DEPARTMENT OF SPEECH PATHOLOGY & AUDIOLOGY

THE DEVELOPMENTAL OUTCOMES OF CHILDREN PREVIOUSLY ENROLLED IN A RISK-BASED HEARING SCREENING PROGRAMME

RUMAANA BHAM

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Contents

Declaration

I, Rumaana Bham, hereby declare that this submission is my own work except as indicated in the references and acknowledgements. I am liable for the content of this study and the conclusions presented. To the best of my knowledge no part of this dissertation has been previously submitted for a degree at any other university or institution.

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Rumaana Bham Date

Scholarly Activities

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List of Abbreviations

ABR: Auditory Brainstem Response

AABR: Automated Brainstem Response

AAP: The American Association of Paediatrics

ACOG: The American Congress of Obstetricians and Gynaecologists

ADOS-G: Autism Diagnostic Observation Schedule-Generic

APGAR: Appearance, Pulse, Grimace, Activity and Respiration

ASD: Autism Spectrum Disorder

ASQ: Ages and Stages Questionnaire

BSID-II: The Bayley Scales of Infant Development – Second Edition

CMV: Cytomegalovirus

CMJAH: Charlotte Maxeke Johannesburg Academic Hospital

CPAP: Continuous Positive Airway Pressure

Denver II: The Denver Developmental Screening Test

DPOAE: Distortion Product Otoacoustic Emissions

DRC: Democratic Republic of Congo

ENT: Ear, Nose and Throat

EBT: Exchange Blood Transfusion

ECD: Early Childhood Development

ECI: Early Childhood Intervention

ECMO: Extracorporeal Membrane Oxygenation

EHDI: Early Hearing Detection and Intervention

ELBW: Extremely Low Birth Weight

ETI: Endotracheal Intubation

FARS: Friedreich Ataxia Rating Scale

FRS: Fetal Retinoid Syndrome

GMFM-88: Gross Motor Function Measure-88

HIE: Hypoxic Ischaemic Encephalopathy

HIV: Human Immunodeficiency Virus

HEI: HIV Exposed Infected

HEU: HIV Exposed Uninfected

HPCSA: The Health Professionals Council

HRR: High-Risk Registry

ICARS: International Cooperative Ataxia Rating Scale

IDEIA: Individuals with Disabilities Education Improvement Act

IPPV: Intermittent Positive Pressure Ventilation

JCIH: The Joint Committee on Infant Hearing

KMC: Kangaroo Mother Care

LBW: Low Birth Weight

MDT: Multidisciplinary

MOU: Midwife Obstetric Units

NICU: Neonatal Intensive Care Unit

NIH: National Institutes of Health

NNFU: Neonatal Follow-Up

NNJ: Neonatal Jaundice

OAE: Otoacoustic Emissions

PCR: Polymerase Chain Reaction

PDMS-II: Peabody Developmental Motor Scale

PEDS: The Parents' Evaluation of Developmental Status

PEDS: DM: The Parents' Evaluation of Developmental Status: Developmental Milestones

PHC: Primary Health Care

PTT: Phototherapy Treatment

RDS: Respiratory Distress Syndrome

RMMCH: Rahima Moosa Mother and Child Hospital

RTHB: The Road to Health Booklet

RVD: Retroviral Disease

SAM: Severe Acute Malnutrition

SARA: The Scale for the Assessment and Rating of Ataxias

SDQ: The Strengths and Difficulties Questionnaire

TEOAE: Transient Evoked Otoacoustic Emissions

TNHS: Targeted Newborn Hearing Screening

UNHS: Universal Newborn Hearing Screening

VLBW: Very Low Birth Weight

VRA: Visual Response Audiometry

WAIS- R: The Wechsler Adult Intelligence Scale Revised

WISC-III: The Wechsler Intelligence Scale for Children III

WHO: The World Health Organization

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Definition of Terms: Glossary

Acute Gastroenteritis (AGE): Inflammation of the gastrointestinal tract/ infectious diarrhoea (John Brown University, 2000).

Anaemia: A condition where your blood is not able to carry enough oxygen to meet the needs of your body (The World Health Organization, 2019).

Anotia: A birth defect where the external ear is missing completely (Centers for Disease Control and Prevention, 2018).

Apnoea: The temporary cessation of breathing, especially during sleep (The Royal Children's Hospital Melbourne, 2017).

Appearance, Pulse, Grimace, Activity, and Respiration (APGAR) score: A score based on a screening test to determine a newborn's condition at birth (The American College of Obstetricians and Gynaecologists, 2019).

Chorioamnionitis: Inflammation of the fetal membranes due to a bacterial infection (Baney-Mohammed, 2018).

Congenital Hearing Loss: A hearing loss present at birth (Alexiades & Hoffman, 2008). A congenital hearing loss represents a spectrum of underlying pathologies, which include environmental and genetic causes (Mikulec, 2009).

Continuous Positive Airway Pressure (CPAP): A type of mechanical ventilation which is usually given via a face mask (The Royal Children's Hospital Melbourne, 2017).

Cryptorchidism: Testicles that do not descend (Urology Care Foundation, 2019).

Developmental Delay: When a child does not reach their developmental milestones at the expected times. It is an ongoing major or minor delay in the process of development (University of Michigan, 2019).

Developmental Difficulty: A temporary delay in development, where a child catches up to their expected developmental level (University of Michigan, 2019).

Delayed-onset/ acquired or progressive hearing loss: A hearing loss that develops any time after birth, which may or may not be genetic in origin (Bhaya, Sperling & Madell, 2004). Stich-Hennen and Bargan (2010) views a delayed-onset hearing loss as presenting by three years of age, after presenting with normal hearing at birth.

Extracorporeal Membrane Oxygenation (ECMO): A treatment that uses a pump to circulate blood through an artificial lung back into the bloodstream (Larsson, Forsman, Hedenqvist, Östlund, Hultman, et al., 2017).

Exchange blood transfusion (EBT): A treatment to counteract serious jaundice or hyperbilirubinemia (Larsson, Forsman, Hedenqvist, et al., 2017).

Extremely Low Birth Weight (ELBW): A birth weight of 999 grams or less (Mabhandi, Ramdin & Ballot, 2019).

Febrile Seizures: A seizure associated with a high body temperature (Child Neurology Foundation, 2019).

Hyperbilirubinemia: A condition where the total serum bilirubin rises above the 95th percentile for one's age (Porter & Dennis, 2002).

Intermittent Positive Pressure Ventilation (IPPV): A type of mechanical ventilation involving endotracheal intubation (The Royal Children's Hospital Melbourne, 2017).

Low Birth Weight (LBW): A birth weight between 1500 – 2499 grams (Mabhandi, Ramdin & Ballot, 2019).

Micrognathia: Small jaw (Children's Hospital of Philadelphia, 2014).

Microtia: A congenital deformity where the pinna (external ear) is underdeveloped (Centres for Disease Control and Prevention, 2018).

Monitoring/ surveillance: Process of collecting, analysing and using information to track an individual's progress (Mtshali, 2015).

Neonatal Jaundice (NNJ): Jaundice lasts for about 10 days with a rapid rise of serum bilirubin up to 12 mg/dL in term infants. In preterm infants jaundice lasts for about two weeks, with a rapid rise of serum bilirubin up to 15 mg/dL (Bujandric & Grujic, 2016).

Otalgia: Ear pain (Ferri, 2013).

Otorrhea: Drainage exiting the ear (Ferri, 2013).

Otitis Media: Middle ear infection (Ferri, 2013).

Otitis Media with Effusion: Fluid (effusion) in the middle ear, without an infection (Ferri, 2013).

Perinatal Asphyxia: Deprivation of oxygen to a newborn infant that lasts long enough during the birth process to cause physical harm, usually to the brain (Gillam-Krakauer & Gowen Jr, 2018).

Phototherapy (PTT): Treatment method for neonatal jaundice (Larsson, Forsman, Hedenqvist, et al., 2017).

Phimosis: A condition where the foreskin is too tight to be pulled back over the head of the penis (Ashfield, Nickel, Siemens, MacNeily & Nickel, 2003).

Screening: occurs once off at certain periods and is more in-depth than monitoring and surveillance (Centers for Disease Control and Prevention, 2018).

Stenosis of the External Ear: Narrowing of the ear canal (Ferri, 2013).

Very Low Birth Weight (VLBW): A birth weight between 1000 – 1499 grams (Mabhandi, Ramdin & Ballot, 2019).

Abstract

Background: South Africa's health care system is facing numerous challenges; such as a high burden of infectious diseases, restricted resources and limited manpower within the public sector. These health care challenges have not fully allowed the successful implementation of early identification of hearing loss. This may place children at a higher risk of late identification of possible hearing developmental delays, and appropriate developmental surveillance systems are not evident. In addition, different risk factors may demonstrate variable expressivity over time, and thus, it is important to monitor the development of children who were classified as high-risk at birth.

Aims: The main aim of the current study was to determine the developmental outcomes of children who were considered high-risk at birth and previously enrolled in a risk-based newborn hearing screening programme.

Method: This study was a descriptive, cross-sectional, prospective cohort design with an integration of retrospective aspects. Data was collected through a general development screening measure (the Parents' Evaluation of Developmental Status), a hearing and communicative checklist, and a hospital file review. Sixty seven caregivers of children who were part of a risk-based newborn hearing screening study were purposefully selected. Data was analysed using descriptive and statistical analysis. Statistical analysis included the Fisher's exact test.

Results: The most frequently occurring case history factors in the current study sample included; preterm birth (100%), low birth weight classes (100%), prolonged hospital stay (95.5%), increased bilirubin levels (neonatal jaundice and hyperbilirubinemia) (86.6%), ototoxic medication (79.1%) and mechanical ventilation (26.9%). Results from the Parents' Evaluation of Developmental Status (PEDS) revealed caregivers most commonly expressed concerns relating to behaviour (25.4%), expressive language (20.9%) and social-emotional development (11.9%). A significant proportion of children in the current study did not meet their fine motor milestones (68.7%), followed by receptive language (44.8%) and expressive language milestones (40.3%). Statistically significant relationships were established between the PEDS measure and bilirubin treatment (p=0.048), and ototoxicity (p=0.008). Furthermore, the relationship between APGAR scores and gross motor milestones (p=0.002), and length of hospital stay and receptive language milestones (p=0.039) was considered statistically significant. Of the four children who did not meet their audiological milestones,

all presented with one or more of the following case history factors; preterm birth, low birth weight classes, prolonged hospital stay, neonatal jaundice mostly requiring phototherapy treatment, HIV exposed infected/ exposed uninfected, mechanical ventilation, ototoxic medication and respiratory distress syndrome.

Conclusion: Findings from the current study may be used to inform risk-based surveillance protocols at follow-up clinics. Clinical implications include suggesting case history factors which are possibly associated with delayed general and hearing development outcomes. Findings may include strengthening the collaboration between paediatricians, audiologists and other allied healthcare professionals when conducting developmental monitoring. In addition, it may aid in encouraging audiologists to monitor certain risk factors more than others in an overburdened healthcare system. Findings may also inform the feasibility and possible time periods to conduct risk-based surveillance programmes within the South African context.

Key words: Development, Risk Factors, Delayed-Onset, Hearing Loss, Risk-Based, Surveillance, Monitoring, Screening, South Africa.

Chapter 1: Background and Rationale

1.1. Background

Hearing significantly affects children's speech-language, educational, and behavioural outcomes (Lieu, 2018). The World Health Organization (WHO) (2018a) reported that in 2018, 466 million people were thought to have a disabling hearing impairment. Approximately 432 million (93.0%) are adults, and 34 million (7.0%) are children. In South Africa, the prevalence of hearing loss is estimated to be four to six in every 1000 live births in the public sector, and three in every 1000 within the private sector (Swanepoel, Störbeck & Friedand, 2009).

South Africa's health care system has been reported to face numerous challenges such as a high burden of infectious diseases, limited resources and manpower shortages within the public sector (Swanepoel, et al., 2009). These health care challenges have not fully allowed for early identification of hearing loss as an area that has been successfully implemented. This may have, and may continue to influence service provision for early detection of hearing impairment. It has been estimated that 90.0% of infants within the South African context don't receive a newborn hearing screening (Theunissen & Swanepoel, 2008; Meyer & Swanepoel, 2011). A large population of children may therefore be at risk for late identification of hearing impairment which may further delay the enrolment in an early intervention programme. An undetected or late detected hearing impairment often results in the critical period for language acquirement to be missed. Delays in speech, language and cognitive development can therefore persist, which can later affect ones scholastic performance and employment prospects (US Preventive Services Task Force, 2008; Swanepoel, 2009).

In an effort to minimize the negative consequences associated with an undetected hearing loss, The Health Professionals Council of South Africa's (HPCSA) (2018) recent guidelines advocates for universal newborn hearing screening (UNHS), and the "1-3-6" early hearing detection and intervention (EHDI) plan. This EHDI plan comprises of; hearing screening of all infants by one month of age, ensuring diagnostic evaluation by three months of age, and enrolling infants with a hearing loss in early intervention services by six months of age. These plans are however not always successfully implemented within the South African context. Kanji (2016a) suggests targeted newborn hearing screening (TNHS)/ risk-based hearing screening, which utilizes a high-risk registry (HRR) to be implemented as an interim solution towards UNHS in South Africa. The Joint Committee on Infant Hearing (JCIH) (2007) has also proposed risk-based hearing screening as the primary method to identify children who may be at risk of developing a delayed-onset hearing loss and require monitoring of their hearing. Risk-based monitoring/ targeted surveillance is imperative as passing a hearing screening at birth may not be sufficient to preclude the possibility of paediatric hearing loss, particularly in certain high-risk cases (Mann, Cuttler & Campbell, 2001).

There is however conflicting literature with regard to the relevance and applicability of the risk factors on the current HPCSA (2018) HRR. The Health Professionals Council of South Africa (HPCSA) (2018) HRR for congenital and delayed-onset hearing loss is similar to the JCIH (2007) guidelines, as there is a paucity of South African research establishing contextually relevant risk-factors for hearing loss. Such studies are however needed as the profile and hearing outcomes of neonates differs per country, as risk factors may be influenced by the infrastructure, community and diseases present in different contexts during different time periods (Kanji & Khoza-Shangase, 2012). In addition, risk factors manifest differently over time. Key differences have been noted between countries, and especially in

terms of toxoplasmosis and extracorporeal membrane oxygenation (ECMO) (Beswick, Driscoll & Kei, 2012; Vos, Senterre, Lagasse, Group & Leveque, 2015).

Risk factors may also demonstrate variable expressivity in terms of age of onset. The Joint Committee on Infant Hearing (JCIH) (2007) therefore recommends that infants who present with a risk factor for delayed-onset hearing loss should have at least one audiologic evaluation by 24 – 30 months of age. Evaluation should occur at any point where caregivers or professionals express concern regarding communication development. The Health Professionals Council of South Africa (HPCSA) (2018) has also identified risk factors for delayed-onset hearing loss which is similar to those of the JCIH (2007), however, with the addition of contextually relevant factors. The Health Professionals Council of South Africa (HPCSA) (2018) has advocated for caregivers to be informed of these risk factors at antenatal clinics and for these risk factors to be recorded on the Road to Health Booklets (RTHB), given to caregivers when their children are born. Caregivers should return if concern arises.

The Health Professionals Council of South Africa (HPCSA) (2018) has also identified a smaller set of five risk factors for hearing loss which should be continuously surveyed, a set of risk-factors associated with progressive-onset hearing loss and the age range they occur in. It is recommended that children who present with these risk factors should be referred for a behavioural audiological assessment at eight to 10 months of age, and surveillance protocols should be developed according to the age of onset of the progressive hearing loss. Studies establishing the relevance of these risk-factors are however internationally based, which re-emphasises the importance of constant and longitudinal research on children who have undergone newborn hearing screening but may be at risk for hearing impairment as this will inform relevant and sensitive audiological surveillance protocols within our context.

1.2. Rationale

While it is important to address and monitor the hearing development of the high-risk population, the majority of the HPCSA (2018) risk factors for hearing loss have also been associated with general developmental delays (fine motor, gross motor, receptive language expressive language, social-emotional and self-help delays), which calls for co-ordinated general developmental monitoring systems as well. Maternal and/ or infant Human Immunodeficiency Virus (HIV) for example presents on the HPCSA (2018) HRR and has a large body of evidence associating it with adverse developmental milestones in numerous domains such as fine and gross motor, speech and language, cognitive and behavioural domains (Sherr, Croome, Castaneda, Bradshaw & Romero, 2014; Laughton, Cornell, Boivin, & Van Rie, 2013; Mwaba, Ngoma, Kusanthan & Menon, 2015; Sherr, Hensels, Tomlinson, Skeen & Macedo, 2018; Baillieu & Potterton, 2008; Lowick, Sawry & Meyers, 2012).

The South African context is complex, with numerous factors such as poverty, malnutrition and high rates of infection and poor caregiver education (Grantham-McGregor, et al., 2007) which places children at a higher risk of presenting with adverse developmental outcomes. There has recently been two studies which have analysed the long-term developmental outcomes of high-risk neonates within the South African context. Firstly, a study by Ramdin, Ballot, Rakotsoane, Madzudzo, Brown, et al., (2018) studied the developmental outcomes of late preterm infants at nine to 12 months, and then again at 15 to 18 months of age in Johannesburg. Secondly, a study conducted by Ballot, Ramdin, Rakotsoane, Agaba, Chirwa, et al., (2017) analysed the long-term developmental outcomes of very low birth weight (VLBW) infants in Johannesburg. The authors from both studies suggest that both groups of children are considered a vulnerable population who are at an increased risk for developmental difficulties, and therefore long-term developmental monitoring is needed for this population. There is limited research available on improving

early detection of developmental difficulties or delays through developmental surveillance or screening tools, as well as limited research on the implementation of such tools in low and middle income countries (Dekker, 2011).

In an effort to monitor the general development of children in South Africa, there is currently a parent administered screening tool being utilized which is included as part of The Road to Health Booklet (RTHB) (Naidoo, Avenant & Goga, 2018). This tool is used to monitor child health, growth and development (Naidoo, Avenant & Goga, 2018). Evidence suggests however that this screening tool failed to identify more than half of infants at risk for developmental delay or disorders, and no clear referral process has been specified by this tool (Van der Linde, Glascoe, Louw & Vinick, 2015). There is therefore a need for revision or replacement of this tool.

Most of the research regarding the long-term developmental and audiological monitoring is internationally based, hence a dearth of published research in South Africa exists. There is also a lack of follow-up studies on children who are considered high-risk at birth, in terms of their long term developmental and audiological outcomes following newborn hearing screening. Such information is however crucial to ensure appropriate monitoring and intervention during the early childhood development (ECD) phase where children can maximize their developmental outcomes. It is this period which can assist in improving delays and therefore improving the overall quality of life of these children. In order to ensure early identification of general and audiological developmental delays for children considered high-risk at birth, structured developmental monitoring and audiological surveillance systems are necessary. These systems must utilize appropriate measures and time frames for early detection and intervention of general developmental and hearing delays.

Chapter 2: Overview and Literature Review

This chapter provides a contextual background of this study, with a focus on early childhood intervention (ECI) and early hearing detection and intervention (EHDI) within the South African context. The principals of the *Developmental System Model (2005)* which addresses the components of the ECI system will be discussed. In addition, this chapter introduces relevant risk factors for hearing impairment and developmental delay by providing outcomes from relevant studies. These findings highlight the importance of audiological surveillance and developmental screening. In conclusion, this chapter discusses methods and tools used to monitor those with hearing impairment and/ or developmental delay.

2.1. Early Childhood Intervention (ECI)

Early childhood development (ECD) refers to very young children (ages zero to eight years) (WHO, 2017). This period is considered critical because the brain develops most rapidly and has a high capacity for change. The foundation for health and wellbeing for one's life is laid during this early development period (Ali, 2013).

Early Childhood Intervention (ECI) is therefore critical to maximize children's developmental outcomes. The health sector has an important role to play in ECI, as it has the potential and opportunity to reach many families and children. Services include supporting children with developmental delays or disabilities. This is ensured by providing therapy and/ or education, counselling, assistance and support to access services, for example, schooling and childcare (Early Childhood Intervention Australia, 2017). Early Childhood Intervention (ECI) as a result encourages child development, well-being and family involvement.

In an effort to address all the components and complexities of ECI, Guralnick (2005) proposed a *Developmental System Model.* This model describes the relevant structural components of the ECI system. Guralnick (2005) explains that when a child is born with a

biological risk, for example, prematurity, a stress is placed on the family patterns of interaction. This model aims to minimize the stressors from creating negative family patterns of interaction to helping maintain the family's strengths instead. Another component of this model includes advocating for effective and efficient community-based screening programmes and referral components. Hence, children who are considered at risk should be enrolled in a risk-based monitoring/ surveillance programme. Points of access must also be established and available for children to access ECI.

The differences between monitoring, surveillance and screening programmes is apparent, with screening being the most in-depth. Nevertheless, all three methods, monitoring, surveillance and screening, are therefore useful when used in combination and also form relevant structural components of the ECI system according to the *Developmental System Model*.

Samuels, et al., (2012) study suggests that the *Developmental System Model* has great potential for ECI service delivery in South Africa. There are however gaps within our current ECI system. Samuels, et al., (2012) further explains that developmental screening programmes are only available at certain health institutions, for example, *Chris Hani Baragwanath Academic Hospital,* and Primary Health Care (PHC) clinics. Developmental screening is in addition not made necessary by law, and there are no systematic developmental monitoring and surveillance systems in both the public and private health care sectors (Samuels, et al., 2012). Furthermore, access to specialist services are also a challenge, as it may be difficult for families in rural parts of the country to access these services (Samuels, et al., 2012). Reasons for these gaps may include a lack of leadership, the absence of a national database on children at risk for developmental delay, the absence of clear protocols for developmental screening, the low priority status of childhood development, and the variations in family's access to health services (Samuels, et al., 2012). As a result,

Samuels, et al., (2012) study has highlighted areas for reformation, the need for political recognition and the need for investment in young children in South Africa.

This study aimed to explore risk-based surveillance through monitoring of developmental milestones in children who were previously considered high-risk at birth and enrolled in a risk-based newborn hearing screening programme. This study consequently focused on children who were born with a biological risk. Certain aspects of the current study fall within Guralnick (2005) *Developmental System Model*, and address certain gaps noted in Samuels, et al., (2012) study. Inevitably aspects such as family needs, referral components, risk-based monitoring, surveillance, and screening programmes are addressed and utilized. However, whilst the *Developmental Systems Model* addresses identifying developmental delays, the current study addressed developmental difficulties, including delayed-onset hearing loss. Addressing the hearing development is necessary, as hearing significantly affects children's speech-language, educational, and behavioural outcomes (Borton, Mauze & Lieu, 2010). In addition, passing a hearing screening at birth may not be sufficient information to preclude the possibility of paediatric hearing impairment, particularly in certain high-risk cases (Mann, et al., 2001). This emphasises the importance of risk-based monitoring/ targeted surveillance for possible delayed-onset of hearing loss, which forms part of Early Hearing Detection and Intervention (EHDI).

2.2. Early Hearing Detection and Intervention (EHDI)

Early Hearing Detection and Intervention (EHDI) is a fundamental step to reduce the negative consequences associated with the psychosocial, scholastic and social-emotional development in children with hearing impairment. The Joint Committee on Infant Hearing (JCIH) (2007) has encouraged EHDI for infants with hearing impairment. Early hearing detection and intervention (EHDI) allows for as little time as possible to lapse between the

onset of hearing impairment, its detection and subsequent management or intervention (Olusanya, et al., 2007). This will minimize auditory deprivation while maximally stimulating auditory development during the peak period for neural growth (Olusanya, et al., 2007). Since early auditory stimulation is the foundation for optimal speech and language development in the first year of life, the main goal of EHDI is to maximize linguistic and communicative competence, and literacy development for children who are deaf or hard of hearing (JCIH, 2007; HPCSA, 2018). The JCIH (2007) position statement has provided important principles and protocol suggestions which should form the foundation for effective EHDI. In South Africa, the Health Professionals Council (HPCSA) has noticed the importance of EHDI, and as a result has developed an EHDI position statement (HPCSA, 2018).

