴Department Ear Science Centre, School of Surgery, University of Western Australia, Nedlands, Australia

^hEar Science Institute. Subiaco, Australia

Background: Childhood hearing loss is a global epidemic most prevalent in low- and middle-income countries where hearing healthcare services are often inaccessible. Referrals for primary care services to central hospitals add to growing lists and delays the time-sensitive treatment of childhood hearing loss.

Aim: To compare a centralised tertiary model of hearing healthcare with a decentralised model through district hearing screening for children in the Western Cape province, South Africa.

Setting: A central paediatric tertiary hospital in Cape Town and a district hospital in the South Peninsula region.

Methods: A pragmatic quasi-experimental study design was used with a 7-month control period at a tertiary hospital (June 2019 to December 2019). Decentralising was measured by attendance rates, travelling distance, number of referrals to the tertiary hospital and hearing outcomes. There were 315 children in the tertiary group and 158 in the district group. Data were collected from patient records and an electronic database at the tertiary hospital.

Results: Attendance rate at the district hospital was significantly higher (p < 0.001). Travel distance to the district hospital was significantly shorter (p < 0.001). Number of referrals to the tertiary hospital decreased significantly during the intervention period (p < 0.001). Most children in both the tertiary and district groups (78.7% and 80.4%, respectively) passed initial hearing screening bilaterally.

Conclusion: Hearing screening should be conducted at the appropriate level of care to increase access, reduce patient travelling distances and associated costs and reduce the burden on tertiary-level hospitals.

Keywords: childhood hearing loss; decentralisation; hearing healthcare; low- and middleincome countries: otoacoustic emissions.

Study III –

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Outcomes of children with sensorineural hearing loss fitted with binaural hearing aids at a pediatric public hospital in South Africa

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ABSTRACT

Keywordz Objective: To describe hearing aid outcomes for children with bilateral sensorineural hearing loss (SNHL) at a Hearing aid outer pediatric public hospital in South Africa in terms of daily use and oral/aural performance. Materials and methods: Retrospective review of clinical data and caregiver reported outcomes of children aged 0-13 years with bilateral SNHL at one-month and three-months post-fitting. Oral/aural performance was Sensoringural hearing loss Data logging PEACH questions measured by the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire. Multiple linear regression was used to evaluate factors associated with hearing aid use. Thematic analysis was applied for qualitative caregiver-reported outcomes. Study sample: Sixty-eight children with confirmed bilateral SNHL who were fitted with binaural air-conduction hearing aids at Red Cross War Memorial Children's Hospital in Cape Town, South Africa, between January 2017 and December 2019. Result: Average daily hearing aid use increased significantly (p < 0.05) from one-month (5.0; 3.0 SD; range 0.3–14.0) to three-months post-fitting (5.9; 3.4 SD; range 1.1–16.8). Average PEACH scores were higher in Quiet (73.4%) than in Noise (69.6%). More than half (52.2%) of children required review based on their overall percentage PEACH scores. Higher average daily hearing aid use was significantly associated with higher overall PEACH scores (p < 0.05). Neuro-typically developing children had significantly higher hearing aid use than children with additional disabilities (p < 0.001). Qualitative caregiver feedback revealed themes pertaining to advantages and barriers to hearing aid use. Conclusion: Outcomes of children with SNHL fitted with binaural hearing aids at a pediatric public hospital in South Africa demonstrated increased average daily hearing aid use from one-month to three-months post-fitting. Aural/oral performance was typical for one in two children. Children with additional disabilities had significantly poorer hearing aid use and aural/oral performance requiring more support for this vulnerable group to realize sufficient benefit from hearing aid use.