The Health Professionals Council of South Africa (HPCSA) (2018) EHDI position statement principles state that firstly, a physiologic measure should be used to conduct hearing screening in the NICU, high care ward or Kangaroo Mother Care (KMC) ward, well-baby nurseries, during immunization visits at PHC clinics or through postnatal follow-up visits at Midwife Obstetric Units (MOUs). This initial hearing screen should occur by one month of age for infants screened within hospital screening programmes, and by six weeks for clinic-based programmes. Secondly, all infants must be given access to an effective referral system if they don't *pass* the initial or any subsequent screen. This is to be able to confirm the presence of a hearing loss by three months of age for those assessed within hospital-based screening programmes. This is also to confirm the presence of a hearing loss for infants enrolled via screening programmes linked to immunisation visits by no later than four months of age. Thirdly, all infants with confirmed permanent hearing loss within hospital-based screening programmes receive intervention services before six months of age and before eight months of age for those infants identified through screening programmes

linked to immunisation visits. The JCIH (2007) and the HPCSA (2018) principles are therefore largely based on the "1-3-6" EHDI plan which consists of three goals; screening all infants by one month of age, ensuring diagnostic evaluation by three months of age and enrolling infants with a hearing loss in early intervention services by six months of age.

Principles further include, ensuring all infants who *pass* the initial hearing screening, but present with risk factors for progressive, late-onset, bilateral hearing loss, auditory disorders and/ or speech-language delay receive ongoing monitoring by caregivers. Caregivers must therefore be informed of the risks, and communication developmental milestones. Infant and family rights should also be ensured through ethical practice in terms of informed choice and consent, and protection of hearing screening evaluation and intervention results. Information regarding screening and possible follow-up appointments or management must be managed by an integrated information system.

Both the JCIH (2007) and HPCSA (2018) position statements have recommended universal newborn hearing screening (UNHS) for effective early detection of hearing impairment. However, UNHS is currently not being successfully implemented in South Africa's health care context, as only 7.5% of public hospitals provide some form of newborn hearing screening, and less than 1% provide UNHS (Meyer & Swanepoel, 2011; South African National Treasury, 2010). Meyer and Swanepoel (2011) further suggest that within the private healthcare sector, only 53% of obstetric units are offered newborn hearing screening, and only 14% were a part of UNHS. As a result, more than 90% of infants within the South African context may not undergo newborn hearing screening and are at-risk for possible late identification of hearing loss (Theunissen & Swanepoel, 2008; Meyer & Swanepoel, 2011). This consequently affects the "1-3-6" EHDI plan, and the subsequent timing of management. In addition, Khoza-Shangase, Kanji, Petrocchi-Bartal and Farr (2017) evaluated the challenges encountered during implementation of UNHS at a secondary level

public hospital in South Africa, through a combination of questionnaires, and face-to-face semi-structured interviews. Results revealed that the implementation of a UNHS programme was unattainable due to four fundamental challenges. Firstly, there were a limited number of audiologists available to screen the neonates daily (Khoza-Shangase, Kanji, Petrocchi-Bartal & Farr, 2017). Secondly, malfunctioning of screening equipment, and therefore it was suggested that more than one set of screening equipment would be required within a secondary level public hospital setting (Khoza-Shangase, Kanji, Petrocchi-Bartal & Farr, 2017). In addition, the influence of ambient noise, and early discharge of neonates from the ward (Khoza-Shangase, Kanji, Petrocchi-Bartal & Farr, 2017). These challenges raise many questions as to whether other protocols should be implemented e.g. hearing screening performed by nursing staff, and implementing more behavioural screening measures for newborns. Kanji (2016a) suggests that targeted newborn hearing screening (TNHS)/ risk-based hearing screening should be implemented as an interim solution towards UNHS in South Africa.

A risk-based hearing screening programme involves screening of newborns with known risk factors for hearing impairment (Kanji, 2016a). The current study is a follow-up study on infants who had been enrolled in a risk-based newborn hearing screening study (Kanji, 2016b).

2.3. Risk Factors Associated with Hearing Impairment

The characteristics that contribute to the presence of hearing loss are termed as risk factors (Dumanch, Holte, O'Hollearn, Walker, Clark, et al., 2017). The Joint Committee on Infant Hearing (JCIH) has revised their position statement from 1982 (JCIH, 1982), to compile their most recent position statement in 2007. The Joint Committee on Infant Hearing (JCIH) (2007) and HPCSA (2018) has identified risk factors associated with congenital

hearing loss, delayed-onset hearing loss, progressive hearing loss and those requiring continued audiological surveillance.

The JCIH (2007) high-risk registry (HRR) has identified 11 risk factors related to congenital hearing loss. These include;

- Caregiver concern regarding hearing, speech, language, or developmental delay
- Family history of permanent childhood hearing loss
- Findings associated with a syndrome known to include sensorineural or permanent conductive hearing loss
- Craniofacial anomalies, including those involving the pinna, ear canal, ear tags, ear pits, and temporal bone anomalies
- Postnatal infections associated with sensorineural hearing loss
- Head trauma, especially basal skull/ temporal bone fracture requiring hospitalization
- Neurodegenerative disorders such as Hunter syndrome, or sensory motor neuropathies, such as Friedreich ataxia and Charcot-Marie-Tooth syndrome
- Chemotherapy
- Neonatal intensive care admission for more than five days
- Neonatal indicators such as Extracorporeal Membrane Oxygenation (ECMO), hyperbilirubinemia requiring exchange blood transfusion, mechanical/ assisted ventilation and exposure to ototoxic medication
- In-utero infections such as CMV, herpes, rubella, syphilis and toxoplasmosis

The Health Professionals Counsel of South Africa (HPCSA) (2018) has adopted the same HRR as the JCIH (2007), with the addition of maternal and/or infant Human Immunodeficiency Virus (HIV), recurrent or persistent otitis media with effusion for at least

three months, and infants who present with a unilateral *refer* result during a screening protocol targeting bilateral hearing loss.

Nine of the JCIH (2007) risk factors included in the HRR are primarily focused on delayed-onset hearing loss, and are therefore used to identify and monitor infants who *pass* their initial hearing screen but are at risk for developing hearing loss later in childhood. These include;

- Parental or caregiver concern regarding hearing, speech, language or developmental delay
- Family history of permanent childhood hearing loss
- ECMO
- CMV
- Postnatal infections associated with sensorineural hearing loss, including bacterial meningitis
- Syndromes associated with progressive hearing loss, for example, neurofibromatosis, osteopetrosis and Usher's syndrome
- Neurodegenerative disorders, for example, Hunter Syndrome, or sensorimotor neuropathies such as Friedreich's ataxia and Charcot-Marie-Tooth syndrome
- Head trauma
- Chemotherapy

The Health Professionals Council of South Africa (HPCSA) (2018) has also considered the above mentioned nine risk factors for delayed-onset hearing loss. With the addition of maternal and/or infant HIV infection, recurrent or persistent otitis media with effusion for at least three months, and infants with a unilateral *refer* result during a screening protocol targeting bilateral hearing loss.

The HPCSA (2018) has in addition considered recent international studies (Wood, Davis & Stutton, 2013; Molloy, Wake, Poulakis, Barker & Goldfeld, 2014) and has further recommended continued audiological surveillance for young children who *pass* the newborn hearing screening but have risk factors which include;

- Down's syndrome
- Other syndromes known to be associated with a hearing loss, for example, Treacher Collins syndrome, Pendred syndrome and CHARGE syndrome
- Craniofacial anomalies
- Congenital infections, for example, toxoplasmosis, rubella and CMV
- If following a protocol that utilizes an automated brainstem response (AABR), to ensure surveillance of those children in the NICU who *refer* in both ears at otoacoustic emission (OAE) and *pass* in both ears at AABR

The HPCSA (2018) has also considered risk factors associated with progressive-onset hearing losses, and have included these for continuous surveillance. Table 2.1 below displays the risk factors associated with progressive-onset hearing losses and the age of onset.

Table 2.1

The Risk Factors Associated with Progressive Hearing Losses and the Ages of Onset according to the HPCSA (2018)

Age Range	Risk Factor
Birth to five years	Autosomal recessive disorders
	X-linked disorders
	Jervell and Lange-Nielsen syndrome
	Perinatal events
	Congenital CMV
	Congenital rubella
	Mucopolysaccharidoses.
Five to 10 years	Autosomal dominant disorders
	Osteogenesis imperfecta
	Alport syndrome
	Alstrom syndrome
	Marshall syndrome
	Noonan syndrome
10 to 20 years	Otosclerosis
	Usher syndrome Type 3
	Mitochondrial
	Down's syndrome
	Turner syndrome
	Norrie syndrome
	Congenital syphilis
	Autoimmune
	Noise
Any age	Bacterial meningitis
	Ototoxic medication
	Widened vestibular aqueducts
	Tumours
	Trauma

The risk factors discussed have been associated with hearing, with little focus on development as a whole, which the current study aimed to explore. Creating awareness of the risk factors associated with delayed-onset hearing loss and other general developmental delays can create better communication and referrals among health care professionals, and ECI within the South African context.

2.4. Risk Factors Associated with General Developmental Delays

2.4.1 Known risk factors associated with hearing impairment and general developmental delays.

Thirty to 40% of children with confirmed hearing loss present with other developmental delays (Karchmer & Allen, 1999). General developmental delays for the purpose of the current study refers to fine motor, gross motor, receptive language, expressive language, social-emotional, and self-help milestone concerns. As children transition into the school context, there is an increase in demand within all general development domains. Table 2.2 below displays risk factors associated with delayed-onset hearing losses according to the HPCSA (2018), and research associating it with general developmental delays.

Table 2.2

Risk Factors Associated with Congenital and/ or Delayed-Onset Hearing Losses, and General Developmental Delays

Risk-Factor	Author	Study Place	Sample Size	Assessment Measure	Domains Delays were Detected
Parental or caregiver concern regarding hearing, speech, language, and/ or developmental delay	(Glascoe, 2003).	The United States of America	472 parents and their children (21 months to 8 years old)	The Parents' Evaluation of Developmental Status (PEDS)	Behavioural Emotional
Extracorporeal membrane oxygenation (ECMO)	(Hanekamp, Mazer, van der Cammen-van, et al., 2006).	Netherlands	98 children (5 years old)	Assessments performed by paediatricians/ neonatologists, physiotherapists, psychologists and speech therapists	Neuromotor Neuropsychological Speech and language
Rubella	(Toizumi, Nguyen, Motomura, Nguyen, Pham, et al., 2017).	Vietnam	41 children (median age 23.0 months)	Ages and Stages Questionnaire (ASQ) The Denver Developmental Screening Test (Denver II)	Personal-social Fine motor Gross motor Language Problem solving

				Modified Checklist for Autism in Toddlers	Sensory integration
Neurofibromatosis	(Soucy, Gao, Gutmann & Dunn, 2012).	The United States of America	66 children (less than 8 years of age)	The Parents' Evaluation of Developmental Status (PEDS)	Fine motor Gross motor Premath delays
Usher syndrome	(Dammeyer, 2012).	Denmark	26 children (0 – 17 years of age)	Autism Diagnostic Observation Schedule-Generic (ADOS-G) The Strengths and Difficulties Questionnaire (SDQ)	Mental Behavioural Visual Language
Friedreich's ataxia	(Vavla, Arrigoni, Nordio, De Luca, Pizzighello, et al., 2018).	Italy	21 patients (patients were divided into two age groups; 12 – 16 years and 16 – 50 years)	The Scale for the Assessment and Rating of Ataxias (SARA) International Cooperative Ataxia Rating Scale (ICARS) The Wechsler Intelligence Scale for Children III (WISC-III) or The	Motor Sensory Global development

				Wechsler Adult Intelligence Scale Revised (WAIS- R) The neurological section of the Friedreich Ataxia Rating Scale (FARS)	
Down's syndrome	(Malak, Kostiukow, A., Krawczyk-Wasielewska, Mojs & Samborski, 2015).	Poland	79 children (average age 6 years 4 months)	Gross Motor Function Measure-88 (GMFM-88)	Motor
Head trauma	(Keenan, Hooper, Wetherington, Nocera & Runyan, 2007).	The United States of America	112 children (2 to 3 years of age)	The Mullen Scales of Early Learning	Cognitive
Chemotherapy	(Bornstein, Scrimin, Putnick, Capello, et al.,	Italy	61 children (aged four years and	The Bayley Scales of Infant Development—	Cognitive Language

	2012).		below)	Second Edition (BSID-II)	Motor
Infant Human Immunodeficiency Virus (HIV) infection	(Jelsma, Davids & Ferguson, 2011)	South Africa	44 children (aged 35 –74 months)	Peabody Developmental Motor Scale (PDMS-II)	Gross motor Fine motor
	(Laughton,Cornell, Boivin & Van Rie, 2013)	South Africa The United States of America Netherlands Canada Uganda Thailand	All children spanning to adolescents	File review	Cognitive Visual-Spatial
	(Baillieu & Potterton, 2008)	South Africa	40 children (aged 18 to 30 months)	Bayley Scales of Infant Development II (BSID II)	Cognitive Global language Gross motor
	(Lowick, Sawry & Meyers, 2012)	South Africa	30 children (aged 5 – 6 years)	Griffiths Mental Development Scales – Extended Revised Version	Cognitive

				(GMDS-ER)	
Craniofacial anomalies	(Speltz, Endriga, Hill, Maris, Jones, et al., 2000).	The United States of America	29 children (aged either 3, 12 or 24 months)	Bayley Scales of Infant Development (BSID)	Cognitive Expressive language Psychomotor

Of the above mentioned risk factors, HIV is one of the most contextually relevant risk factors in the present study. South Africa has the largest HIV epidemic in the world, and the total number of persons living with HIV has increased from an estimated 4,64 million in 2002 to 7,97 million by 2019 (Statistics South Africa, 2019). The HIV virus affects the central nervous system as a result of the actual virus and/ or opportunistic infection caused by immunodeficiency (Ballieu & Pottertaon, 2008).

The literature has associated cognitive delays for all children affected with HIV regardless of whether they have been on treatment or not. When analysing the above HIV studies in further detail, Baillieu and Potterton (2008) investigated the outcomes of HIV positive children who did not receive anti-retroviral (ART) treatment. Cognition was noted to be significantly affected, which is a result of structural changes to the brain due to the effect on the central nervous system (Baillieu & Potterton, 2008). However, cognitive delays were also noted on children who were on ART for more than one year (Lowick, Sawry & Meyers, 2012). Despite successful ART, 90% of the infected children had significant delay, which may be a result of the direct neurotoxic effects on the central nervous system resulting in structural damage (Lowick, Sawry & Meyers, 2012). Similarly, literature reviewed by Laughton Cornell, Boivin & Van Rie (2013) revealed that these children did not perform well on cognition tests, processing speed, visio-spatial tasks and were at a higher risk of psychiatric and mental health problems.

These studies further revealed that children who did not receive ART treatment and who were perinatally infected were more likely to present with delays in other developmental domains as well. Baillieu and Potterton's (2008) study revealed children who did not receive ART treatment to present with language delays, especially with regards to expressive language as a result of the central nervous system difficulties. Motor delays were also found

to be significantly delayed, especially relating to gross motor skills as a result of poor strength (Baillieu & Potterton, 2008). Children who were perinatally affected with detectable viral load and ART initiation less than six months of age had an increased risk of language impairment (Laughton Cornell, Boivin & Van Rie, 2013). Other risk factors for language impairment combined with cognitive or hearing impairment were race and caregiver's education status (Laughton Cornell, Boivin & Van Rie 2013). This at-risk population's development can therefore be affected by numerous factors, with their development being heterogeneous. This highlights the need for audiologists to be aware of the possible influence of risk factors for hearing impairment on other domains of general development.

2.4.2 Risk factors associated with general developmental delays. There are a number of risk factors which the HPCSA (2018) have not associated with hearing impairment, but are associated with general developmental delays. Some of these risk factors include; prematurity, low birth weight, sepsis, meningitis, and neonatal jaundice. Risk factors also include poverty and maternal education which will be discussed later in the chapter.

In terms of prematurity, Lecuona, Van Jaarsveld, van Jaarsveld and Van Heerden (2017) reports that premature infants are at risk of sensory processing difficulties and developmental delays because of an immature central nervous system and possible episodes of medical instability, discomfort, pain and stress during the first weeks or months after birth. Lecuona, et al., (2017) analysed the development of 24 infants in South Africa who were premature, presented with a low birth weight of between 750 – 1499 g, a gestational age of 26 – 36 weeks, corrected 4 – 10 months, and did not receive any previous therapy or presented with any other conditions. The developmental status was determined with the *Bayley Scales of Infant and Toddler Development III*, the *Test of Sensory Functions in Infants and the Infant/ Toddler Sensory Profile*. All infants experienced difficulties in terms of low

registration, sensory sensitivity and low thresholds, as well as sensory avoiding behaviour. Those who received intervention improved in all areas of development, with significant improvement noted in cognitive, receptive and expressive communication, and fine motor, gross motor and social-emotional development.

Similar results were obtained by Ferreira, Mello, and Silva (2013) who evaluated 194 infants who presented with VLBW and neonatal sepsis and a 12 month corrected age. *The Bayley Scale of Infant Development II* revealed neonatal sepsis to be an independent risk factor for neuromotor delay, but not cognitive development. In addition, Dangor, Lala, Cutland, Koen, Jose, et al., (2015) evaluated the neurological sequelae in infants exposed to Group B Streptococcus (GBS) which is a leading cause of sepsis and meningitis in South Africa. Infants with GBS were 21.48 times more likely to have neurological sequelae (Dangor, Lala, Cutland, et al., 2015). The results of these studies highlight that although not all high-risk factors are significantly associated with a hearing loss, they may be associated with delays in other developmental domains.

When analysing high-risk neonates who present with increased levels of bilirubin, there is conflicting evidence. Wilar, Masloman, Lestari and Tjeng (2010) found a statistically significant relationship between hyperbilirubinemia in term infants and developmental delay. However, numerous other studies (Vandborg Hansen, Greisen, Jepsen, Ebbesen, 2012; Heimler & Sasidharan, 2010; Fallah Karimi, Bafrooee, 2013) did not find a relationship between increased levels of bilirubin and developmental delay. This can be related to the level and duration of bilirubin that influences the severity of the developmental delays (Wusthoff and Loe,2015).

There is compelling evidence to suggest that the HPCSA (2018) risk-factors for congenital and/ or delayed-onset hearing loss, are also risk factors for general developmental

delays. It is important for audiologists to consider the child as a whole, and be aware of risk factors that may not necessarily be associated with audiological delays, but are associated with other general developmental delays for the high-risk population. This will provide a more holistic picture of the child, which can positively affect intervention and long term outcomes.

2.5. Audiological Risk-Based Monitoring/ Targeted Surveillance

Different risk factors for hearing loss may demonstrate variable expressivity over time. For example, certain clinical indicators of syndromic sensorineural hearing loss are not apparent at birth and have a variable delayed age of onset (Kochhar, Hildebrand & Smith, 2007). Secondly, in certain cases such as cerebral palsy there may be a delayed-onset of hearing impairment as part of the aging process or because of co-morbidities such as acquired neurological conditions (Haak, Lensk, Hidecker, Li & Paneth, 2009). Thirdly, there is a no direct relationship between specific risk factors such as neonatal bilirubin levels and the average hearing thresholds (Panahi, et al., 2013). Therefore, these children could *pass* their initial hearing screening and *refer* later, which emphasizes the need for monitoring children with various amounts of bilirubin levels. In addition, as seen in Lü, et al., (2011) study, children who previously *passed* their newborn hearing screening can present with delayed-onset hearing loss. The Joint Committee on Infant Hearing (JCIH) (2007) has therefore proposed risk-based monitoring/ targeted surveillance as the primary method to monitor hearing in children who may be at risk of developing a postnatal hearing loss.

However, there are conflicting findings within literature as to whether the JCIH (2007) risk-based surveillance factors are appropriate. A study by Vos, et al., (2015) determined the level of evidence and strength of recommendation for monitoring each risk factor. Results revealed the following evidence for levels of association between a given risk

factor and delayed-onset hearing loss. Strong levels of association were noted for CMV, toxoplasmosis, syphilis, or rubella. Moderate levels of association included a family history of hearing loss, hyperbilirubinemia, meningitis or ECMO therapy. Weak levels of association included low Appearance, Pulse, Grimace, Activity, and Respiration (APGAR) scores, and very low levels included congenital herpes, low birth weight (LBW), NICU stay, assisted/ mechanical ventilation or ototoxic drugs. Whilst some of these results are in agreement with Beswick, Driscoll, and Kei (2012), differences also exist.

Beswick, et al., (2012) results revealed strong levels of association for congenital CMV or ECMO, weak levels of association for genetic mutations, respiratory distress syndrome (RDS), or family history of hearing loss, and no association for LBW or congenital toxoplasmosis. The differences in findings between these two studies are apparent. The studies discussed highlight and reinforce that the profile and hearing outcomes of neonates differs per country. Risk factors may be influenced by the infrastructure, community and diseases present in different contexts during different time periods (Kanji & Khoza-Shangase, 2012). Since risk factors may manifest differently, further large-scale research to examine the association between risk factors and postnatal hearing status is needed. Furthermore, risk-based hearing monitoring/ targeted surveillance is also important.

The Joint Committee on Infant Hearing (JCIH) (2007) recommends that all infants with a risk factor for hearing loss should receive at least one audiological assessment by 24 to 30 months of age, with more frequent and earlier monitoring of those with certain factors, for example, craniofacial anomalies, and a family history of childhood hearing loss. In addition to risk factor monitoring, it is recommended that the family doctor should monitor all children for developmental milestones and auditory skills, and should consider caregiver distresses about the child's speech and language abilities. The current study therefore

followed this recommendation and reviewed paediatricians' notes and considered caregivers' concerns.

In order for children to be effectively monitored by their caregiver, trained personnel to advise and empower caregivers to cautiously monitor their child's hearing ability and communicative development, against the milestones for normal speech and language development is needed. Relying on caregivers' concern has however proven to not be an effective method of surveillance (Wood & Sutton, 2013; Mehl & Thomson, 2002). Wood and Stutton (2012) findings suggest that caregivers often present with poor risk factor knowledge associated with hearing impairment. In addition, targeted surveillance is not viewed as significant amongst caregivers. These values are related to a sense of security caregivers feel from their child passing their newborn hearing screening test. Furthermore, of the families that do attend targeted surveillance appointments 0.35% present with permanent childhood hearing impairment (Wood & Stutton, 2012). The remaining children present with non-permanent hearing impairment or a mild permanent childhood hearing impairment which did not meet the criteria for the screen target condition (Wood & Stutton, 2012). Evidence suggests that continuous re-evaluation of targeted surveillance programmes are needed in order to not overburden hospital resources and caregivers with numerous, and in some cases, unnecessary appointments (Wood & Stutton, 2012).

In Queensland, Australia, Beswick, Driscoll, Kei and Glennon (2012a) report that most children referred for risk-based monitoring/ targeted surveillance are seen at audiology for a one-off appointment at nine to 12 months old, with the exception of children who have family history or congenital infection as a risk factor. Children with family history as a risk factor are seen at six months old, then every six months until two years old, with a discharge assessment performed at three years old. Children with congenital infection as a risk factor are seen at three months old, six months old, and then every six months until two years old.

Assessments performed at these appointments depend on the child's age and developmental level. Assessments may include otoscopy, tympanometry, Auditory Brainstem Response (ABR), Visual Response Audiometry (VRA), Transitory Evoked Otoacoustic Emissions (TEOAEs), Distortion Product Otoacoustic Emissions (DPOAEs) and play audiometry. Children are discharged from the risk-based monitoring/ targeted surveillance programme when they have completed their appointment series and frequency-specific information has been obtained for each ear. For the children referred for risk-based monitoring/ targeted surveillance, the largest proportion of referrals from the study were generated from family history of a hearing loss, LBW, and prolonged ventilation. For the risk factors reported in children who developed postnatal hearing loss, 46.4% reported family history of a hearing loss as a risk factor, and 19.6% reported a syndrome or prolonged ventilation as a risk factor.

It is uncertain as to whether this approach would be feasible in South Africa as a high follow-up return rate is required and there may be the risk of missed cases if an initial, in-hospital screening is not performed, and if the caregivers don't attend the follow up appointments. In Kanji (2016b) study, a significant decrease in follow-up return rate for the six month diagnostic assessment was associated with; migration to another country, province or city, caregiver employment, and financial difficulties, for example, insufficient money for transport. Scheepers and le Roux (2014) suggest that caregiver's lack of knowledge of screen outcome and follow-up recommendations are most commonly related to follow-up default. These are important factors to consider when implementing a risk-based surveillance programme within the South African context.

A more feasible risk-based surveillance programme within the South African context could be to implement preschool hearing screening, which includes children aged three to five years of age. Hall (2016) explains that since the "1-3-6 rule" is not always successful it is important to perform preschool screening for hearing loss. This allows for identification of

hearing loss for children who did not receive a newborn hearing screen. Furthermore, it will identify children with delayed-onset or progressive hearing loss. This approach could be successful within the South African population where 90% of children are not receiving a newborn hearing screening (Theunissen & Swanepoel, 2008; Meyer & Swanepoel, 2011). Further studies are needed to assess the success of conducting preschool hearing screening, highlighting the importance of the current study. Ultimately, the JCIH (2007) recommends that guidelines indicating timelines should be adapted based on what is practical and suitable for practice.

The HPCSA (2018) recommends that due to the overburdened health care system in South Africa, children who present with risk factors for delayed-onset hearing loss should be monitored by their caregivers. Caregivers must therefore be informed of these risk factors at antenatal clinics. In addition, for these risk factors to be recorded on the Road to Health Booklets (RTHB), which is given to caregivers when their children are born. Furthermore, children who present with one or more of the five risk factors for continued audiological surveillance should be referred for a behavioural audiological assessment at eight to 10 months of age. In addition, surveillance protocols should be developed according to the ages of onset for the risk factors for progressive-onset hearing loss in children, as detailed in table 2.1.

Although co-ordinated guidelines specifying time frames for audiological monitoring is necessary, the same is needed in terms of developmental screening. Co-ordinated monitoring and/ or screening programmes allows for communication between audiologists, paediatricians, and other health care providers involved and is essential for the diagnosis and management of the high-risk population (Umrigar, et al., 2017).

2.6. Developmental Monitoring/ Surveillance and Screening

The American Academy of Paediatrics (AAP) (2001) recommends the use of surveillance and screening measures for the identification of children who would require developmental assessment and evaluation. However, as discussed previously, in South Africa co-ordinated developmental screening programmes are only available at certain health institutions/ clinics (Samuels, et al., 2012). This has implications for ECI within the South African context.

According to Almeleh, et al., (2016) two thirds of children below the age of six years are still not receiving the essential services that they require during the ECD period in South Africa. In addition, 63%, of young children in South Africa live in poverty and their development (physical, cognitive and emotional) is compromised because appropriate services are not received. In South Africa, many factors such as poverty and poor maternal education may influence developmental outcomes (Grantha-McGregor, et al., 2007). Ohonba, Ngepah and Simo-Kengne (2019) suggest that maternal education strongly influences child health outcomes in South Africa, which may be due to maternal educational levels having been found to be associated with improved compliance to ECI programmes (Diener, Zick, McVicar, Boettger & Park, 2017; Holte, Walker, Oleson, Spratford, Moeller et al., 2012).

The average age of identification of children with developmental delays in South Africa is not known. Evidence suggests that in developing countries, less than half of the eligible children are identified before entering school (Aly, Taj & Ibrahim, 2010). This could be especially true for South Africa, due to the lack of coordinated monitoring/ surveillance programmes.

There is a need for developmental monitoring now more than before, especially since advances in neonatal care has allowed for the survival of high-risk neonates. Claas, et al., (2011) explain that high-risk neonates are prone to a range of long term complications. For

example, improved rates of survival of VLBW, and especially extremely low birthweight (ELBW) infants may be associated with increased rates of neurodevelopmental handicap. Ramdin, et al., (2018) analysed the long-term developmental outcomes of high-risk neonates, more specifically late preterm infants at nine to 12 months, and then again at 15 to 18 months of age. Results revealed that 7.1% of the children presented with evidence of developmental disability. Although this was not considered a statistically significant, results could have been affected by the small sample size of 73 children. Furthermore, Ballot, Potterton, Chirwa, Hilburn and Cooper (2012) in their study analysed the long term outcome of VLBW neonates in South Africa. Results revealed that the prevalence of cerebral palsy and severe handicap is low, and is similar to that reported from other developing countries. However, this low rate of handicap may reflect the low survival rate of infants with a birth weight below 900 grams. Results further suggested that VLBW infants in this setting are a high-risk population likely to have learning and other difficulties at school going age and warrants long-term follow up. These results are also supported by more recent findings that suggest VLBW infants in South Africa are at risk of developmental delay and require long term neurodevelopmental follow-up (Ballot, et al., 2017).

Ongoing long term developmental follow-up utilizing appropriate monitoring and screening tools is therefore necessary for the high-risk neonate population. Especially since children's development is fundamentally cumulative in nature and therefore, the early years of life are the foundation for later development (Ali, 2013), including hearing development. There is still however a paucity of literature regarding the long term developmental outcome of all high-risk neonates, and not just the LBW population, in developing countries. This highlights the importance of the current study. This study is a follow-up study of high-risk neonates who were previously enrolled in a risk-based hearing screening programme, and aimed to explore the developmental outcomes of these children.

Employing monitoring and screening tools at PHC settings can facilitate early identification of children with developmental delays (Brothers, Glascoe & Robertshaw, 2008). This potentially assists families of all socio-economic standings to obtain early intervention for their children (United Nations International Children's Emergency Fund, 2013). Comprehensive tracking and follow-up systems are however essential to ensure that children are not only identified through screening, but also return for the appropriate assessments and intervention (Wang, et al., 2009). Various studies have, however, reported poor follow-up return rates (Wang, et al., 2009; Giannoni & Kass, 2010). Low participation rates lead to diminished effectiveness of early intervention programmes.

Reichert, Collet, Eickmann and Lima (2015) discuss that child developmental monitoring and surveillance is important as it is a fast and cost-effective application measure, for detecting children with risk of development delay.

2.6.1 Evaluation of developmental outcome measures. Many development screening tools have been developed and validated internationally. *The Denver Developmental Screening Test* (DENVER II), *Ages and Stages Questionnaire* (ASQ), and the *Parents' Evaluation of Developmental Status* (PEDS) and its accompanying tool the *Parents' Evaluation of Developmental Status: Developmental Milestones* (PEDS:DM) have the largest body of supporting evidence of screening tools that ranges from birth to kindergarten (8 years of age). There are a number of other screening tools being used in South Africa in both public and private sectors. These screening tools include; *The Bayley Scales of Infant and Toddler Development III (BSID III), The PEDS tools* (Abdoola, Swanepoel, Van Der Linde & Glascoe, 2019), as well as *The Developmental Milestone Checklist*, and *The Guide to Monitoring Child Development* (Sabanathan, Wills & Gladstone, 2015). Furthermore, Marlow, Servili and Tomlinson (2019) reviewed screening tools that are best applicable in detecting Autism Spectrum Disorder (ASD), and developmental delays in infants in low and

middle income countries. Their results revealed 10 tools which show promise for use across settings. Three tools specifically for ASD were *The Modified Checklist for Autism in Toddlers, Revised Follow-Up* (M-CHAT-R/F); *The Pictorial Autism Assessment Schedule* (PAAS); *Three-item Direct Observation Screen* (TIDOS). Seven were more for general developmental delays; *The Caregiver Reported Early Development Index* (CREDI); *The evaluation of the Guide for Monitoring Child Development* (GMCD); *The INTERGROWTH-21st Neurodevelopment Assessment*; *The Malawi Developmental Assessment Tool* (MDAT); *Rapid Neurodevelopmental Assessment* (RNDA); *The Ten Question Screening Instrument* (TQSI); and a *12-month screener*.

According to Van der Linde, et al., (2015) the only developmental screening tool which has been implemented nationally by the department of health in South Africa is integrated as part of the RTHB (Naidoo, Avenant & Goga, 2018), which is given to parents when their baby is born. This developmental checklist is a parent administered measure which is used to monitor and promote early child health, growth and development (Harrison, et al., 2005).

The accuracy of the RTHB integrated screening measure was compared against the PEDS in a study by Van der Linde, et al., (2015) in South Africa. Results indicated that the RTHB screening measure was ineffective as it failed to identify more than half of infants at risk of developmental delays or disorders within the PHC context. In addition, no clear referral process has been specified for the RTBH screening measure (Van der Linde, et al., 2015). The PEDS and PEDS: DM were however able to identify the children with developmental delays or disorders and provided a referral pathway. The nationally implemented developmental screening tool in South Africa therefore requires revision or replacement so that early detection and intervention can be implemented (Van der Linde, et al., 2015). Furthermore, for at-risk children to experience improved outcomes and reach their

fullest potential. Developmental screening also provides a platform for other preventative strategies such as parental training (Van der Linde, et al., 2015).

In addition, Abdoola, Swanepoel, Van der Linde and Glascoe (2019) compared the PEDS tools smartphone application and the *Bayley Scales of Infant and Toddler Development III (BSID III)* to detect developmental delays in 174 children aged 3 – 18 months in a primary health care clinic in South Africa. These authors did not report the tools to be effective when used in isolation due to the high-risk nature and young age cohort. The current study made use of the PEDS in conjunction with a hearing and communication checklist, and a hospital file review (where available).

The Parents' Evaluation of Developmental Status (PEDS) was therefore best suited for the current study as it also has a large body of evidence confirming the accuracy, validity and reliability of the tool (Van der Linde, et al., 2015). The Parents' Evaluation of Developmental Status (PEDS) has been validated in 20 studies during 2001 to 2010 in which a total of 7213 children were assessed (Macy, 2012). The PEDS: DM has been standardised, has excellent levels of validity and reliability, and sensitivity and specificity (83% and 84% respectively) (Van der Linde, et al., 2015). Furthermore, a recent study confirmed that use of the PEDS tools is feasible in South Africa (Silva, 2010) and that it is cost-effective.

On analysis of the actual measure and according to the Parents' Evaluation of Developmental Status (PEDS) (2013a), the PEDS tool is a combination of the PEDS and the PEDS: DM. *The Parents' Evaluation of Developmental Status* (PEDS) is a 10 item measure focused on eliciting and addressing parents' concerns. Whereas the PEDS: DM consists of six to eight items, which replaces informal milestones checklists (that are possible contributor to under detection of children with delays and disabilities), with an accurate one that has distinct cut offs for problematic versus typical development (PEDS, 2013a). Each item on the PEDS and PEDS: DM addresses different developmental domains; fine motor, gross motor,

expressive language, receptive language, self-help, and social-emotional. Failure on an item predicts delays in that domain. The tests take less than 10 minutes to administer and score (PEDS, 2013a).

The Parents' Evaluation of Development Status (PEDS) has been translated into various languages, including Zulu. The Zulu PEDS was however not be used in the current study as results from Van der Merwe, Cilliers, Maré, Van der Linde and Le Roux, (2017) study indicated that 54% of participants preferred the PEDS in English over the PEDS in Zulu. This indicates a skewed preference towards English with only minor associations between language preference, age, education and home language.

There is additional value in using the PEDS screening tools as obtaining information from caregivers was reported as the most important aspect of a clinical evaluation in the area of child development (Mbuyi, 2016). Mbuyi (2016) compared caregivers' evaluation of developmental status and professional assessment for early detection of developmental disorders in infants born at term with birth asphyxia at Tembisa hospital. *The Parents' Evaluation of Development Status* (PEDS) was utilized and results revealed that the PEDS is more likely to identify developmental delays or disorders than routine professional assessment. The routine professional assessment used in isolation presented with a risk of under-detection of developmental issues. Furthermore, it could result in lack of referral of infants for comprehensive developmental assessment and early intervention.

Since there are no coordinated developmental monitoring and surveillance systems currently in place in the South African public and private healthcare sector (Samuels, et al., 2012), it is important that improvements are made. This is imperative as ECD screening is an important strategy for promoting child well-being. The current study aimed to determine the long term developmental outcomes of the high-risk population using the PEDS measures.

This chapter has highlighted the health care challenges affecting ECD within the South African context. Risk-based monitoring and surveillance has been identified as a possible method in overcoming some of these challenges to ensure early identification of hearing loss. There is however no standard monitoring system being implemented in detecting developmental delays, and there is a paucity of literature in establishing contextually relevant risk factors for delayed hearing and general development delays within the South African context. There is also a lack of follow-up studies on children who were considered high-risk at birth, in terms of long term developmental and audiological outcomes following newborn hearing screening. Such studies are imperative to ensure appropriate monitoring and intervention during the ECD phase.

Chapter 3: Methodology

This chapter provides a description of the research aims, design, sampling methods and research instrumentation used, and process followed. Results from the pilot study are presented and the data analysis method is described. The chapter ends with a discussion of reliability, validity and ethical considerations that were considered and adhered to in this study.

3.1. Aims of the study

3.1.1 Primary aim. The main aim of the current study was to determine the developmental outcomes of children who were considered high-risk at birth and previously enrolled in a risk-based hearing screening programme.

3.1.2 Sub-aims.

- To describe the case history factors of the participants' children, who were considered high-risk at birth.
- To describe the general development of the participants' children (this includes fine motor, gross motor, expressive language, receptive language, self-help, and social-emotional domains, and other concerns).
- To describe the audiological development of the participants' children.
- To determine the relationship between case history factors at birth, and the current general and audiological development of the participants' children.

3.2. Research Questions

What are the general and audiological developmental screening outcomes in a group of children considered high-risk at birth and previously enrolled in a risk-based hearing screening programme?

Is there a relationship between case history factors at birth, and the current general and audiological development of children who were considered high-risk at birth and enrolled in a risk-based hearing screening programme?

3.3. Research Design

A descriptive, cross-sectional, prospective cohort design was employed with an integration of retrospective aspects. As defined by Julius and Wright (2002), a descriptive study addresses one or more descriptive research questions. Its main aim is to develop or add on to an existing body of knowledge, and therefore usually builds upon knowledge obtained from other studies. In quantitative, descriptive research the aim is to describe the data and characteristics about the phenomena (Offredy & Vickers, 2010). This research design was selected to specifically meet the current study's aims of describing the current general and audiological development of children who were considered high-risk at birth and enrolled in a risk-based hearing screening programme. A descriptive study also involves the exploration of suspected associations between variables (Julius & Wright, 2002).

The primary aim of a cross-sectional study is to analyse the characteristics of a given population at a certain point in time (Liu, 2008). Cross-sectional studies are therefore mostly used to determine the frequency distribution of certain outcomes, and whether associations between two variables exist (Liu, 2008), for example, whether there is an association between a particular factor and a certain outcome (Martin, 2012). This study followed-up on a group of children who were considered high-risk at birth, and aimed to explore whether an association exists between certain risk factors and delayed-onset developmental difficulties, including delayed-onset hearing loss.

A cohort study is a form of research design which longitudinally observes a particular group of individuals who share a common attribute (Hagger-Johnson, 2014). All children in

the current study shared a common attribute as they were all considered high-risk at birth. A cohort study can be conducted prospectively and/ or retrospectively. According to MacGill (2016) in a prospective cohort design, participants are followed-up to determine the development of a possible impairment. In a retrospective cohort design, the researcher looks at data that already exists, and attempts to recognise risk factors for certain impairments. Both prospective cohort and retrospective cohort designs are however subject to biased.

Biases associated with prospective cohort studies include; imprecise risk assessment because of the lack of understanding into error rates (Trucco & Cavallin, 2006), the inability to contemplate combinational events (Marx & Slonim, 2003), and unreliable risk identification because of judgement inconsistency (Bonnabry, et al., 2005). Possible biases of retrospective cohort designs include; underreporting (Johnson, 2003; Olsen, et al., 2007), recall bias (Hassan, 2006), and incomplete data (Barach & Small, 2000). In order to reduce these biases, there is potentially an advantage of using prospective and retrospective designs to triangulate data (Hayes, Slater & Snyder, 2008).

Although both prospective and retrospective deign methods are individually valuable, Kessels-Habraken, Van der Schaaf, De Jonge, Rutte and Kerkvliet (2009) revealed that the triangulation of design methods can possibly improve the efficiency of analysis, as well as create a better and less bias picture of risks. The integration of design methods was therefore useful for the current study as it allowed the researcher to identify risk factors possibly associated with delayed general and audiological developmental delays in a less bias manner. The results from an integration method could also ultimately support hospital management in setting their priorities for patient safety and assigning resources to the most imperative problems (Battles, Dixon, Borotkanics, Rabin-Fastmen & Kaplan, 2006). This is particularly useful within the South African health care system where there is restricted resources and limited manpower (Swanepoel, et al., 2009; Swanepoel, 2006).

3.4. Participants

3.4.1 Sampling. A non-probability, purposive sampling method was utilized. Purposive sampling involves the deliberate selection of individuals, on the basis of a predefined criteria (DePoy & Gitlin, 2011). The current study deliberately selected caregivers of children who were participants in the risk-based hearing screening study by Kanji (2016b). This study aimed to explore risk-based hearing screening within a developing country, through early detection of hearing loss in high-risk neonates, within an academic hospital complex in Gauteng, South Africa.

The study by Kanji (2016b) comprised 325 high-risk neonates from *Charlotte Maxeke Johannesburg Academic Hospital* (CMJAH) and *Rahima Moosa Mother and Child Hospital* (RMMCH). Of the 325 participants, 22.2% were from CMJAH and 77.8% were from RMMCH. The current study only selected participants from RMMCH, as it was reported that there was a much better follow-up return rate noted at this hospital. The current study's sample is therefore representative of caregivers whose children were enrolled in a risk-based newborn hearing screening study, which was conducted at RMMCH during 2013 - 2015.

3.4.2 Inclusion and exclusion criteria. Children of the caregivers had to have attended the initial and repeat hearing screening and/ or the diagnostic audiological assessment as part of the study by Kanji (2016b). Consent also had to be obtained from the caregivers.

3.4.3 The sample. A total of 187 caregivers were contacted to participate in the current study; 71 that attended the diagnostic assessment, and 116 that attended the repeat hearing screen in the risk-based newborn hearing screening study by Kanji (2016b).

The sample consisted of 67 caregivers whose children were enrolled in a risk-based newborn hearing screening programme. The caregivers' evaluation of their child's

development was assessed in this study, and therefore the primary participants in the current study were the caregivers.

3.4.4 Participant recruitment. Participants were recruited through retrospective means from contact details collected as part of the study by Kanji (2016b). All caregivers of the children who met the inclusion criteria were contacted telephonically by the research supervisor who was the researcher of Kanji (2016b) study to inform them about the current study. If caregivers were willing to participate in the current study, verbal consent was obtained by the research supervisor for their contact details to be shared with the current researcher. The number of caregivers who could not be contacted, and their reasons were provided by the research supervisor for the current researcher to document. The current researcher contacted the caregivers who agreed to participate. A suitable time was arranged at RMMCH for the general development screening and audiological surveillance appointment. Written consent was then obtained on the predetermined date and time.

3.4.5 Description of study site. The current study was conducted at RMMCH, which is a government and academic hospital located in Coronationville, Johannesburg. This hospital provides services for hospital admissions, where wards include; general wards, neonatal wards, Kangaroo Mother Care (KMC) wards and a NICU. A paediatric outpatient department (POPD), and paediatric specialist clinics, for example, neonatal follow-up clinics were also included (The University of the Witwatersrand, 2019).

3.5. Research Instrumentation

The measures that were used to conduct this investigation included:

3.5.1 The Parents' Evaluation of Developmental Status (PEDS) and The Parents' Evaluation of Developmental Status: Developmental Milestones (PEDS: DM). The PEDS is a 10 item measure centred on eliciting and addressing parents' concerns. The PEDS: DM

consists of six to eight items, which replaces informal milestones checklists with an accurate one that has distinct cut offs for problematic versus typical development (PEDS, 2013a). Each item on the PEDS and PEDS: DM addresses different developmental domains namely; fine motor, gross motor, expressive language, receptive language, self-help, and social-emotional. Failure on an item predicts delays within that domain. The PEDS tools have evidence confirming the accuracy, validity and reliability of the tool (Van der Linde, et al., 2015). As mentioned previously, the accuracy of the RTHB integrated screening measure was compared against the PEDS in a study by Van der Linde, et al., (2015) in South Africa PHC context. Van der Linde, et al., found the PEDS measures to identify children with developmental delays or disorders more accurately than the RTHB screening measure. In addition, a clear referral pathway was provided by the PEDS measures. A recent study by Silva (2010) has confirmed the accuracy of the PEDS tools in South Africa, and its cost-effectiveness.

Both measures were accessed and scored via 'PEDS online'. Researchers are encouraged to use the online measure as the scoring is accurate and automated, and it generates an exportable database of results that can be concatenated with other measures or study protocols (PEDS, 2014). Both tests, the PEDS and PEDS: DM, take less than 10 minutes to conduct and score (Glascoe & Robertshaw, 2010).

The researcher firstly conducted the PEDS, and thereafter conducted the PEDS: DM. Depending on the child's corrected age, the appropriate PEDS: DM form was selected as depicted in Table 3.1 below. The researcher was not given permission to replicate or reproduce the PEDS resources in any way, and therefore these forms will not be included in the appendices.

Table 3.1

The Appropriate PEDS: DM Form to use for the Corrected age of the Participants' Children

PEDS: DM Form	Corrected Age Range
Form G	17 – 19 months
Form H	20 – 22 months
Form J	23 – 25 months
Form K	2 years, 2 months – 2 years, 4 months
Form L	2 years, 5 months – 2 years, 9 months
Form M	2 years, 10 months – 3 years, 2 months
Form N	3 years, 3 months – 3 years, 7 months
Form P	3 years, 8 months – 4 years
Form Q	4 years, 1 month – 4 years, 5 months

A few questions on the PEDS: DM (particularly from form N, P and Q) require the child to be present, which wasn't always possible in the current study. The author of the PEDS was contacted directly, and permission was given to adapt such questions so that these questions can be phrased to the participants in a hypothetical case situation (Appendix E). For example, "if you asked your child…" In terms of the effects on scoring, parent-report versus hands-on administration have excellent inter-rater reliability (PEDS, 2013b). If the caregiver stated, "I don't know", "never tried" or "I'm not sure" (or similar answers), the researcher would mark the answer options as "no". The author of the PEDS further suggested that a caregiver not knowing what their child can do also gives insight into the caregiver's relationship with the child and tasks that the child is exposed to.

The Parents' Evaluation of Developmental Status (PEDS) (2015) describe the interpretation of results as follows:

Results from the PEDS indicated parental predictive and non-predictive concerns, with specific reference to the domains which were assessed. Following analysis, an evidence-based pathway was provided which indicated the child's developmental level based on the type/ amount of parental concerns. Table 3.2 was adapted from Glascoe (2018) and demonstrates what each path represents.

Table 3.2

The Interpretation of the PEDS Main and Sub-Pathways

Main and Sub-pathways	Interpretation
A	**High risk for developmental difficulties** Caregiver has multiple developmental and frequent behavioural concerns
A-1	Need for speech-language evaluation
A-2	Need for testing by developmental or school psychologist
B	**Moderate risk for developmental difficulties**
B-1	Caregivers with non-developmental concerns (for example, sleeping, or vision)
B-2	Caregivers have at least one predictive developmental concern
C	**Elevated risk for developmental difficulties**
C-1	Children four and a half years of age and older Additional behavioural/ emotional screening required
C-2	Children younger than four and a half years of age Council caregivers about their concerns and monitor progress
E	**Low risk for developmental or behavioral problems**

Secondly, results from the PEDS: DM revealed the developmental area at which the child was performing at a less than optimal level. Results were categorized according to the domains milestones that have been met and unmet. Ultimately, a result and a brief

recommendation was provided. For example, at-risk for development/ mental health problems, and a brief recommendation was provided.

Hearing was not considered by the PEDS measures, and therefore a hearing and communicative checklist was additionally used in the current study.

3.5.2 Hearing and communicative checklist. One of the components of the *Developmental Systems Model* which forms the basis of ECI, includes the creation of effective and efficient screening programs, and monitoring/ surveillance programmes. In order to meet this models requirements the *"How Does Your Child Hear and Talk"* developmental milestones provided by The American Speech-Language-Hearing Association (ASHA, 2006), was used as a hearing and communicative checklist (Appendix F).

This checklist identifies ages at which children should be demonstrating certain audiological skills. The researcher asked the participants the questions, and responses were recorded as either yes/ no. For example, at 3 to 4 years, "does your child hear you when you call from another room?" If the participant responded with no to any of the questions relating to the expected milestone at that age, it would indicate audiological concern. Caregiver concern of audiological delays must be taken seriously as Störbeck and Young (2016) suggest that it can decrease the age of hearing loss identification.

The checklist format was created so that it can potentially be used to assist paediatricians with appropriate referrals. According to the HPCSA (2018) paediatricians are responsible for monitoring the general health and well-being of the infant. This includes the initiation of referrals for medical speciality evaluations necessary to determine hearing loss aetiology, monitoring of middle-ear status, and reviewing risk indicators of the infant. Paediatricians are also required to ensure periodic audiological assessment for infants with risk factors for progressive and/ or late onset hearing loss, and monitor developmental

milestones. Utilizing early childhood audiological monitoring through a checklist within the paediatric settings will therefore create an easily accessible monitoring/ surveillance system for EHDI.

3.5.3 Hospital file review. Initially the paediatric hospital files, and if necessary, the audiology hospital files were planned on being reviewed. However, one hospital file which includes all children's medical information is utilized at RMMCH.

The researcher therefore accessed the hospital files of the children in the current study. The paediatricians' notes were important to analyse as it revealed the case history factors that classified the children as high-risk neonates, as well as their medical history thereafter. It therefore assisted the researcher in to gaining better insight into the general development of the children. According to Ali (2013) children's development is fundamentally cumulative in nature and therefore, the early years of life are the foundation for later development. Information from the files were analysed according to a file review checklist (Appendix H). The hospital file review checklist (Appendix H) consisted of the following subheadings; general information, significant medical history at birth, and neonatal follow-up (NNFU) appointments.

The general information section captured information regarding demographic details e.g. date of birth, gender, home language etc. The significant medical history at birth section captured the case history information at birth. In addition, At RMMCH, follow-up appointments are usually booked six weeks after discharge, and then four months after that until around one year of age. Information captured at NNFU appointments usually include; age, weight, length, head circumference, diet, immunisation, developmental milestones, general examination, HIV status, ophthalmology and hearing test results. Information relating to re-admissions and out-patient appointments at RMMCH were also recorded.

The file review was also done to assess if any of the children returned to the audiology department after discharge from the risk-based newborn hearing screening programme. Audiological concerns and outcomes from the file review were also documented according to a checklist (Appendix I). This checklist documented whether the child *passed/ referred* their initial and repeat hearing screens, results from their diagnostic assessment (if conducted) as part of Kanji (2016b) study. The checklist also included information relating to the initial reason as to why the child returned to the audiology clinic, as well as the outcome of any reported concerns. The researcher captured this information utilizing a summary form (Appendix J).

3.6. Research Procedure

Data collection commenced once ethical clearance was granted by the Human Research Ethics Committee of the University of the Witwatersrand (medical) (Appendix A) and permission was obtained from the research site, RMMCH (Appendix B), as displayed in figure 1 below. Although planned for, the use of a trained interpreter was not necessary as all caregivers were proficient in English

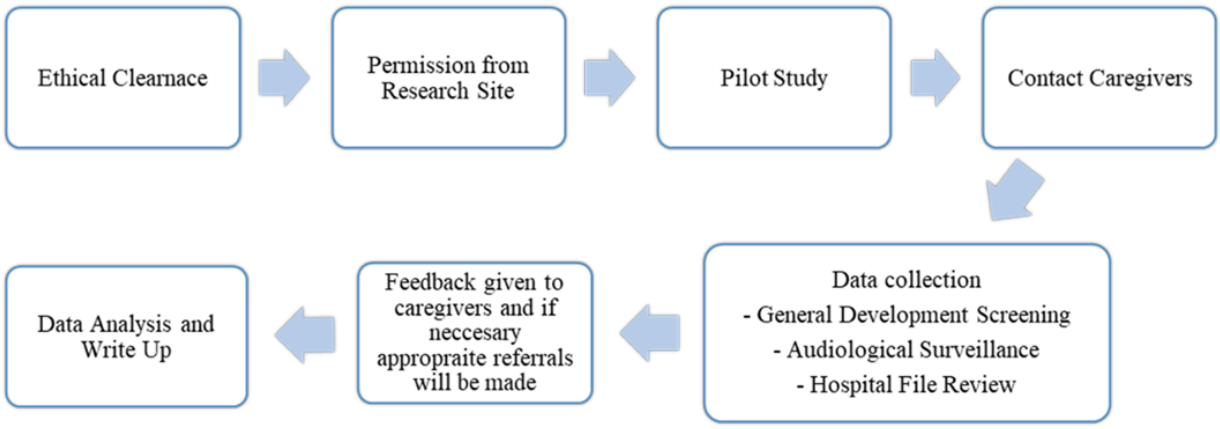

Figure 1. The Research Procedure of the Current Study.

3.6.1 Pilot study. Once ethical clearance was received and permission from the research site was granted, a pilot study was conducted. Blessing and Chakrabarti (2009)

emphasise the importance of trialling a research approach, to identify potential problems that may affect the quality and validity of the results. Possible changes may include formulating less ambiguous questions or adding other methods to capture certain aspects in more detail. A pilot study usually involves one or two cases, and the setup should be as close to possible to the setup of the intended study (Blessing & Chakrabarti, 2009).

3.6.2 General development screening. The researcher and participants met at RMMCH on the predetermined date and time, for the general development screening and the audiological surveillance appointment. Prior to data collection, information letters regarding the study (Appendix C), and written consent forms (Appendix D) agreeing to participate in the study were given to caregivers. If the participants had any questions regarding the study, they would be answered. Participants were also informed that all information would remain confidential.

Since the majority of the risk-factors present on the HPCSA (2018) HRR for delayed-onset hearing loss have also been associated with other general developmental delays, the PEDS and PEDS: DM was utilised to assess the general development of the children in the study.

3.6.3 Audiological surveillance. The *"How Does Your Child Hear and Talk"* developmental milestones provided by the *American Speech-Language-Hearing Association* (ASHA, 2006), was used as a hearing and communicative checklist (Appendix F).

Results from the PEDS, PEDS: DM and hearing and communicative development checklist was thereafter interpreted, and feedback was given to the participants. If participants had not already been referred by the paediatrician or sought out the necessary assistance that may be required, appropriate referrals were made. Referrals were made by utilizing a referral letter (Appendix G).

3.6.4 Hospital file review. The researcher captured information on the hospital file review checklist (Appendix H). If the hospital file was not available, the researcher verbally requested permission from the participants to provide the information detailed on the file review checklist (Appendix H). If the participants expressed any uncertainty regarding the information they had provided, the researcher obtained verbal consent for the information to be cross-checked by the research supervisor who had previously obtained consent to review their child's file for the same information as part of her study. Information was matched by hospital number provided by the researcher to ensure anonymity.

3.7. Pilot Study Aims and Results

3.7.1 Pilot study aims. The current study's pilot study was therefore conducted on two caregivers attending the neonatal follow-up clinic at RMMCH, and their children's hospital files. Aims for the pilot study included; testing the adequacy of the research measures, determining caregiver understanding of the research measures, determining the time period that the research measures will take to conduct and identifying logistical problems with procedures.

3.7.2. Pilot study results.

3.7.2.1 Demographic profile of the participants from the pilot study. The pilot study comprised two caregivers attending the NNFU clinic, namely 'pilot 1' and 'pilot 2'. Both participants were female and South African. One participant attained a primary education, and the other attained a secondary education respectively. A summary of the demographic details of the participants from the pilot study are provided in table 4.1 below.

Table 4.1

The Demographic Details of the Participants in the Pilot Study

	Pilot 1	Pilot 2
Gender	Female	Female
Age	26	32
Race	Black African	Coloured
Nationality	South African	South African
Highest Level of Education	Primary (completed Grade 7)	Secondary (completed Matric)
Home Language	isiZulu	Sepedi

3.7.2.2 Demographic profile and the case history factors of the participants' children from the pilot study. The child of the participant, 'pilot 1' was female and 1 year and 1 month in terms of age. In terms of case history factors, this child was born full-term, presented with a normal birth weight and normal APGAR scores. Furthermore, this child stayed in hospital for a period of 17 days, presented with a retroviral disease (RVD) status of HIV exposed (HEI) and meningitis.

The child of the participant, 'pilot 2', was male and had a chronological age of 1 year 4 months, and an adjusted age of 1 year 1 month. In terms of case history factors, the child was born preterm at 28 weeks, and presented with a LBW. This child in addition presented with normal APGAR scores, stayed in hospital for a period of 21 days and presented with an HIV status of infected unexposed (HEU). Lastly, this child was ventilated with intermittent positive pressure ventilation (IPPV) for a period of one day. A summary of the children's demographic details and case history factors are provided in table 4.2 below.

Table 4.2

The Demographic Details and Case History Factors of the Participants' Children in the Pilot study

	Pilot 1	Pilot 2
Gender	Female	Male
Chronological Age	1 year 1 month	1 year 4 months
Adjusted Age	1 year 1 month	1 year 1 month
Birth Weight	Normal	Low Birth Weight
APGAR scores	Normal	Normal
Length of Hospital Stay	17 days	21 days
HIV Status	Exposed	Unexposed
Neurological Conditions	Meningitis	None
Ventilation	No	Yes

3.7.2.3 General developmental screening outcomes from the pilot study. The results from the PEDS revealed that only the participant from 'pilot 1' expressed a medical concern, relating to her child's seizures. The results from the PEDS further revealed that both children presented on path B-2, which indicates a moderate risk for developmental concern, and often behavioural concerns. Results from the PEDS: DM revealed that both children did not meet their gross motor milestones.

3.7.2.4 Hospital file review outcomes from the pilot study. The hospital file review revealed that both participants returned for the NNFU appointments, and have thus far attended two appointments.

On analysis of the NNFU notes, paediatricians commented on movement and physical development, communication and language skills, social-emotional development and feeding. The hearing development was only commented on once for the child from, 'pilot 1'. In addition, the neurodevelopmental milestones were not commented on for both children.

The paediatricians suggested that both children presented with developmental concerns. More specifically, the child from 'pilot 1' was being managed for seizures, and was referred to the dietician for severe acute malnutrition (SAM), and the physiotherapist for the regression of milestones. The child from, 'pilot 2', was referred to the physiotherapist for delayed motor milestones. Lastly, none of the children from the pilot study were re-admitted to hospital or returned as out-patients for any concerns at RMMCH.

3.7.2.5 Audiological history of the children in the pilot study. The file review revealed that only the child from 'pilot 1' received a newborn hearing screen. This child presented with an initial hearing screen result of, *'pass'*. No further audiological testing/ surveillance was done for either of the children.

3.7.2.6 Hearing and communicative checklist outcomes from the pilot study. Results from the hearing and communicative checklist indicated that both participants did not express any audiological concern. Furthermore, both children met their audiological milestones for their corrected age. These results correlate with the newborn hearing screening result of, *'pass'*, for the child from, 'pilot 1'. Towards the end of the appointment the researcher gave advice as to how to promote development. All necessary professionals were being seen, and therefore no referrals were made.

3.7.2.7 Participant understanding of the research measures. Both participants' home languages were languages other than English, and received either a primary or secondary education. Nevertheless, the participants were proficient in English and did not require any assistance in understanding the researcher, questions from the PEDS measures, and the hearing and communicative checklists.

3.7.2.8 Time periods the research measures took to conduct. The pilot study revealed that the PEDS would take five to 10 minutes to conduct, depending on the amount of detail provided by the participants. The PEDS: DM took five minutes to conduct, and the hospital

file review took 10 to 15 minutes to conduct. This was also dependent on the amount of detail reported on in the file. Each appointment therefore took an average of 20 to 30 minutes to conduct.

3.8. Data Analysis

All data analysed in the current study was done by a statistician using the SPSS version 25 statistical program.

In order to address the first sub-aim, information obtained from the hospital file review and data obtained from Kanji (2016b) study was analysed descriptively. According to LAERD Statistics (2013) descriptive statistics helps describe, show or summarize data in a meaningful way such that, for example, patterns might emerge from the data. It also describes and comments on the data obtained, and allows for the opportunity of comparison of results to be made among the subjects. Descriptive statistics allowed the current researcher to determine the most and least common case history factors that the participants' children presented with in the current study.

In order to address the second sub-aim of the current study, data obtained from the PEDS, PEDS: DM and hospital file review was analysed descriptively. The data obtained from the PEDS addressed participants' general development concerns. Based on these concerns the 'PEDS online' specified domains where concerns arose, and the child's level of risk according to a pathway. Secondly, the 'PEDS online' analysed the results from the PEDS: DM and revealed which developmental domains children met and unmet their milestones. Thirdly, data obtained from the hospital file review revealed developmental and medical concerns detected at NNFU appointments, out-patient visits and re-admissions. Descriptive statistics allowed the current researcher to establish the most and least common developmental domain/s where caregiver concerns arose, pathways children presented on and

domains where milestones were unmet. In addition, the most and least common medical and developmental concerns reported in the hospital file.

The hearing and communicative checklist was used in the current study to address the third sub-aim of describing the participants' children's audiological development. Results were also analysed using descriptive statistics. Whether participants presented with audiological concern, if the child returned for further testing, and the overall outcomes as to whether audiological milestones were met, were all descriptively analysed by frequency.

In order to address the last sub-aim of the current study, the Chi-square test was initially used to determine if an association exists between the case history factors at birth and the current general and audiological development of the children. According to Jawlik (2016) the Chi-Square test is used to determine if there is a significant relationship between two categorical variables. When sample sizes are however small and unbalanced, it is worth considering the Fisher's exact test *p*-value, instead of the chi-square test, for more reliable conclusions to be drawn (Pallant, 2011). Secondly, when the expected frequencies are 5 or more in the two way contingency tables, the Fisher's exact test should be utilized (Pallant, 2011). The current study's data meets both of these criteria, and therefore the Fisher's exact test *p*-value was considered in the current study, and not the chi-square test. There is a long tradition of using the *p*-value of 0.05 as the cut-off for rejection or acceptance of the null hypothesis (Mehta & Patel, 1996).

3.9. Validity and Reliability

Validity is defined as the degree to which an instrument correctly measures what it purports to measure (Kimberlin, & Winterstein, 2008). Reliability refers to the degree to which a research instrument consistently yields the same results if it is used in the same situation on repeated occasions (Heale & Twycross, 2015). More specifically, content validity

ensures that the instrument sufficiently covers all the content that it should with respect to the variable (Heale & Twycross, 2015). In the current study, the PEDS and the PEDS: DM covered all general development domains, and the hearing and communicative checklist ensured all audiological milestones were addressed for each age group. Triangulation of data was also ensured through inclusion of a hospital file review. In the instance where hospital files were not available, the researcher verbally requested permission from the participants to provide the information detailed on the file review checklist (Appendix H). If participants expressed uncertainty about the information they had provided, the researcher obtained verbal consent for the information to be cross-checked by the research supervisor who had previously obtained consent to review their child's file for the same information as part of her study.

Secondly, construct validity refers to whether one can draw inferences about test scores related to the concept being analysed (Heale & Twycross, 2015). *The Parents Evaluation of Development Status* (PEDS) and the PEDS: DM is a formal test with a large body of evidence confirming the accuracy, validity and reliability of the tool, as well as in the South African population (Van der Linde, et al., 2015; Silva, 2010) and therefore inferences can be drawn from the results. Lastly, validity was ensured through standard test administration. The same researcher administrated all monitoring, surveillance and screening measures, and reviewing of all files, and therefore data capturing errors were eliminated. In addition, since the 'PEDS online' was utilized, scoring was accurate and automated which improved the validity and reliability of the study.

With regards to reliability, an argument could be made that the current study has poor reliability, because of the recall bias associated with the caregiver reporting from their experience. However, since this study is prospective cohort in nature; following up on the same caregivers reduced the likelihood of recall bias, as data on the exposure and the

outcome are collected in real time (El-Masri, 2014). In addition, because of the variety of measures used in this study, the researcher applied methodological triangulation and therefore results were cross-checked. Methodological triangulation of data reduces the threat to reliability, and captures the intricacy of the study, and offers the prospect of enhanced confidence (Bryman, 2010).

3.10. Ethical Considerations

Ethical clearance was obtained from the Human Research Ethics Committee (Medical), clearance number (M180468) (Appendix A). Only once the ethics certificate and permission letter (Appendix B) was received did the research commence. The ethical principles used in healthcare ethics often include; justice, autonomy, non-maleficence, and beneficence (Summers, 2014). The current study utilized the same principles.

Firstly, non-maleficence refers to not doing harm to others (Summers, 2014). This was done by ensuring participants did not experience any discomfort, inconvenience or expense, and only benefited from participating in the current study. To ensure participants were at ease, all aspects of the study were explained. Furthermore, participants were encouraged to ask questions and were reassured that results would remain confidential. Participants were also reimbursed with transport money worth R50.

Secondly, beneficence refers to taking positive and direct steps to benefit others and promote good (Summers, 2014). To ensure that participants benefited positively from the appointment, appropriate advice and referrals were made when necessary. All participants were given advice as to how to promote their child's development.

Thirdly, the principle of autonomy refers to respecting others' rights to make choices and actions based on their values (Summers, 2014). Participants were given the choice to

participate in the current study. Furthermore, they were ensured of their right to withdraw from the study at any point, with no negative consequences.

Lastly, since children being studied were considered high-risk at birth, they form part of a vulnerable group of children, and therefore the principle of justice was considered. This principle advocates that individuals should not bear an unfair share of the burdens of participating in research, nor should they be unethically excluded from the benefits of participating in the research (Panel on Research Ethics, 2012). All participants in the current study were therefore given equal amounts of time, attention, and management. In addition, the researcher paid close attention to detail from the beginning of the study through to study termination and presentation of results. This was to ensure minimum harm and maximum benefit of the current study.

Chapter 4: Results

This chapter aims to present the findings obtained from all three measures; the general developmental screening measures (the PEDS and PEDS: DM), the hearing and communicative checklist and the hospital file review. Results were descriptively analysed for the first three sub-aims, and statistically analysed for the last sub-aim. All data has been portrayed in tables and graphs. Furthermore, results will be described in accordance to the respective sub-aims.

4.1. Main Study Results

A total of 187 caregivers were contacted to participate in the current study; 71 that attended the diagnostic assessment, and 116 that attended the repeat hearing screen in the risk-based newborn hearing screening study by Kanji (2016b). The current study sample consisted of 67 participants whose children were considered high-risk and enrolled in a risk-based hearing screening programme at RMMCH. One hundred and twenty (64.2%) caregivers contacted therefore did not participate in the current study.

Reasons for not participating varied, with the most common reason being a change in contact numbers for 87 (72.5%) caregivers. Figure 2 displays the reasons as to why caregivers contacted did not participate in the current study.

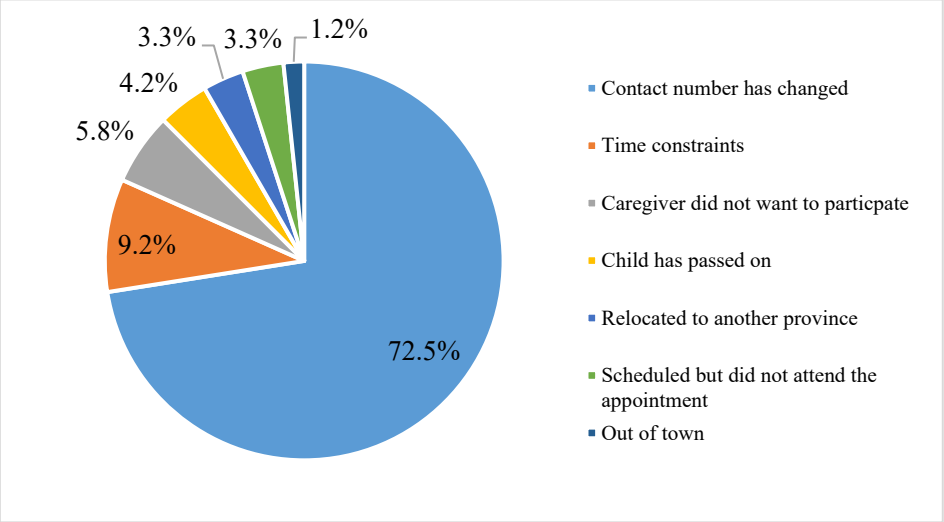

Figure 2. Reasons as to why Caregivers Contacted were not able to participate in the Current Study.

4.1.1 Demographic profile of the participants. Of the 67 participants, the majority were female, (95.5%), and three (4.5%) were male. The mean age of the participants was 34.6 years (*SD* = 5.98 years; range years 20 – 49). Figure 3 below displays the percentage of participants in each age category.

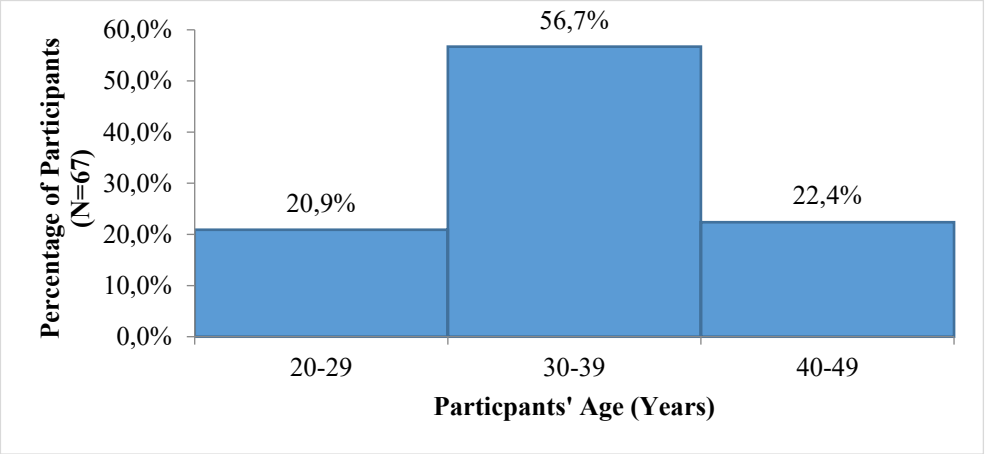

Figure 3. The Distribution of the Ages of the Participants.

In terms of race, the majority of the participants, 57 (85.1%) were Black African, followed by seven (10.4%) being Coloured. Two participants (3.0%) were White, and one (1.5%) was Indian. In terms of nationality, the majority of the participants, 49 (73.1%) were South African, followed by 14 (20.9%) who were Zimbabwean, two (3.0%) Ethiopian, one

(1.5%) from the Democratic Republic of Congo (DRC), and one (1.5%) was Nigerian. Analysis of home language revealed, the majority (20.9%) spoke isiZulu, whilst 12 (18%) spoke Ndebele, eight (12%) spoke English, eight (12%) spoke Tswana, and eight (12%) spoke Sepedi. Afrikaans, Siswati, Amhoric, Tsonga, Xhosa, French, Shona, Igbo and Sotho were spoken by three or less participants, as displayed in figure 4 below.

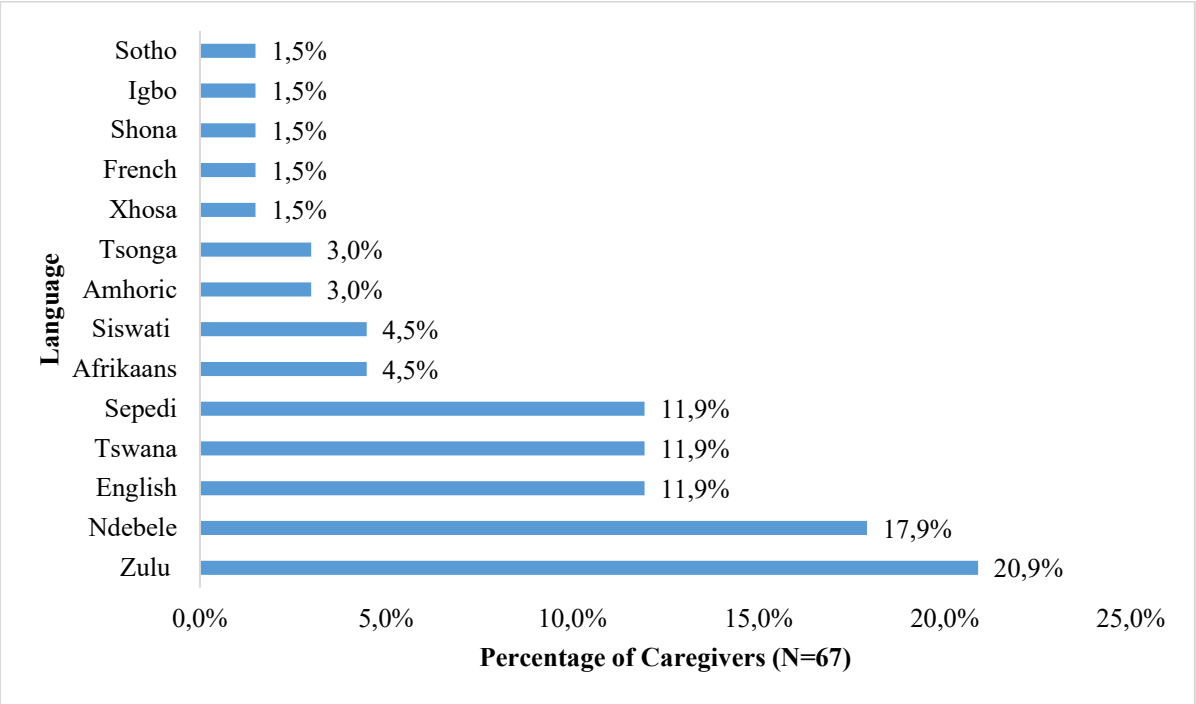

Figure 4. The Distribution of the Home Languages of the Participants.

In terms of education, 52 (77.6%) of the participants received a secondary education, and 15 (22.4%) received a tertiary education. Figure 5 provides further details as to the highest level of education that the participants attained.

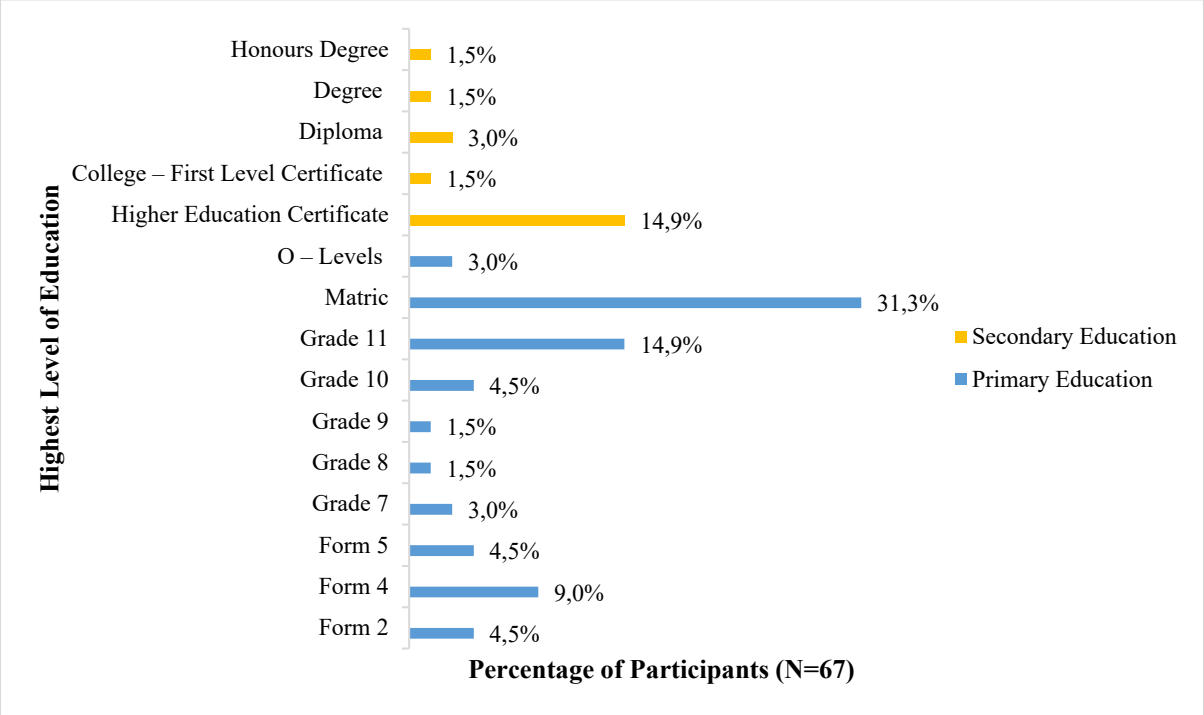

Figure 5. The Distribution of the Highest Level of Education of the Participants.

4.1.2 Demographic profile of the participants' children. Each participant had one child enrolled in the risk-based NHS programme, resulting in a total of 67 children. Thirty nine (58.2%) of the participants' children were female, and 28 (41.8%) were male.

In terms of chronological age, the mean chronological age was 49.2 months (*SD* = 5.1 months; range months 41 – 61). Figure 6 displays the number of children in each age category.

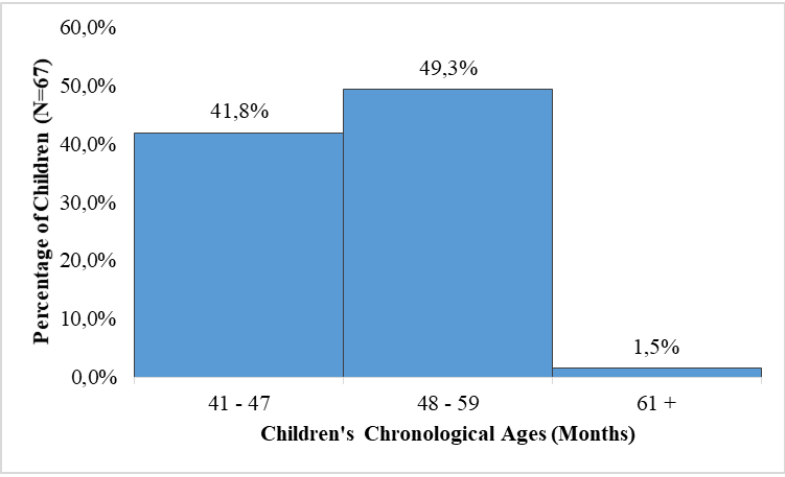

Figure 6. The Distribution of the Chronological Ages of the Participants' Children.

All of the children in the current study were preterm, and therefore the mean adjusted age was 47.2 months (*SD* = 5.1 and range months of 38 – 59). A representation of the distribution is provided in figure 7 below.

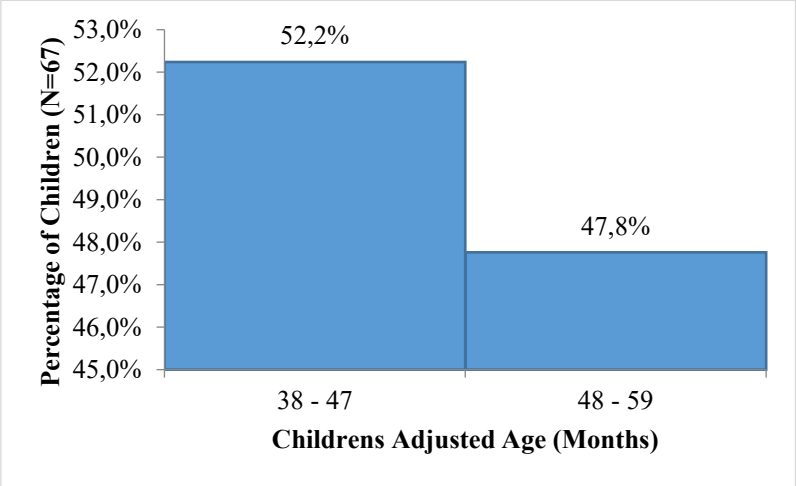

Figure 7. The Distribution of the Adjusted ages of the Participants' Children.

4.1.3 Description of the case history factors of the participants' children. In order to address the first sub-aim of the current study, descriptive statistics was used to analyse results. Case history information was captured from the children's hospital files at RMMCH. Hospital files were only found for 28 (41.8%) of the 67 children. The remaining 39 files were reported to have been considered old and therefore bundled and taken away for storage to another site. As a result, these files could not be located. In these cases, the researcher verbally requested permission from the participants to provide the information detailed on the file review checklist (Appendix H). Participants agreed to this, and also voluntarily provided the researcher with the Road to Health Booklet (RTHB) to verify the birth and medical history which would have been documented in the file. When participants expressed uncertainty about any of the information they had provided, the researcher obtained verbal consent for the information to be cross-checked by the research supervisor who had previously obtained consent to review their child's file for the same information as part of her

study. Information was matched by hospital number provided by the researcher to ensure anonymity.

Baseline case history information obtained included; gestational age, birth weight, APGAR scores, length of hospital stay, in-utero infections, bilirubin level and treatment, HIV exposure, family history of hearing loss, ventilation and the type, ototoxic medication and the type of medication, neurological conditions, syndromes, and any additional case history information. Figure 8 below displays the most and least common case history factors present in the current study. The percentages do not sum to 100% since some children presented with more than one risk factor.

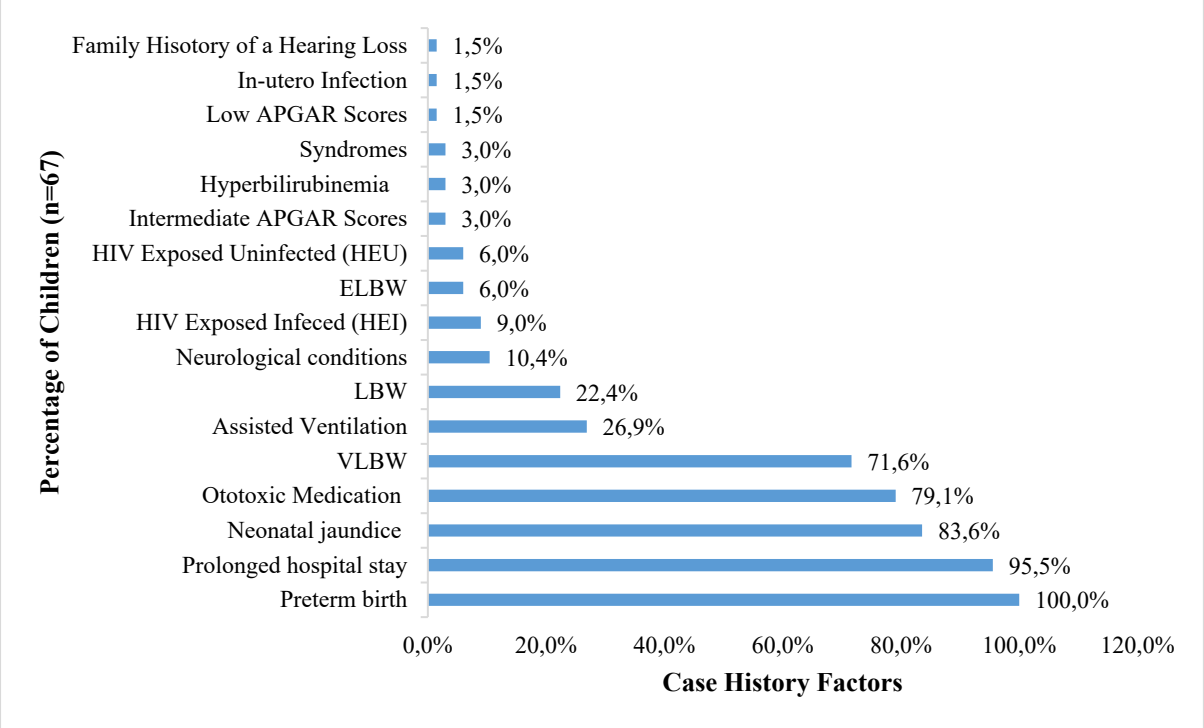

Figure 8. The Most and Least Common Case History Factors Present in the Current Study.

4.1.3.1 Gestational age. Guidelines by The American Congress of Obstetricians and Gynaecologists (ACOG) (2013) were used to categorise the gestational age of the high-risk neonates in Kanji (2016b) study. Full term was considered as 39 – 40 weeks, early term was categorized as 37 – 38 weeks, preterm was anything less than 37 weeks, and late term was 41

weeks. In the current study all 67 (100%) children were considered preterm at birth. More specifically, the mean gestational age was 31 weeks (*SD* = 1.9 and range weeks 26 – 36).

4.1.3.2 Birth weight. According to The World Health Organization (2018b) a normal birth weight is considered 2500g or more, a LBW is considered 1500 – 2499 grams, a very low birth weight (VLBW) is considered 1000 – 1499 grams and an extremely low birth weight (ELBW) is 999 grams or less. All children in the current study presented with low birth weight classes, with the majority of born with a VLBW (71.6%), followed by 15 (22.4%) who were born with a LBW. Only four (6.0%) were ELBW.

4.1.3.3 Length of hospital stay. The total length of hospital stay included time spent in the NICU, high care and/ or Kangaroo Mother Care (KMC) wards. The mean length of stay was 27.5 days (*SD* = 15.6 days, range days 8 – 87). A prolonged hospital stay for children with medical complexities is defined as 10 or more days (Gold, Hall, Shah, Thomson, Subramony, et al., 2016). Sixty-four (95.5%) children in the current study presented with a hospital stay longer than 10 days. Figure 9 displays the distribution of the length of hospital stay.

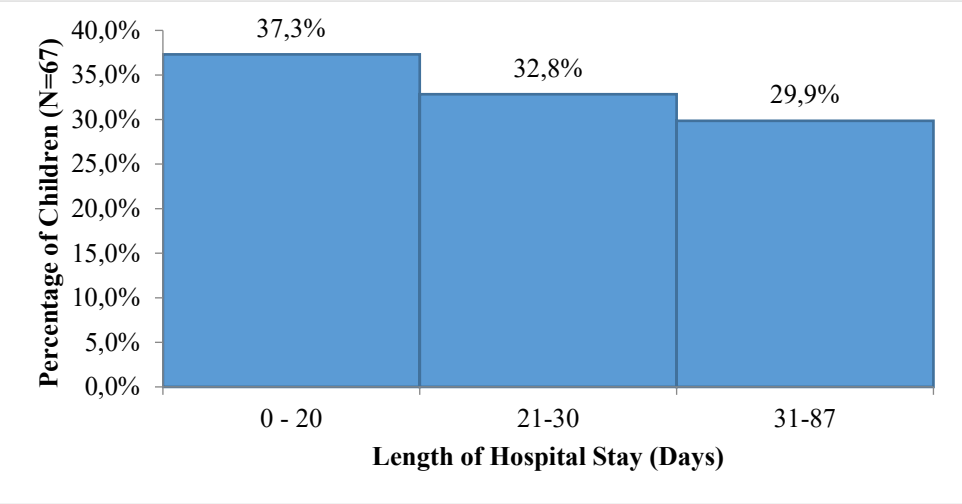

Figure 9. The Distribution of Length of Hospital Stay of the Participants' Children.

4.1.3.4 Bilirubin levels and treatment. Of the 67 children in the current study, 58 (86.6%) presented with increased bilirubin levels. Neonatal Jaundice (NNJ) refers to jaundice which lasts for 10 days with a rapid rise of serum bilirubin up to 12 mg/dL in term infants (Bujandric & Grujic, 2016). In preterm infants jaundice lasts for about two weeks, with a rapid rise of serum bilirubin up to 15 mg/dL (Bujandric & Grujic, 2016). Hyperbilirubinemia refers to the condition where the total serum bilirubin rises above the 95th percentile for one's age (Porter & Dennis, 2002).

Of the 67 children who presented with increased bilirubin levels, 56 (83.6%) presented with NNJ and of these 54 (80.6%) received PTT, and the treatment for two was unknown. Furthermore, two (3.0%) children presented with hyperbilirubinemia of which both received PTT and EBT. The number of EBT was one for both of these children.

4.1.3.5 Ototoxic medication. A large number of children in the current study, (n = 55, 82.1%) received ototoxic medication. Table 4.3 below displays the types of ototoxic medication received and the number of children who received them.

Table 4.3

The Types of Ototoxic Medication Received and the Number of Children who received them

Type of ototoxic medication	Number of Children who received the medication (n=55)
Gentamycin	31 (56.4%)
Imipenem	10 (18.2%)
Vancomycin	4 (7.3%)
Erythromycin	1 (1.8%)
Gentamycin and Vancomycin	1 (1.8%)
Gentamycin and Imipenem	2 (3.6%)
Gentamycin and Amphotericin	2 (3.6%)
Imipenem and Furosemide	1 (1.8%)
Erythromycin and Imipenem	1 (1.8%)
Gentamycin, Imipenem and Vancomycin	1 (1.8%)
Gentamycin, Imipenem and Fluromycin	1 (1.8%)

4.1.3.6 Mechanical ventilation. Case history information revealed that 18 (26.9%) of the 67 children received mechanical ventilation. On further analysis of the types of mechanical ventilation received, 17 (94.4%) received continuous positive airway pressure (CPAP), one child (5.6%) received IPPV, and two (11.1%) received both CPAP and IPPV.

The mean number of days that the children received CPAP was 17 days (SD = 1.9 days, range days 1 – 8). The mean number of days that children received IPPV was 5.2 days (SD = 1.2 days, range days 1 – 9 days). Figure 10 illustrates the number of days that ventilation was received for CPAP and IPPV respectively.

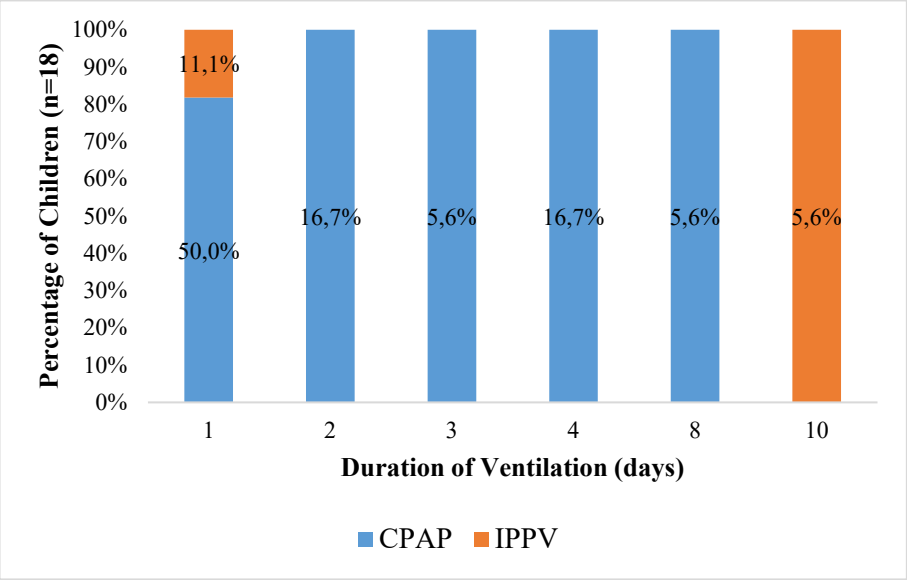

Figure 10. The Number of Days that CPAP and IPPV was received for the children who received Mechanical Ventilation.

4.1.3.7 HIV exposure. With regards to HIV exposure, the majority of the children (n = 57; 85.1%) were considered unexposed, six (9.0%) were HIE, and 4 (6.0%) were HEU. According to Garibyan and Avashia (2013) the polymerase chain reaction (PCR) technique is a simple, yet elegant, enzymatic assay, which allows for the amplification of a specific DNA fragment from a complex pool of DNA. This allows for the diagnosis of many infectious diseases. A PCR result of negative could either indicate that the pathogen is present, or not present. When not present the PCR could be presenting with a false-negative result as a result of poor DNA extraction or reaction performance. As a result, it is not certain what the true HIV status is if the four children who presented with HEU in the current study, as there was no further recent HIV testing performed according to the hospital file or reported by the caregiver.

4.1.3.8 APGAR scores. In Kanji (2016b) study, the classification of APGAR scores were determined at five and 10 minutes by the researcher and a consultant paediatrician. Scores were categorized using guidelines by The American Congress of Obstetricians and Gynaecologists (ACOG) (2006). The majority of the children (n = 64) in the current study

presented with normal APGAR scores (score between seven and ten). In addition, two (3.0%) children presented with intermediate APGAR scores (scores between four and six) and one (1.5%) child presented with low APGAR scores (between zero and three).

4.1.3.9 Neurological conditions. Seven children (10.4%) presented with neurological conditions. More specifically, one child (1.5%) presented with each of the following conditions; interventricular haemorrhage (IVH) Grade II – III, IVH Grade I on left and IVH Grade II on Right, IVH Grade IV, perinatal asphyxia, queried perinatal asphyxia, queried perinatal asphyxia due to low APGAR scores, and queried mild ventriculomegaly.

4.1.3.10 Syndromes. Two (3.0%) of the children from the risk-based newborn hearing screening study presented with a syndrome or features of a syndrome, namely Isotretinoin syndrome, and low set ears and micrognathia (small jaw).

4.1.3.11 In-utero infections. One child (1.5%) presented with an in-utero infection at birth in the current study, more specifically congenital syphilis.

4.1.3.12 Family history of hearing loss. One child (1.5%) presented with a family history of a hearing loss in the current study.

4.1.3.13 Additional medical history information. Additional case history information captured included; children presented with respiratory distress syndrome (RDS) (n = 11; 16.4%), anaemia (n = 2; 3.0%), hypoglycaemia (n = 1; 1.5%), chorioamnionitis (n = 1; 1.5%) nosocomial sepsis (n = 1; 1.5%), MRSA sepsis (n = 1; 1.5%), and apnoea (n = 1; 1.5%).

4.1.3.14 Description of the audiological history. Children of participants in the current study were previously enrolled in a risk-based newborn hearing screening study. In this study, the children underwent initial and repeat hearing screening. Screening measures included TEOAEs, DPOAEs, AABR, and high frequency tympanometry when needed. If necessary re-screening was conducted, and a diagnostic assessment was conducted for those who *referred* at the repeat hearing screening. Diagnostic assessments for those who *referred*

included ABR testing. Those who *passed* the screening were booked for VRA testing at six months corrected age.

In the current study, all 67 children attended the initial and repeat hearing screens. With regards to the initial hearing screen, 52 (77.6%) of the children *passed,* 15 (22.4%) *referred*, and one (1.5%) presented with *inconclusive* results. Results from the repeat hearing screening revealed that 60 (89.6%) of the children *passed*, six (9.0%) *referred*, and one (1.5%) had *inconclusive* results. With regards to the diagnostic assessment, 41 (61.2%) of the children attended and 26 (38.8%) did not attend. All 41 (61.2%) of the children who attended the diagnostic assessment presented with hearing within normal limits. Figure 11 below displays the results from the initial and repeat hearing screening for the children in the current study.

It must be noted that one (1.5%) child *referred* both the initial and repeat hearing screen, but did not attend the diagnostic assessment thereafter. Furthermore, one (1.5%) child *referred* the initial screen, presented with *inconclusive* results at the repeat hearing screen, but did not attend the recommended re-screen.

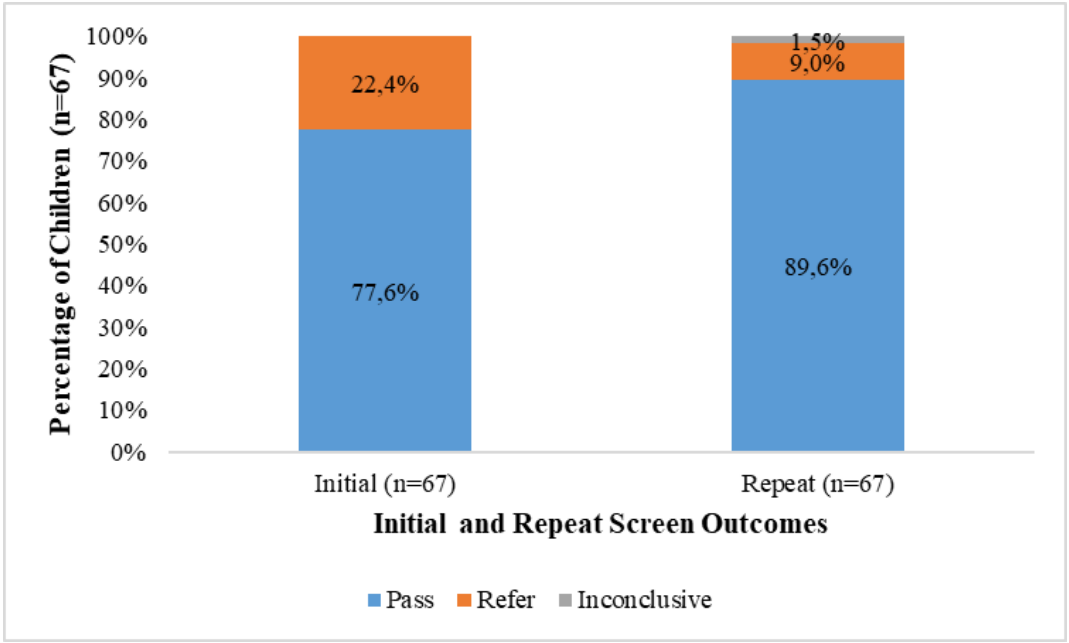

Figure 11. The Results of the Newborn Initial and Repeat Hearing Screening.

4.1.4 Description of the General Developmental Profile of the Participants' Children.

The Parents' Evaluation of Developmental Status (PEDS) measures was used to describe the general development of the children in the current study, and address the second sub-aim of the current study. In addition, of the files that were located, more recent medical concerns and developmental concerns reported from the NNFU visits, outpatient visits and re-admissions was also recorded, to gain better insight into the general development of the children.

4.1.4.1 Results obtained from the Parents' Evaluation of Developmental Status (PEDS). In terms of participants' concerns, the PEDS revealed that the most common domain that participants expressed concern in were 'other' (n = 31). This was followed by concerns in behaviour (n = 17), expressive language (n = 14), and social -emotional (n = 8) domains. The less common domains were corner arose was, gross motor (n = 7), fine motor (n = 1), self-help (n = 1) and lastly receptive language (n = 0) domains. Figure 12 below illustrates the percentage of concerns the participants expressed within each general developmental domain. Table 4.4 provides further details regarding the concerns classified under the 'other' domain.

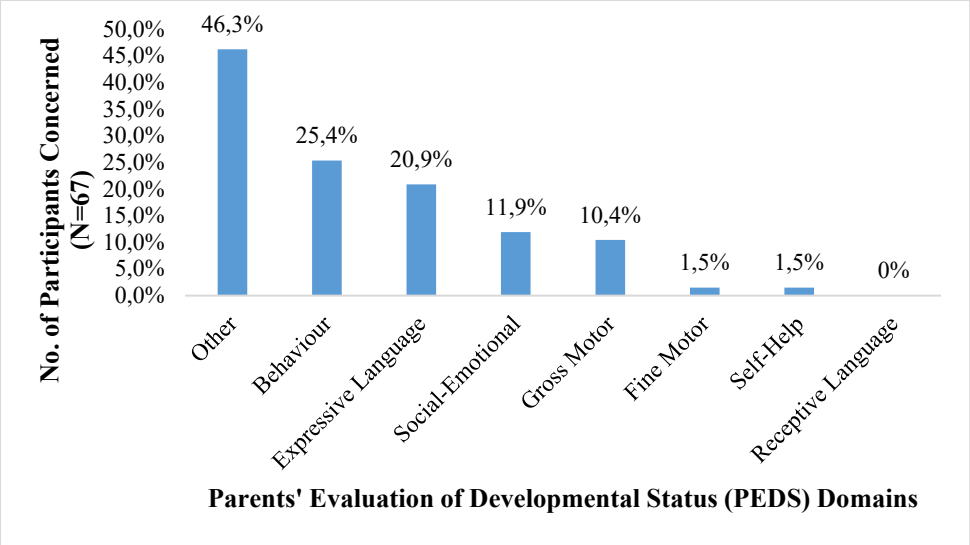

Figure 12. The Percentage of Participants who Expressed Concern within each PEDS Developmental Domain.

Table 4.4

The Details regarding the Participants' Concerns under the PEDS domain of 'other'

'Other' concerns	Number of children (n =31)
Visual/ eye concerns • Right eye pulls inwards when concentrating on an object • When looking to the side, the child presents with a squint • Both eyes constantly tear • Right eye tears	4 (22.2%)
Fussy eater	4 (22.2%)
Tonsillitis	2 (11.1%)
Unresolved allergic rhinitis	2 (11.1%)
Influenza	2 (11.1%)
Skin concerns	2 (11.1%)
Concentration concerns	2 (11.1%)
Growth concerns	2 (11.1%)
Decreased weight	2 (11.1%)
Queried sleep apnoea	1 (5.6%)
Queried HIV diagnosis	1 (5.6%)
Ring worm	1 (5.6%)
Seizures	1 (5.6%)
Vomiting	1 (5.6%)
Recurrent diarrhoea	1 (5.6%)
Cryptorchidism	1 (5.6%)
Neck problem	1 (5.6%)
Poor hair growth	1 (5.6%)

In terms of expressive language concerns, 14 (20.9%) participants expressed concern. More specifically, six (9.0%) reported concerns regarding their child communicating in short word utterances, four (6.0%) reported articulation errors, two (3.0%) reported unintelligible

speech, one (1.5%) reported stuttering concerns, and one (1.5%) reported their child to not be communicating meaningfully. With regards to gross and fine motor concerns, table 4.5 specifies further details.

Table 4.5

The Details Regarding the Participants' Concerns under the PEDS Domains of Gross and Fine Motor Skills

Motor Concerns	Number of Children (n = 8)
Gross Motor Concerns (n = 7)	
Delayed motor milestones	*1 (12.5%)*
Started walking at two and a half years old	
Gait concerns	
Toe-walking	1 (12.5%)
Right foot and arm bends outwards, and therefore she falls when walking and running	1 (12.5%)
Clumsy and frequent tripping	*1 (12.5%)*
Balance concerns	
Difficulty maintaining balance when performing a task	*1 (12.5%)*
Post-injury concerns	
Both feet previously fractured, and currently struggling to kick	*1 (12.5%)*
Right leg was previously broken and currently painful when touched	*1 (12.5%)*
Fine Motor Concerns (n = 1)	
Difficulty holding a pencil and drawing	*1 (12.5%)*

The Parents' Evaluation of Developmental Status (PEDS) provided a path indicating the level of risk children presented on based on the participants' concerns. Each path subdivides based on the constellations of concerns. Table 4.6 below indicates the pathways, interpretation, and the number and percentage of children within each pathway.

Table 4.6

The Parents' Evaluation of Developmental Status (PEDS) Pathway Results and Interpretation

Pathway	Pathway interpretation	Number of children (N = 67)
A	High risk Parents have multiple developmental and behavioural concerns	11 (16.4%)
A-1	Indicates the need for a speech-language evaluation	5 (7.5%)
A-2	Indicates the need for further testing by a developmental or school psychologist	6 (9.0%)
B	Moderate risk	23 (34.3%)
B-1	Health concerns	9 (13.4%)
B-2	Numerous behavioural concerns	14 (20.9%)
C	Elevated risk for behavioural/ mental health problems	13 (19.4%)
C-1	Children four and a half years of age and older	2 (3.0%)
C-2	Children younger than four and a half years of age	11 (16.4%)
E	Low Risk for developmental and behavioural problems	20 (29.9%)

4.1.4.2 Results obtained from the Parents' Evaluation of Developmental Status (PEDS: DM). Results from the PEDS: DM further described the general development by assessing which domains children met or unmet their milestones. With regards to the most and least common domains that milestones were unmet, fine motor milestones was the most common, (n = 46; 68.7%). This was followed by receptive language milestones which were unmet by 30 (44.8%) children, expressive language milestones which were unmet by 27 (40.3%) children, and social-emotional milestones which were unmet by 26 (38.8%) children.

Among the least common domains presenting with unmet milestones were self-help (n = 16; 23.9%) and gross motor milestones (n = 9; 15.8%).

It must be noted that when conducting the PEDS: DM, form S and R was chosen to be used by the 'PEDS online' for 10 of the children. When these forms were utilized, the gross motor milestones were not assessed. This could have affected the representation of unmet gross motor milestones in the current study.

4.1.4.3 Results obtained from Neonatal Follow-up (NNFU) Appointments. In terms of return rate to the NNFU clinics, of the 28 files that were located, 25 (89.3%) of the participants attended the NNFU appointments. The mean number of appointments attended was 4.2 (*SD* = 2.8; range of 1 – 11). Although 39 files were not located, all 39 of these participants reported to have attended the NNFU appointments. Therefore, only three (4.5%) participants in the current study are reported to have not attended the NNFU appointments.

Of the 28 hospital files that were located, 12 (42.8%) files reported medical or developmental concerns at the NNFU clinics. Of the 39 files that were not located, one (2.6%) participant reported a medical concern, and one (2.6%) participant reported a developmental concern detected at the NNFU clinics. Overall, 14 (20.9%) of the 67 children were reported to present with either a medical or developmental concern at the NNFU clinic. More specifically, nine (64.3%) medical concerns and five (35.7%) developmental concerns, of which one was speech-language related and six were general developmental delay related. Table 4.7 below displays the specific medical and developmental concerns, and the frequency and percentage of children who presented with each concern.

Table 4.7

The Medical and Developmental Concerns Detected at the Neonatal Follow-Up (NNFU) Appointments

Medical and developmental concerns	Number of children who presented with the medical condition (n = 14)
Medical concerns	
Bronchitis	1 (7.1%)
Bowel obstruction	1 (7.1%)
Dandy Walker Malformation	1 (7.1%)
Rhinitis	3 (21.4%)
Lower respiratory tract infection	1 (7.1%)
Decreased weight	2 (14.3%)
Failure to thrive	1 (7.1%)
Developmental concerns	
Developmental delay	2 (14.3%)
Delayed speech milestones	1 (7.1%)
Delayed motor milestones	1 (7.1%)

4.1.4.4 Results obtained from out-patient visits. Thirty three (49.3%) children were reported to have returned as out-patients at RMMCH. The mean number of times returned was 1.4 times (*SD* = 1.9 times, and range 1 – 11 times). Figure 13 below displays the number of times participants returned with their children as out-patients. Specific reasons as to why participants returned as out-patients is displayed in table 4.8.

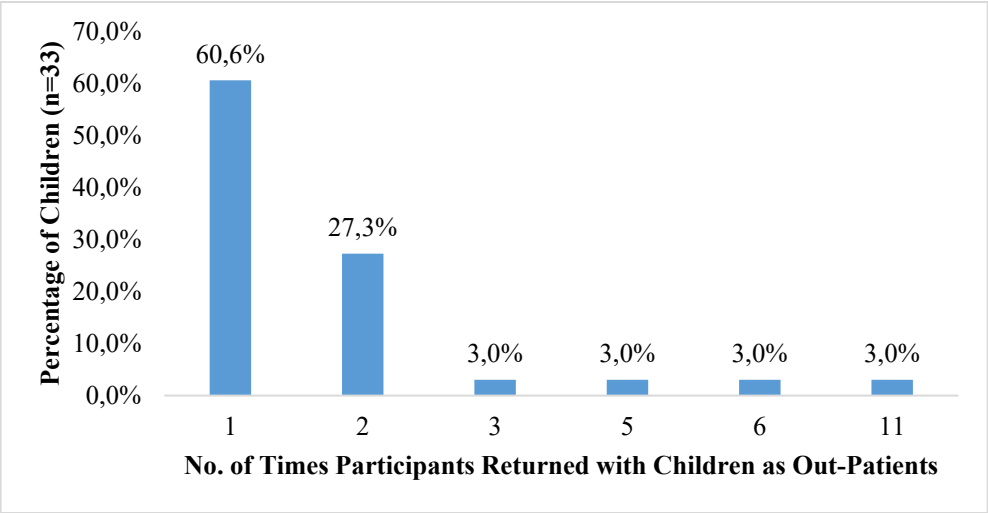

Figure 13. The number of times participants returned with their children as out-patients

Table 4.8

Reasons as to why Participants Returned as Out-Patients with their Children

Reason returned as out-patient	Number of children (n = 33)
Medical and/ or Ear, Nose and Throat (ENT) related	19 (57.6%)
Otitis media	1 (3.0%)
Tonsillitis	1 (3.0%)
Tonsillitis and lower respiratory tract infection	1 (3.0%)
Tonsillitis and upper respiratory tract infection	2 (6.0%)
Tonsillitis, upper respiratory tract infection, rhinitis, asthma and bronchopneumonia	1 (3.0%)
Tonsillitis, upper respiratory tract infection, rhinitis, inflamed adenoids, bronchopneumonia, and inflamed ear	1 (3.0%)
Lower respiratory tract infection	2 (6.0%)
Upper respiratory tract infection and constipation	1 (3.0%)
Rhinitis	1 (3.0%)
Bronchitis and asthma	1 (3.0%)
Bronchitis, acute gastroenteritis (AGE), and eczema	1 (3.0%)
Bronchitis and eczema	1 (3.0%)
Acute gastroenteritis (AGE) and phimosis	1 (3.0%
Nasal regurgitation	1 (3.0%)
Seizures	2 (6.0%)

Hand burns	1 (3.0%)
Developmental related	3 (9.1%)
Decreased weight	2 (6.0%)
Language and fine motor delay	1 (3.0%)
Musculoskeletal	3 (9.1%)
Foot fracture	1 (3.0%)
Elbow injury	1 (3.0%)
Broken leg	1 (3.0%)
Visual related	3 (9.1%)
Eyesight concern	1 (3.0%)
Pink eye	1 (3.0%)
Eye tearing	1 (3.0%)
Medical and developmental related	1 (3.0%)
Tonsillitis, sleep apnoea and developmental delay	1 (3.0%)
Medical, motor, diet and visual related	1 (3.0%)
Otitis media, bronchitis, AGE, toe walking, and squint	1 (3.0%)
Developmental and visual related	1 (3.0%)
Conjunctivitis and developmental delay	1 (3.0%)
Other	2 (6.1%)
Dysphagia	1 (3.0%)
Sexual abuse	1 (3.0%)

4.1.4.5 Results obtained from re-admissions. Twenty one (31.3%) of the 67 children were reported to have been re-admitted to hospital. More specifically, 13 (61.9%) of the hospital files located reported re-admissions, and 8 (38.1%) participants whose children's hospital files were not located, reported their children to have been re-admitted.

In terms of number of re-admissions, 16 (76.2%) children were re-admitted once and five (23.8%) were re-admitted twice. Table 4.9 below specifies the reasons and the number of children re-admitted in the current study.

Table 4.9

Reasons as to why Children were Re-Admitted

Reason for readmission	Number of children (n = 21)
One re-admission	
Bronchitis	5 (23.8%)
Apnoeas	1 (4.8%)
Bowel obstruction	2 (9.5%)
Lower respiratory tract infection	1 (4.8%)
Febrile seizures	1 (4.8%)
Abdominal distention	1 (4.8%)
Diarrhoea	1 (4.8%)
Eye operation for bilateral squint	1 (4.8%)
Stomach hernia repaired	1 (4.8%)
Elbow operation	1 (4.8%)
Finger re-attached	1 (4.8%)
Two re-admissions	
Neonatal sepsis and pericardial effusion	1 (4.8%)
Apnoeas and neonatal sepsis	1 (4.8%)
Upper respiratory tract infection and bronchospasm	1 (4.8%)
Apnoeas and vomiting	2 (9.5%)

4.1.5 Description of the Audiological Development of the Participants' Children. In order to address the third sub-aim of the current study, descriptive statistics was used to analyse results from the hearing and communicative checklist. Of the 67 participants, 12 (17.9%) expressed audiological concern. Reasons for concern included; listening difficulties, especially when occupied with another task (n = 3), previous ear trauma (n = 2), ignoring others (n = 2), delayed speech and complaints of itchy ears (n = 1), lump behind ear (n = 1), questionable ear tag (n = 1), questionable grommets still in situ (n = 1), and excessive cerumen (n = 1).

Furthermore, three (25.0%) of the 12 children whose caregivers expressed audiological concern did not meet their auditory milestones according to the hearing and communicative checklist. These participants' concerns included; their child ignoring others (n = 2), and one participant reported delayed speech and a complaint of itchy ears. It must be noted that one child whose caregiver didn't express audiological concern also did not meet her audiological milestones. As a result, four (6.0%) of the 67 children in the current study didn't meet their auditory milestones according to the hearing and communicative checklist.

All participants, whether having expressed audiological concern or not, did not seek further audiological testing after the newborn risk-based hearing screening and/ or diagnostic assessment that formed part of the study by Kanji (2016b). Three of the 12 caregivers reported to have not sought further audiological assistance as they assumed the hearing was fine based on the newborn hearing screening results. Five of the 12 caregivers who expressed concern did not report a reason for not seeking further audiological assistance.

4.1.6 The Relationship between Case History Factors at Birth, and the Current General and Audiological Development of the Participants' Children.

In order to address the fourth sub-aim of the current study, the Fisher's exact test was performed to determine if a relationship exists between the case history factors at birth and the general development (results from the PEDS and PEDS: DM measures), and the audiological development (results from the hearing and communicative checklist). In cases where the sample was small, statistically significant relationships could not be established. In these cases, significant findings that the researcher observed will still be reported on. Lastly, the combination of case history factors associated with auditory milestones not being met was analysed.

When analysing the relationships between case history factors and PEDS pathways, statistically significant relationships were established between bilirubin treatment and PEDS

pathways ($p = 0.048$), and ototoxicity and PEDS pathways ($p = 0.008$). Observed results revealed a large proportion from of children considered VLBW presented on path A (high risk for developmental delay) (n = 9; 81.8%). Both children presenting with hyperbilirubinemia requiring PTT and EBT also presented on path A. Table 4.10 displays the case history factors, and the number of children presenting on each PEDS pathway. Significant values obtained from the Fisher's exact test are also displayed.

Analysis of the relationships between case history factors and the gross and fine motor milestones met and unmet according to the PEDS: DM was then analysed. The sample size of the gross motor milestones assessed was 57 and not 67, due to certain forms been used by the 'PEDS online', as mentioned previously. Results revealed a statistically significant relationship between APGAR scores and gross motor milestones ($p = 0.002$). Observed results revealed children considered VLBW are more likely not meet their gross motor milestones (n = 7; 77.8%). Both children presenting with hyperbilirubinemia requiring PTT and EBT did not meet their fine motor milestones. A large proportion of children who received ototoxic medication did also not meet their fine motor milestones (n = 35; 76.1%). Table 4.11 displays the case history factors and the number of children who met and did not meet their fine and gross motor milestones according to the PEDS: DM. Significant values obtained from the Fisher's exact test are also displayed.

When analysing the relationships between case history factors and the receptive and expressive milestones, results revealed a statistically significant relationship between length of hospital stay and receptive language milestones ($p = 0.039$). Observed results revealed a large proportion of children presenting with NNJ did not meet their receptive language milestones (n = 25; 92.6%) and expressive language milestones (n = 21; 100%). Both children presenting with hyperbilirubinemia requiring PTT and EBT did not meet their receptive language milestones. A large proportion of children exposed to ototoxic medication

did not meet both their receptive language milestones (n = 24; 80.0%) and expressive language milestones (n = 22; 81.5%). Table 4.12 displays the number of children who met and did not meet these milestones, as well as the significant values obtained from the Fisher's exact test.

When analysing the relationships between case history factors and the self-help and social emotional milestones, results revealed a statistically significant relationship between ventilation and social-emotional milestones ($p = 0.025$). Table 4.13 displays the number of children who met and did not meet these milestones, as well as the significant values obtained from the Fisher's exact test.

The relationship between caregiver audiological concern, and auditory milestones met and unmet were also analysed. The small sample size of children who did not meet their auditory milestones did not allow for statistically significant relationships to be established. Observed results revealed children presenting with VLBW were more likely to present with caregiver concern (n = 10; 83.3%) and children presenting with NNJ (n = 9; 100%). In addition, all four children who did not meet their auditory milestones presented with NNJ and received ototoxic medication. A large proportion of the children who did not meet their auditory milestones presented with VLBW (n = 3; 75.0%) and received PTT (n = 3; 5.4%). Table 4.14 displays the number of children who presented with caregiver audiological concern, the number of children who did not meet their auditory milestones. Significant values obtained from the Fisher's exact test are also displayed.

Table 4.10

The Number and Percentage of Children Presenting with certain Case History Factors and Presenting on each PEDS pathways, and the Significant Values obtained from the Fisher's Exact Test

Case History Factor	No. on Path A (High Risk) (%)	No. on Path B (Moderate Risk) (%)	No. on Path C (Elevated Risk) (%)	No. on Path E (low Risk) (%)	*df*	Fisher's Exact Test *p*-Value
Birth Weight					6	0.878
LBW (n = 15)	2 (18.2%)	5 (21.7%)	3 (23.1%)	5 (25.0%)		
VLBW (n = 48)	9 (81.8%)	17 (73.9%)	8 (61.5%)	14 (70.0%)		
ELBW (n = 4)	0 (0%)	1 (4.3%)	2 (15.4%)	1 (5.0%)		
Total (N = 67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		
Length of Hospital Stay					6	0.888
8 – 20 days (n = 25)	3 (27.3%)	8 (34.8%)	6 (46.2%)	8 (40.0%)		
21 – 30 days (n = 22)	3 (27.3%)	9 (39.1%)	3 (23.1%)	7 (35.0%)		
31 – 87 days (n = 20)	5 (45.5%)	6 (26.1%)	4 (30.8%)	5 (25.0%)		
Total (N=67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		
Bilirubin Level					3	0.057
Neonatal Jaundice (NNJ) (n = 56)	8 (80.0%)	21 (100%)	10 (100.0%)	17 (100%)		
Hyperbilirubinemia (n = 2)	2 (20.0%)	0 (0%)	0 (0%)	0 (0%)		
Total (N = 58)	*10 (100%)*	*21 (100%)*	*10 (100%)*	*17 (100%)*		
Bilirubin Treatment					3	0.048
Phototherapy Treatment (PTT) (n = 54)	7 (77.8%)	21 (100%)	9 (100%)	17 (100%)		
Phototherapy and exchange blood transfusion (EBT) (n = 2)	2 (22.2%)	0 (0%)	0 (0%)	0 (0%)		
Total (N = 56)	*9 (100%)*	*21 (100%)*	*9 (100%)*	*17 (100%)*		

Ototoxic Medication					3	0.008
Received medication (n = 55)	8 (72.7%)	23 (100%)	8 (61.5%)	16 (80.0%)		
Did not receive medication (n = 12)	3 (27.3%)	0 (0%)	5 (38.5%)	4 (20.0%)		
Total (N = 67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		
Mechanical Ventilation					2	0.538
Received (n = 18)	3 (27.3%)	8 (34.8%)	4 (30.8%)	3 (15.0%)		
Did not receive (n = 49)	8 (72.7%)	15 (65.2%)	9 (69.2%)	17 (85.0%)		
Total (N = 67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		
HIV Status					6	0.261
Unexposed (n = 57)	8 (72.7%)	21 (91.3%)	10 (76.9%)	18 (90.0%)		
Exposed Infected (n = 6)	1 (9.1%)	1 (4.3%)	3 (23.1%)	1 (5.0%)		
Exposed Uninfected (n = 4)	2 (18.2%)	1 (4.3%)	0 (0%)	1 (5.0%)		
Total (N = 67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		
APGAR Scores					6	0.337
Normal (n = 64)	11 (100%)	21 (91.3%)	12 (92.3%)	20 (100%)		
Intermediate (n = 2)	0 (0%)	2 (8.7%)	0 (0%)	0 (0%)		
Low (n = 1)	0 (0%)	0 (0%)	1 (7.7%)	0 (0%)		
Total (N =67)	*11 (100%)*	*23 (100%)*	*13 (100%)*	*20 (100%)*		

Table 4.11

The Number and Percentage of Children who presented with Certain Case History Factors, who met and did Not Meet their Fine and Gross Motor Milestones according to the PEDS: DM, and the Significant values obtained from the Fisher's Exact Test

	Fine Motor Milestones				Gross Motor Milestones			
Case History Factor	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value
Birth Weight			2	0.218			2	0.585
LBW (n = 15)	2 (9.5%)	13 (28.3%)			13 (27.1%)	1 (11.1%)		
VLBW (n = 48)	18 (85.7%)	30 (65.2%)			32 (66.7%)	7 (77.8%)		
ELBW (n = 4)	1 (4.8%)	3 (6.5%)			3 (6.3%)	1 (11.1%)		
Total (N = 67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		
Length of Hospital Stay			2	0.091			2	0.216
8 – 20 days (n = 25)	4 (19%)	21 (45.7%)			19 (39.6%)	4 (44.4%)		
21 – 30 days (n = 22)	10 (47.6%)	12 (26.1%)			18 (37.5%)	1 (11.1%)		
31 – 87 days (n = 20)	7 (33.3%)	13 (28.3%)			11 (22.9%)	4 (44.4%)		
Total (N=67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		
Bilirubin Level (n=58)			1	0.540			1	1.000
Neonatal Jaundice (NNJ) (n = 56)	20 (100%)	36 (94.7%)			40 (95.2%)	6 (100%)		
Hyperbilirubinemia (n = 2)	0 (0%)	2 (5.3%)			2 (4.8%)	0 (0%)		
Total (N = 58)	*20 (100%)*	*38 (100%)*			*42 (100%)*	*6 (100%)*		
Bilirubin Treatment			1	0.544			1	1.000
Phototherapy Treatment (PTT) (n = 54)	19 (100%)	35 (94.6%)			40 (95.2%)	6 (100%)		
Phototherapy and exchange blood transfusion (EBT) (n = 2)	0 (0%)	2 (5.4%)			2 (4.8%)	0 (0%)		

Total (N = 56)	*19 (100%)*	*37 (100%)*			*42 (100%)*	*6 (100%)*		
Ototoxic Medication			1	0.086			1	1.000
Received medication (n = 55)	20 (95.2%)	35 (76.1%)			38 (79.2%)	7 (77.8%)		
Did not receive medication (n = 12)	1 (4.8%)	11 (23.9%)			10 (20.8%)	2 (22.2%)		
Total (N = 67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		
Mechanical Ventilation			1	0.701			1	0.202
Received (n = 18)	5 (23.8%)	13 (28.3%)			10 (20.8%)	4 (44.4%)		
Did not receive (n = 49)	16 (76.2%)	33 (71.7%)			38 (79.2%)	5 (55.6%)		
Total (N = 67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		
HIV Status			2	0.471			2	0.171
Unexposed (n = 57)	19 (90.5%)	38 (82.6%)			43 (89.6%)	7 (77.8%)		
Exposed Infected (n = 6)	2 (9.5%)	4 (8.7%)			3 (6.3%)	0 (0%)		
Exposed Uninfected (n = 4)	0 (0%)	4 (8.7%)			2 (4.2%)	2 (22.2%)		
Total (N = 67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		
APGAR Scores			2	0.683			2	0.002
Normal (n = 64)	20 (95.2%)	44 (95.7%))			48 (100%)	6 (66.7%)		
Intermediate (n = 2)	0 (0%)	1 (2.2%)			0 (0%)	2 (22.2%)		
Low (n = 1)	0 (0%)	1 (2.2%)			0 (0%)	1 (1.8%)		
Total (N = 67)	*21 (100%)*	*46 (100%)*			*48 (100%)*	*9 (100%)*		

Table 4.12

The Number and Percentage of Children who presented with Certain Case History Factors, who met and did Not Meet their Receptive Language and Expressive Language Milestones according to the PEDS: DM, and the Significant Values obtained from the Fisher's Exact Test

	Receptive Language Milestones				Expressive Language Milestones			
Case History Factor	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value
Birth Weight			2	0.528			2	0.550
LBW (n = 15)	9 (24.3%)	6 (20.0%)			7 (17.5%)	8 (29.6%)		
VLBW (n = 48)	27 (73.0%)	21 (70.0%)			30 (75.0%)	18 (66.7%)		
ELBW (n = 4)	1 (2.7%)	3 (10.0%)			3 (7.5%)	1 (3.7%)		
Total (N = 67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		
Length of Hospital Stay			2	0.039			2	0.153
8 – 20 days (n = 25)	12 (32.4%)	13 (43.3%)			11 (27.5%)	14 (51.9%)		
21 – 30 days (n = 22)	17 (45.9%)	5 (16.7%)			15 (37.5%)	7 (25.9%)		
31 – 87 days (n = 20)	8 (21.6%)	12 (40.0%)			14 (35.0%)	6 (22.2%)		
Total (N=67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		
Bilirubin Level (n=58)			1	0.120			1	0.530
Neonatal Jaundice (NNJ) (n = 56)	31 (100%)	25 (92.6%)			35 (94.6%)	21 (100%)		
Hyperbilirubinemia (n = 2)	0 (0%)	2 (7.4%)			2 (5.4%)	0 (0%)		
Total (N = 58)	*31 (100%)*	*27 (100%)*			*37 (100%)*	*21 (100%)*		
Bilirubin Treatment			1	0.211			1	0.532
Phototherapy Treatment (PTT) (n = 54)	30 (100%)	24 (92.3%)			34 (94.4%)	20 (100%)		
Phototherapy and exchange blood transfusion (EBT) (n = 2)	0 (0%)	2 (7.7%)			2 (5.6%)	0 (0%)		

Total (N = 56)	*30 (100%)*	*26 (100%)*			*36 (100%)*	*20 (100%)*		
Ototoxic Medication			1	0.755			1	1.000
Received medication (n = 55)	31 (83.8%)	24 (80.0%)			33 (82.5%)	22 (81.5%)		
Did not receive medication (n = 12)	6 (16.2%)	6 (20.0%)			7 (17.5%)	5 (18.5%)		
Total (N = 67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		
Mechanical Ventilation			1	0.406			1	0.781
Received (n = 18)	8 (21.6%)	10 (33.3%)			10 (25.0%)	8 (29.6%)		
Did not receive (n = 49)	29 (78.4%)	20 (66.7%)			30 (75.0%)	19 (70.4%)		
Total (N = 67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		
HIV Status			2	0.078			2	0.291
Unexposed (n = 57)	34 (91.9%)	23 (76.7%)			36 (90.0%)	21 (77.8%)		
Exposed Infected (n = 6)	3 (8.1%)	3 (10%)			3 (7.5%)	3 (11.1%)		
Exposed Uninfected (n = 4)	0 (0%)	4 (13.3%)			1 (2.5%)	3 (11.1%)		
Total (N = 67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		
APGAR Scores			2	1.000			2	0.491
Normal (n = 64)	35 (94.6%)	29 (96.7%)			39 (97.5%)	25 (92.6%)		
Intermediate (n = 2)	1 (2.7%)	1 (3.3%)			1 (2.5%)	2 (3%)		
Low (n = 1)	1 (2.7%)	0 (0%)			0 (0%)	1 (3.7%)		
Total (N = 67)	*37 (100%)*	*30 (100%)*			*40 (100%)*	*27 (100%)*		

Table 4.13

The Number and Percentage of Children who presented with Certain Case History Factors, who met and did Not Meet their Self-Help and Social-Emotional Milestones according to the PEDS: DM, and the Significant Values obtained from the Fisher's Exact Test

	Self-Help Milestones				Social-Emotional Milestones			
Case History Factor	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value	No. who met milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value
Birth Weight			2	0.357			2	0.835
LBW (n = 15)	11 (21.6%)	4 (25)			10 (25.0%)	5 (19.2%)		
VLBW (n = 48)	38 (74.5%)	10 (62.5)			28 (70.0%)	19 (73.1%)		
ELBW (n = 4)	2 (3.9%)	2 (12.5)			2 (5.0%)	2 (7.7%)		
Total (N = 67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		
Length of Hospital Stay			2	0.937			2	0.858
8 – 20 days (n = 25)	19 (37.3%)	6 (37.5%)			14 (35.0%)	11 (42.3%)		
21 – 30 days (n = 22)	16 (31.4%)	6 (37.5%)			14 (35.0%)	8 (20.8%)		
31 – 87 days (n = 20)	16 (31.4%)	4 (25.0%)			12 (30.0%)	7 (26.9%)		
Total (N=67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		
Bilirubin Level			1	0.374			1	1.000
Neonatal Jaundice (NNJ) (n = 56)	45 (97.8%)	11 (91.7%)			32 (97.0%)	23 (95.8%)		
Hyperbilirubinemia (n = 2)	1 (2.2%)	1 (8.3%)			1 (3.0%)	1 (4.2%)		
Total (N = 58)	*46 (100%)*	*12 (100%)*			*33 (100%)*	*24 (100%)*		
Bilirubin Treatment			1	0.386			1	1.000
Phototherapy Treatment (PTT) (n = 54)	43 (97.7%)	11 (91.7%)			31 (96.9%)	22 (95.7%)		
Phototherapy and exchange blood transfusion (EBT) (n = 2)	1 (2.3%)	1 (8.3%)			1 (3.1%)	1 (4.3%)		

Total (N = 56)	*44 (100%)*	*12 (100%)*			*32 (100%)*	*23 (100%)*		
Ototoxic Medication			1	0.140			1	0.517
Received medication (n = 55)	44 (86.3%)	11 (68.8%)			34 (85.0%)	20 (76.9%)		
Did not receive medication (n = 12)	7 (13.7%)	5 (31.3%)			6 (15.0%)	6 (23.1%)		
Total (N = 67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		
Mechanical Ventilation			1	1.000			1	0.025
Received (n = 18)	14 (27.5%)	4 (25.0%)			15 (37.5%)	3 (11.5%)		
Did not receive (n = 49)	37 (72.5%)	12 (75.0%)			25 (62.5%)	23 (88.5%)		
Total (N = 67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		
HIV Status			2	1.000			2	0.662
Unexposed (n = 57)	43 (83.3%)	14 (87.5%)			35 (87.5%)	21 (80.8%)		
Exposed Infected (n = 6)	5 (9.8%)	1 (6.3%)			3 (7.5%)	3 (11.5%)		
Exposed Uninfected (n = 4)	3 (5.9%)	1 (6.3%)			2 (5.0%)	2 (7.7%)		
Total (N = 67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		
APGAR Scores			2	0.142			2	0.704
Normal (n = 64)	50 (98%)	14 (87.5%)			37 (92.5%)	26 (100%)		
Intermediate (n = 2)	1 (2%)	1 (6.3%)			2 (5.0%)	0 (0%)		
Low (n = 1)	0 (0%)	1 (6.3%)			1 (2.5%)	0 (0%)		
Total (N = 67)	*51 (100%)*	*16 (100%)*			*40 (100%)*	*26 (100%)*		

Table 4.14

The Number and Percentage of Children who presented with Certain Case History Factors, who presented with Audiological Concern, who did not meet their Audiological Milestones, and the Significant Values obtained from the Fisher's Exact Test

Case History Factor	No. who presented with audiological concern (%)	No. who did not present with audiological concern (%)	*df*	Fisher's Exact Test *p*-value	No. who did meet milestones (%)	No. who didn't meet milestones (%)	*df*	Fisher's Exact Test *p*-value
Birth Weight			2	0.753			2	1.000
LBW (n = 15)	2 (16.7%)	13 (23.6%)			14 (22.2%)	1 (25.0%)		
VLBW (n = 48)	10 (83.3%)	38 (69.1%)			45 (71.4%)	3 (75.0%)		
ELBW (n = 4)	0 (0%)	4 (7.3%)			4 (6.3%)	0 (0%)		
Total (N = 67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		
Length of Hospital Stay			2	0.150			2	1.000
8 – 20 days (n = 25)	3 (25.0%)	22 (40.0%)			23 (36.5%)	2 (50.0%)		
21 – 30 days (n = 22)	7 (58.3%)	15 (27.3%)			21 (33.3%)	1 (25.0%)		
31 – 87 days (n = 20)	2 (16.7%)	18 (32.7%)			19 (30.2%)	1 (25.0%)		
Total (N=67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		
Bilirubin Level			1	1.000			1	1.000
Neonatal Jaundice (NNJ) (n = 56)	9 (100%)	47 (95.9%)			52 (96.3%)	4 (100%)		
Hyperbilirubinemia (n = 2)	0 (0%)	2 (4.1%)			2 (3.7%)	0 (0%)		
Total (N = 58)	*9 (100%)*	*49 (100%)*			*54 (100%)*	*4 (100%)*		
Bilirubin Treatment			1	1.000			1	1.000
Phototherapy Treatment (PTT) (n = 54)	8 (100%)	46 (95.8%)			51 (96.2%)	3 (5.4%)		
Phototherapy and exchange	0 (0%)	2 (4.2%)			2 (3.8%)	0 (0%)		

blood transfusion (EBT) (n = 2)								
Total (N = 56)	*8 (100%)*	*48 (100%)*			*53 (100%)*	*3 (100%)*		
Ototoxic Medication			1	0.678			1	1.000
Received medication (n = 55)	11 (91.7%)	44 (80.0%)			51 (81.0%)	4 (100%)		
Did not receive medication (n = 12)	1 (8.3%)	11 (20.0%)			12 (19.0%)	0 (0)		
Total (N = 67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		
Mechanical Ventilation			1	0.158			1	1.000
Received (n = 18)	1 (8.3%)	17 (30.9%)			17 (27.0%)	1 (25.0%)		
Did not receive (n = 49)	11 (91.7%)	38 (69.1%)			46 (73.0%)	3 (75.0%)		
Total (N = 67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		
HIV Status			2	1.000			2	0.103
Unexposed (n = 57)	11 (91.7%)	46 (83.6%)			55 (87.3%)	2 (50.0%)		
Exposed Infected (n = 6)	1 (8.3%)	5 (9.1%)			5 (7.9%)	1 (25.0%)		
Exposed Uninfected (n = 4)	0 (0%)	4 (7.3%)			3 (4.8%)	1 (25.0%)		
Total (N = 67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		
APGAR Scores			2	1.000			2	1.000
Normal (n = 64)	12 (100%)	52 (94.5%)			60 (95.2%)	4 (100%)		
Intermediate (n = 2)	0 (0%)	2 (3.6%)			2 (3.2%)	0 (0%)		
Low (n = 1)	0 (0%)	1 (1.8%)			1 (1.6%)	0 (0%)		
Total (N = 67)	*12 (100%)*	*55 (100%)*			*63 (100%)*	*4 (100%)*		

4.1.6.1 Neurological conditions. The relationship between neurological conditions and current study measures could not be determined, due to the small sample size. Seven children presented with neurological conditions in the current study. Table 4.15 displays the distribution of the children who presented with neurological conditions and results from the PEDS pathways. Table 4.16 displays the number of children presenting with neurological conditions and the milestones met and unmet according to the PEDS: DM.

Table 4.15

The Number and Percentage of Children presenting with Neurological Conditions at birth and the Pathway they presented on according to the PEDS Measure

Neurological Conditions	Pathway (n = 7)		
	A (n = 2)	B (n = 4)	E (n = 1)
IVH Grade II – III	0 (0%)	1 (25.0%)	0 (0%)
IVH Grade I on left, and IVH Grade II on Right	1 (50.0%)	0 (0%)	0 (0%)
IVH Grade IV	1 (50.0%)	0 (0%)	0 (0%)
Perinatal asphyxia	0 (0%)	1 (25.0%)	0 (0%)
Queried perinatal asphyxia	0 (0%)	1 (25.0%)	0 (0%)
Queried perinatal asphyxia due to low APGAR scores	0 (0%)	1 (25.0%)	0 (0%)
Queried mild ventriculomegaly	0 (0%)	0 (0%)	1 (100%)

Table 4.16

The Number and Percentage of Children who presented with Neurological Conditions at birth, and who met and did Not Meet their milestones according to the PEDS: DM

General developmental domain	No. of Children (n = 7)
Fine Motor Milestones	
Milestones met	2 (28.6%)
Milestones not met	5 (71.4%)
Gross Motor Milestones	
Milestones met	4 (57.1%)
Milestones not met	3 (42.9%)
Receptive Language Milestones	
Milestones met	3 (42.9%)
Milestones not met	4 (57.1%)
Expressive Language Milestones	
Milestones met	4 (57.1%)
Milestones not met	3 (42.9%)
Self-Help Milestones	
Milestones met	6 (85.7%)
Milestones not met	1 (14.3%)
Social-Emotional Milestones	
Milestones met	6 (85.7%)
Milestones not met	1 (14.3%)

The hearing and communicative checklist revealed that none of the participants whose children presented with neurological conditions voiced audiological concern. Furthermore, all of the children met their auditory milestones.

4.1.6.2 Syndromes. On further analysis of the child presenting with Isotretinoin syndrome, the PEDS revealed her to present on path A (high risk for developmental delay). Furthermore, the PEDS: DM revealed that this child did meet her receptive language

milestones and did not meet the following; fine motor, gross motor, expressive language, self-help and social-emotional milestones. The hearing and communicative checklist revealed that the caregiver communicated audiological concern; querying whether grommets were still in situ in one of her child's ears. The child met her auditory milestones for her age.

The child who presented with low set ears and micrognathia, presented on path E (low risk for developmental delay) on the PEDS, and did not meet social-emotional milestones according to the PEDS: DM. The hearing and communicative checklist further revealed that the caregiver did not present with audiological concern and that the child did meet her auditory milestones.

4.1.6.3 In-Utero infections. It was observed that the child who presented with congenital syphilis presented on path E on the PEDS (low risk of developmental delay), and did not meet expressive language milestones according to the PEDS: DM. The hearing and communicative checklist further revealed that the caregiver did not present with any audiological concern and the audiological milestones were met for this child.

4.1.6.4 Family history of hearing loss. The one child who presented with a family history of hearing loss presented on path C (elevated risk for developmental delay) on the PEDS measure, and all milestones were met. According to the hearing and communicative checklist, the caregiver did not present with audiological concern and the child met the auditory milestones for her age.

4.1.6.5 Combination of case history factors of children who did not meet their auditory milestones. The combination of case history factors of the four children who did not meet their auditory milestones were analysed and is displayed in table 4.17 below.

Table 4.17

The Case History Factors of the Children who did not meet their Auditory Milestones

Case History Factor	Child 1	Child 2	Child 3	Child 4
Gestational Age (Weeks)	31	30	28	31
Birth Weight	LBW	VLBW	VLBW	VLBW
APGAR Score	Normal	Normal	Normal	Normal
Length of Hospital Stay (Days)	20	16	34	30
Bilirubin Outcome and Treatment	NNJ	NNJ PTT	NNJ PTT	NNJ PTT
HIV Status	Exposed Infected	Exposed Uninfected	Unexposed	Unexposed
Ventilation and Treatment	No	No	Yes CPAP	No
Ototoxic Medication	No	Yes Gentamycin	Yes Gentamycin Vancomycin	Yes Gentamycin
Additional Medical History	No	RDS	RDS	No
Audiological History	*Passed* the initial and repeat hearing screening	*Passed* the initial hearing screen and *referred* repeat hearing screen	*Passed* the initial and repeat hearing screen	*Referred* the initial and repeat hearing screen

As mentioned previously, the small sample size of children who did not meet their auditory milestones did not allow for statistically significant relationships to be established between individual case history factors. Nevertheless, observing the case history factors of these children revealed all of them presented with; preterm birth, low birth weight classes,

normal APGAR scores, prolonged hospital stay and NNJ. A large proportion of children received PTT (n = 3), ototoxic medication (n = 3), HIV status of HEI or HEU (n = 2), ventilation (n = 2) and RDS (n = 2). These children most commonly presented with an overall result of *pass* at the newborn hearing screening (n = 3) and one child *referred* both the initial and repeat hearing screen.

In this chapter, results for case history data; description of the general development profile; audiological development; and the relationship between case history factors and the current general and audiological development of the participants' children were presented. The most frequently occurring case history factors in the current study sample included; preterm birth, low birth weight classes, prolonged hospital stay, increased bilirubin levels, ototoxic medication and mechanical ventilation. Results from PEDS revealed caregivers most commonly expressed concerns relating to behaviour, expressive language, and social-emotional development. According to the PEDS:DM a significant proportion of children did not meet their fine motor, receptive language, and expressive language milestones. Statistically significant relationships were established between the PEDS measure and bilirubin treatment ($p=0.048$), and ototoxicity ($p=0.008$). In addition, the relationship between APGAR scores and gross motor milestones ($p=0.002$), and length of hospital stay and receptive language milestones ($p=0.039$). Of the four children who did not meet their audiological milestones, all presented with one or more of the following case history factors; preterm birth, low birth weight classes, prolonged hospital stay, neonatal jaundice mostly requiring phototherapy treatment, HIV exposed infected/ exposed uninfected, mechanical ventilation, ototoxic medication and respiratory distress syndrome. These findings will be discussed in the next chapter.

Chapter 5: Discussion

5.1. Introduction

This chapter presents a discussion of the results in accordance with the sub-aims of the current study. Relevant literature will be incorporated into the discussion in order to validate the results, and to possibly inform risk-based general developmental monitoring and audiological surveillance within the South African context.

Prior to the discussion of the findings, it must be noted that caregivers' involvement in developmental and audiological monitoring is crucial. The HPCSA (2018) highlights the caregivers' important role by assigning them with the responsibility of monitoring their children, especially when their child presents with a risk-factor for delayed-onset hearing loss. Risk-based monitoring is however only one component for effective early intervention, the second component includes follow-up return (Schoeman & van der Linde, 2017). One hundred and twenty (64.2%) caregivers contacted did not participate in the current study. The most significant reason for poor follow-up was that caregivers' contact details had changed. Various other factors influencing follow-up return is reported in the literature, for example, level of caregiver education, proximity from the intervention centre (De Souza, Sardessai, Joshi, Joshi & Hughes, 2006), and as social-economic status (Kanji & Krabbenhoft, 2018). Hence, caregivers' demographic details may be associated with follow-up and it is therefore important that the profile of the participants in the current study are discussed.

5.2. Demographic Characteristics

5.2.1 Demographic characteristics of caregivers. With regards to age and gender, current findings revealed that 64 of the 67 participants were mothers, and participants were mostly between the ages of 30 – 39 years, with the mean age being 34.6 years. Statistical data reports that the majority of mothers giving birth in South Africa are between the ages 20 to

34 years of age (Statistics South Africa, 2017). Considering that the mean chronological age of the children in the current study was 4 years 1 month, the age of the participants in the current study is in agreement with the statistical data. On further analysis of participant gender, it is suggested that within dual-earning families, the father's interest in their child's well-being is in the form of responsibility or accessibility, which is seen as less time consuming (Kotila, Schoppe- Sullivan & Kamp Dush, 2013). The mother's role was reported to be more demanding, as they serve as the primary care provider and therefore this requires more time. The current study supports this claim as mentioned previously, as significantly larger proportion of mothers attended the developmental screening and audiological surveillance appointment for the purposes of the current study.

As far as participant race was concerned, the current study revealed that the vast majority of participants and children were Black African. This is a true representation of the South African population as Statistics South Africa (2018), reports that the largest race group in South Africa are Black African (46.7 million). Furthermore, the Coloured population is the second largest (5.1 million) followed by the White population (4.5 million), and lastly the Indian/Asian population (1.4 million). This is also a true representation of Coronationville, the location in which RMMCH hospital is located. The Census (2011) revealed 60.12% of the population in Coronationville to be Black African, 38.07% were Coloured, 0.70% were white, 0.64% were Indian/ Asian and 0.47% were 'other' races. Results from the current study correspond with Statistics South Africa (2018). The current study's findings could also be explained by South Africa's unequal past of apartheid still having an effect on race discrimination (Coovadia, Jewkes, Barron, Sanders & McIntyre, 2009), and more non-Whites attending public hospitals such as the research site. Race is an important factor to consider within the EHDI context, as a study by Clements, Barfield, Kotelchuck and Wilber (2008)

suggest that the non-White race is associated with lower social economic statuses and poor follow-up return in early intervention services.

On analysis of nationality and home language, results revealed that the largest proportion of participants were South African and spoke isiZulu. This is a true representation of the South African population, as isiZulu is spoken by 24.6% of the population, and is the most widely spoken language (South African Government, 2018). Numerous other nationalities and home languages were found in the current study, which is also characteristic of the multicultural and multilingual diversity within the South African population (South African Government, 2018).

In terms of level of education, the majority of participants received a secondary education, with the minority receiving a tertiary education. A large number of studies have noted that higher maternal educational levels are significantly associated with improved compliance to ECI and/ or EHDI programmes (Diener, Zick, McVicar, Boettger & Park, 2017; Holte, Walker, Oleson, Spratford, Moeller et al., 2012; De Souza, Sardessai, Joshi, Joshi,et al., 2006). In addition, a recent study by Ohonba, Ngepah and Simo-Kengne (2019) suggests that maternal education strongly influences child health outcomes in South Africa. Audiologists, or any health care professional therefore need to provide extra support and ensure adequate information exchange and education to caregivers. This can possibly ensure effective ECI and EHDI management within the South African context.

5.2.2 Demographic characteristics of participants' children. The demographic details of the children in the study was also analysed prior to analysing their case history factors and their current general and audiological development. With regards to gender, the current study revealed more females than males to be present within the sample. These children were born between 2013 – 2015, and these results are contrary to the statistical data obtained from Statistics South Africa (2015), where more males than females were reported

to be born in South Africa. The literature has established males to be at a higher risk for postnatal complications and mortality (Ballot, et al., 2012; Banga, Barche, Singh, Sheehan and Vasylyeva, 2015). With regards to age, all of the children in the current study were born preterm, and as mentioned previously presented with a mean chronological age of 49.2 months (four years, one month), and a mean adjusted age of 47.2 months (three years, nine months). These findings are inconsistent with the results obtained from Chattopadhyay and Mitra (2015) study who analysed the neurodevelopmental outcomes of high-risk neonates in India, at one to two years of age. Majority of the children in their study sample were born term, 244 (60.7%), with the minority born preterm, 158 (39.3%). The current study's findings are however consistent with studies from high income countries. Suppiej, Rizzardi, Zanardo, Franzoi, Ermani, et al., (2007) examined 206 high-risk neonates in Italy. All 206 neonates examined were born pre-term; 120 were born less than 33 weeks, and 53 were born less than 30 weeks. Similar findings were also noted by Humberg and colleagues (Humberg, Härtel, Paul, Hanke, Bossung, Hartz, et al., 2017) who analysed 2203 high-risk neonates who were considered VLBW infants at birth in Germany. A significant proportion of neonates (n=965), regardless of mode of delivery were born premature, thus highlighting variation of high risk neonatal profiles between countries.

5.3. Case History Factors of Participants' Children

In order to address the first sub-aim of the current study, a discussion of the case history factors of the children will be provided. All of the children in the current study were considered high-risk at birth. Although there is no universal definition of high-risk neonates, Kanji (2016b) viewed her study sample as characteristic of this population as all the children required special health care needs. The birth history which classified these children as high-risk were obtained from the children's hospital files, data obtained from Kanji's (2016b)

study, and the RTHB when hospital files were not accessible. The HPCSA (2008) reports that health records should be kept for a duration of six years as from the date the file became dormant. The current study therefore advocates for appropriate record management, especially since children in the current study are considered high-risk and are more likely to present with developmental and/ or medical concerns after birth. Appropriate record management can also assist retrospective studies in exploring hearing development, such as in Baillieu, Khoza-Shangase and Jacklin (2016) study, and other neurodevelopmental outcomes as analysed in Werner, Dawson, Osterling & Dinno (2000) study.

5.3.1 Preterm birth and low birth weight classes. The most frequently occurring case history factors in the current study sample included; preterm birth, low birth weight classes, prolonged hospital stay, increased bilirubin levels (NNJ and hyperbilirubinemia), ototoxic medication and mechanical ventilation. Among the least frequently occurring case history factors included; neurological conditions, HEI or HEU, intermediate or low APGAR scores, syndromes, in-utero infections and a family history of a hearing loss. These results are consistent with a South African based study by Thompson (2000) who analysed the neurodevelopmental outcomes of high-risk infants in Cape Town. Among the most common case history factors were preterm birth and low birth weight classes. In addition, the current study's findings are inconsistent with the results obtained from Chattopadhyay and Mitra (2015) study, who as mentioned previously, analysed the long term neurodevelopmental outcomes of high-risk neonates in India. Their results revealed that the most common case history factors included; term birth, sepsis and pneumonia. The less prevalent case history factors included preterm birth, low birth weight classes, asphyxia and increased bilirubin levels. These findings implies that the profile of high-risk infants differs per country. Within the South African context, literature and results from the current study suggest that two of the

most prevalent case history factors among high-risk neonates are preterm birth and low birth weight classes.

All 67 children in the current study were born preterm and presented with a low birth weight, with the majority, (71.6%), presenting with a VLBW. A 2012 systematic review estimated that 14.9 million preterm births occurred globally in 2010, equating to 11.1% of all live births worldwide (Blencowe Cousens, Oestergaard, Chou, Moller, et al., 2012). More than 60% of preterm births were in south Asia and sub-Saharan Africa, where 52% of the global live births occur (Blencowe Cousens, Oestergaard, Chou, Moller, et al., 2012). Approximately one million liveborn babies die each year from complications of preterm birth (Lawn, Gravett, Nunes, Rubens, Stanton, et al., 2010) and it is also a major cause of multiple perinatal morbidities (Gladstone, White, Kafulafula, Neilson & van den Broek, 2011). Preterm birth and LBW rates in South Africa are as high as 14.2%, as opposed to 7.0% in developed countries (Howson, Kinney & Lawn, 2012). These current study's findings are also consistent with the literature which suggests that preterm birth and low birth weight classes often co-occur (Fouché, Kritzinger & le Roux, 2018; Schieve, et al., 2002).

There is a paucity of literature analysing the long-term outcomes of preterm, and low birth weight infants within the South African context. Firstly, there has been a study by Kalimba & Ballot (2013) which focused on the outcomes of the late preterm population (born between 34 and 37 weeks gestation). Secondly, with regards to the low birth weight studies in South Africa, the majority are focused on the VLBW population (Ramdin, et al., 2018; Ballot, et al., 2017) and one study has focused on the ELBW population (Bopape-Chinyange, et al., 2016). Of the study samples which were observational in nature (Ramdin, et al., 2018; Kalimba & Ballot, 2013), study samples were small (median sample size n = 89, IQR 73 - 105), as the current study. Although there is a paucity of literature regarding these

populations, preterm birth and low birth weight are highly prevalent within the South African population, highlighting the need for further larger scale studies of this population.

5.3.2 Prolonged hospital stay. On further analysis of the most common case history factors present in the current study, a prolonged hospital stay was the third most commonly reported. Advances in neonatal care has increased the number of children treated in hospital, and those who remain in hospital for longer periods often present with complicated conditions (Ayers, et al., 2007). Several recognized risk factors for prolonged hospital stay include; apnea, at least part of the hospital stay in NICU, use of CPAP, mechanical ventilation, and prematurity (Weisgerber, Lye, Li, Bakalarski, Gedeit, et al., (2011). The total length of hospital stay in the current study included time spent at the NICU, high care and/ or KMC wards. The mean length of stay was 27.5 days (*SD* = 15.6 days, range days 8 – 87). Contrary results were obtained by Bassingthwaighte and Ballot (2013) who studied the outcomes of children 'born before arrival' and weighing more than 500 grams, in South Africa. The mean duration of hospital stay in their study was ±16.4 days. Differences in the studies could be a result of children being less at risk; born before arrival instead of preterm and/ or presenting with normal birth weights. In addition, Weisgerber, Lye, Li, Bakalarski, Gedeit, et al., (2011) studied factors which predict prolonged hospital stay for infants with bronchiolitis in Winconsin. Results revealed that hospital stay can be affected by number of clinical assessments. These findings suggest that there is inconsistency with regards to length of hospital stay of the high-risk population, which can depend on the child's birth weight, gestational age, and other possible compounding case history factors.

5.3.3 Increased bilirubin levels. From further analysis of the most common case history factors in the current study, a large proportion of children presented with increased bilirubin levels. Fifty six children (83.6%) presented with NNJ, and 54 of these children

(80.6%) were known to have received PTT. Furthermore, two (3.0%) children in the current study presented with hyperbilirubinemia and received both PTT and EBT. With regards to prevalence of abnormal bilirubin levels, NNJ prevalence studies are subject to limitation as diagnosing NNJ largely depends on the visual inspection which has been found to be untrustworthy (Keren, Tremont, Luan, & Cnaan, 2009). A recent study by Brits, Adendorff, Huisamen, Beukes, Botha, et al., (2018) aimed to determine prevalence of NNJ in healthy term neonates in a district hospital in Bloemfontein, South Africa. Findings were consistent with the current study's results, as more than half of infants presented with NNJ. It can be assumed that the prevalence of NNJ would be increased within the high-risk population, as seen in the current study.

A higher proportion of NNJ compared to hyperbilirubinemia was found in the current study. These results are consistent with a population-based study in Pakistan where 466 (27.6%) newborns presented with NNJ and the detected rate of hyperbilirubinemia among the1690 newborns was 39.7⁄1000 live births (95% CI 29.3–47.6) (Tikmani, Warraich, Abbasi, Rizvi, Darmstadt, et al., 2010). There is a paucity of recent South African studies indicating the prevalence of hyperbilirubinemia. A study by Ballot and Rugamba (2016) does however suggest that severe neonatal hyperbilirubinemia was not common among infants at *Charlotte Maxeke Johannesburg Academic Hospital* (CMJAH), which the current study supports within the RMMCH context.

5.3.4 Ototoxic medication. In addition to NNJ, a large proportion of children in the current study also received ototoxic medication at birth. In South Africa, a large number of medications which are prescribed to treat cancers, tuberculosis and infections are ototoxic (Wium & Gerber, 2016). The most commonly reported ototoxic medication used in the current study was gentamycin. These results correlate with English, et al., (2004) notion that

gentamycin is one of the most commonly used drugs within the neonatal population. Gentamycin is a commonly used to treat infections such as pneumonia and neonatal sepsis (Cantey, Wozniak & Sanchez, 2015), and its popularity is due to its clinical and cost effectiveness, and easy access (English, et al., 2004). The downfall of gentamycin is that it has the potential for toxicity, more specifically, ototoxicity and nephrotoxicity. With regards to ototoxicity, Pacifici (2015) suggests that gentamycin has the potential to cause a hearing impairment, with a greater potential in preterm neonates. Since all the children in the current study were born preterm, and a large proportion received gentamycin, which highlights the possible need for co-ordinated monitoring of gentamicin serum concentrations and audiological status within this population.

5.3.5 Mechanical ventilation. The last most common case history factor in the current study which will be discussed is mechanical ventilation. Eighteen (26.9%) of the children in the current study received mechanical ventilation. Mechanical ventilation is used to completely or partially assist spontaneous breathing (Hyzy, 2018). Common reasons for mechanical ventilation include; respiratory difficulties from pneumonia such as RDS, viral infections or neurological conditions (Torpy, Campbell & Glass, 2010). In addition, severe heart disease, sepsis or multiple organ failures (Torpy, et al., 2010). The preterm population is especially at risk of developing these morbidities, especially RDS (Thukral, Sankar, Chandrasekaran, Agarwal & Paul, 2016; Velaphi & Rhoda, 2012). Respiratory distress syndrome (RDS) has been reported as the most common reason for mechanical ventilation and the most common cause of mortality in preterm neonates (Ochiai, 2015; Thukral, et al., 2016). All children in the current study were born preterm and these findings indicate the need for appropriate monitoring of this high-risk population.

Results from the current study revealed CPAP to be the most popular use of mechanical ventilation. These results are supported by the numerous South African based studies which have demonstrated the wide use and effectiveness of CPAP within high risk populations (Ballot, Chirwa & Cooper, 2010; Pieper, Smith, Maree, 2003; Jeena, Pillay, Adhikari, 2002). Results are further supported by Ho, Henderson-Smart and Davis (2002) who researched the effect of early versus delayed initiation of ventilation on preterm neonates presenting with RDS. Ho, et al., (2002) results revealed that the early initiation of CPAP decreased the need for intubation for ventilation by 45.3%.

5.3.6 HIV status. Analysis of the least frequently occurring case history factors in the current study revealed that six (9.0%) children presented with an HIV status of exposed, and four (6.0%) were classified as HEU. These results correlate with Powis, Slogrove and Davies (2018) report of South Africa's public health success of a large reduction of number of babies born with HIV over the last decade. The amount of HIV infections in children has decreased from 70 000 in 2003, to 13 000 in 2017.

All children in the current study were considered preterm, and therefore of the four children who presented with an HIV status of HEI, they also presented with a preterm birth. A preterm birth and exposed HIV status has been associated with an increased risk of morbidity and mortality (Ryder, Nsuami, Nsa, Kamenga, Badi, et al., 1994; Abrams, Milner, Kwiek, Mwapasa, Kamwendo, et al., 2004). This is especially important within the current study's context as four children were considered HEU and no further testing was performed, hence, the true HIV status was unknown. Sherman (2015) does not support these findings, and recommends that children who present with a PCR (negative) result at birth should receive another HIV PCR test at 10 weeks of age (not less, as results will be affected by the viral load) in order to identify any intrapartum infections. In addition, The National

Department of Health (2014) suggest that high-risk children who received prolonged prophylaxis should receive a third HIV PCR test at 16 weeks. These findings suggest that ongoing monitoring is needed for children whose HIV status in unclear. This can ensure appropriate early intervention for this population.

5.3.7 APGAR scores. Intermediate and low APGAR scores were less prevalent in the current study as two children presented with intermediate scores and one child presented with low APGAR scores. There is a large body of evidence associating low APGAR scores with neurological manifestations (Montgomery, 2000; Ehrenstein, 2009; Padayachee & Ballot, 2013). The current study supports this notion as one child presented with intermediate APGAR scores and perinatal asphyxia and another presented with queried perinatal asphyxia due to low APGAR scores.

5.3.8 Neurological conditions. On further analysis of the neurological manifestations, seven children in the current study presented with neurological conditions. Within the South African context neurological conditions are becoming increasingly prevalent due to low APGAR scores, but also due to influencing factors such as malnutrition, negative perinatal conditions, malaria, HIV/AIDS and demographic transitions (Silberberg & Katabira, 2006). The most common neurological conditions in the current study included IVH and perinatal asphyxia. There is inadequate prenatal care for majority of African women who deliver prematurely, and their neonates are less likely to receive mechanical ventilation (Howell, Holzman, Kleinman, Wang & Chassin, (2010). There is a 2-fold higher rate of IVH related mortality in African ancestry neonates (Qureshi, Adil, Shafizadeh & Majidi, 2013). The prevalence of IVH in VLBW neonates in South Africa is estimated at 53.0% (Sandler, Cooper, Bolton, Bental & Simchowitz, 1994). The prevalence of perinatal asphyxia in South

Africa according to a study conducted by Bruckmann and Velaphi (2015), ranges from 8.7 to 15 in every 1000 deliveries.

Findings from the current study relating to low APGAR scores and neurological conditions are supported by a South African study (Padayachee & Ballot, 2013) who retrospectively analysed children with intermediate or low APGAR scores and a birth weight greater than 1800g. Results from this study revealed that the prevalence of perinatal asphyxia was 4.7/1 000 live births. However, unlike in the current study Padayachee and Ballot (2013) further found evidence of hypoxic ischaemic encephalopathy (HIE), 3.6/1 000 live births within their sample. Although the sample sizes was small Padayachee and Ballot's (2013) findings suggested perinatal asphyxia to be a significant neurological condition associated with low APGAR and low birth weights within the South African context.

5.3.9 Syndromes. With regards to syndromes, one child presented with Isotretinoin syndrome, and one child presented with low set ears and micrognathia in the current study. According to The National Organization for Rare Disorders (NORD) (2003) Isotretinoin syndrome/ fetal retinoid syndrome (FRS) results from maternal use of retinoids during pregnancy. As a result, mental and physical birth defects occur. Browne, Mason and Tang (2014) further suggest that results on the fetus include craniofacial, central nervous system, cardiovascular and thymic abnormalities. The ear defects include microtia, anotia and stenosis of the external ear.

5.3.10 In-Utero conditions. Analysis of the less frequently occurring case history factors in the current study further revealed that only one child presented with an in-utero infection, more specifically congenital syphilis. These results are in agreement with The National Antenatal Sentinel HIV and Syphilis Survey Report (2015) who revealed a low prevalence of syphilis among pregnant woman in South Africa in 2015, 2.0%. The syphilis

prevalence rate was in addition relatively low across districts in Gauteng, in comparison to other provinces.

The World Health Organization (2007) began a global initiative to reduce mother-to-child transmissions of congenital syphilis. The low incidence of congenital syphilis in the current study suggests success of The WHO (2007) initiative. This finding is supported by Wijesooriya, Rochat, Kamb, Turlapati, Temmerman, et al., (2016) who revealed that the incidence of congenital syphilis has reduced globally from 2008 – 2012, through the use of penicillin, indicating success of the WHO initiative. In addition, higher rates of congenital syphilis has been reported when mothers receive poor antenatal care, or attend few visits (Saloojee, Velaphi, Goga, Afadapa, Steen, et al., 2004). The current study therefore also suggests that the low prevalence of congenital syphilis in the current study sample could also be associated with improved antenatal care at the research site, and a large proportion of participants who returned for their NNFU clinic appointments.

5.3.11 Family history of hearing loss. The last case history factor which will be discussed under this sub-aim is a family history of a hearing loss. One child in the current study presented with a family history of a hearing loss. Most of the research surrounding this case history factor has focused on hearing impairment prevalence studies. For example, Ramma and Sebothoma (2016) study aimed determine factors associated with hearing impairment within a metropolitan area in Cape Town, South Africa. The odds ratio test revealed that those with a family history of a hearing loss were 3.02 times (CI - 1.93, 4.73) more likely to present with a hearing impairment. Contrary results were however obtained by Driscoll, Beswick, Doherty, D'Silva and Cross (2015) who retrospectively studied the prevalence of a family history of a hearing loss for children with congenital and postnatal hearing losses, in Queensland, Australia. A low yield was present for both congenital (1.4%)

and postnatal groups (1.7%). These findings suggest inconsistent results regarding the validity of family history of a hearing loss as a case history factor for hearing impairment. This will be elaborated on in the discussion under the fourth sub-aim.

5.4. Current General Development Profile of Participants' Children

The second sub-aim of the current study was to describe the current general developmental profile of children who were considered high-risk at birth and enrolled in a risk-based hearing screening programme. Although hearing forms part of general development, numerous developmental screening assessments, including the PEDS, do not include hearing as a domain. Therefore 'general development' for the purpose of the current study refers to the domains that the PEDS assessed. These include; fine motor, gross motor, expressive language, receptive language, self-help, and social-emotional domains, and 'other' concerns. The current researcher advocates for understanding the high-risk population's development as a whole, as this can improve intervention outcomes and can encourage the involvement of other disciplines and/ or ensure appropriate referrals are made. A discussion of the participants' concerns from the PEDS will firstly be discussed, followed by a discussion of the PEDS pathways. Thereafter, results from the PEDS: DM will be discussed. Hospital file review results will be used to supplement findings obtained from the PEDS measures.

5.4.1 Caregiver concerns. Results from the PEDS revealed that caregivers of the high-risk population present with widespread concerns ranging from toe-walking, to querying the HIV status of one's child. The most common domain that participants expressed concerns in was 'other', followed by behavioural concerns, expressive language and social-emotional concerns. Among the less frequently occurring domains of concern were gross motor, fine motor and self-help. These results correspond with McLeod, Crowe, McCormack, White,

Wren, et al., (2018) findings, who studied caregiver concerns of four to five year olds at childhood centres in Sydney using the PEDS. Like the current study, the most common areas of caregiver concern included expressive speech, behaviour and social-emotional domains. Amongst the least common domains of concern was school readiness, receptive language, self-help, fine motor and gross motor skills. Findings differ in the sense that McLeod, et al., (2018) did not consider the 'other' domain, and since children were slightly older than the children in the current study, 'school readiness' was also assessed. These results are further supported by Chunsuwan, Hansakunachai and Pornsamrit (2016) who found the most common caregiver concerns within the Thai population via the PEDS was behavioural, social–emotional and expressive language problems.

Caregiver concerns according to Chung, Liu, Chang, Chen, Tang, et al., (2011) are most commonly related to domains that are more explicit. For example, expressive language concerns are easier to detect than a cognitive delay. The current researcher supports Chung, et al., (2011) findings, and believes that behaviour, expressive language and social-emotional concerns are more noticeable concerns to detect than motor, self-help or receptive language concerns, as seen in the current study.

The largest proportion of participant concerns from the current study derived were from the 'other' domain, which most commonly related to visual/ eye and ENT related concerns (unresolved allergic rhinitis and tonsillitis). These results correspond with information obtained from the hospital file review as the majority of the children presented with ENT concerns at NNFU appointments, for example rhinitis. Furthermore, eye concerns, tonsillitis and bronchitis were most commonly reported at out-patient visits, and bronchitis was the most common reason for re-admission. These results are supported by Cox, Huntington, Saada, Epee-Bounya and Schonwald (2010) who suggest that caregivers from lower income families in Boston often used the PEDS as an opportunity to address any

additional concerns. These findings are also supported by Saigal and Doyle (2008) who associated preterm and VLBW infants to be at an increased risk of visual problems in Canada. With regards to ENT and medically related concerns, tonsillitis, allergic rhinitis and bronchitis are reported to be some of the most common paediatric conditions, regardless of whether children are high-risk or not (Nyquist, Gonzales, Steiner & Sande, 1998; Barr, Al-Reefy, Fox & Hopkins, 2014). The current study therefore re-emphasises that the high-risk population presents with a broad variety of conditions during their preschool years, and therefore appropriate developmental and medical screening is needed for this population.

5.4.2 The PEDS outcomes. Results from the PEDS further revealed that a large proportion of children presented on path B (moderate risk for developmental delay). Eleven (16.4%) of children presented on path A, indicating a high-risk for developmental delay. These results differ slightly from the results obtained from Coghlan, Kiing and Wake (2003) study who utilized the PEDS within a day-care setting in Australia. Findings from their study indicated that the largest proportion of children presented on path E (low risk for developmental delay). However, children in Coghlan, et al., (2003) study were not considered high-risk unlike in the current study. Despite the profile of children in the current study and them being considered high-risk, a small proportion presented on path A (high risk for developmental delay). These findings raise questions regarding what constitutes high-risk in different contexts, and whether all high risk factors that are of medical concern necessarily equate to negative consequences in terms of development. It may also highlight the presence of cofounding variables such as caregiver involvement, which may positively or negatively influence outcomes.

In addition, although a small proportion of children in the current study were considered high-risk (path A), the large majority did not meet their milestones on the PEDS: DM measure. This questions caregiver knowledge regarding developmental milestones,

which will be elaborated further later in the chapter. This also indicates the importance of using more than one measure such as both the PEDS and PEDS: DM in combination. This is supported by Abdoola, Swanepoel, Van Der Linde and Glascoe (2019) who recommends using PEDS in combination with other screening measures.

5.4.3 The PEDS: DM outcomes. The Parents' Evaluation of Developmental Status: Developmental Milestones (PEDS: DM) results revealed the domains that were mostly likely to be unmet. A significant proportion of children in the current study, (n = 46; 68.7%) did not meet their fine motor milestones, followed by receptive language milestones (n = 30; 44.8%), and expressive language milestones (n = 27; 40.3%). These results are contradictory to the literature which suggests speech and language delays are the most common developmental delays (Restall & Borton, 2010; McLeod & Harrison, 2009). The current study acknowledges that speech and language delays are highly prevalent, but there is also a need for further focus and caregiver education regarding other developmental domains.

Fine motor milestones and receptive language skills were least reported as concerns amongst caregivers in the current study. However, the PEDS: DM revealed these domains to be amongst the most common domain children were not meeting in terms of their milestones. These findings may suggest poor caregiver knowledge regarding appropriate developmental milestones, especially relating to fine motor and receptive language during the preschool period. These results are supported by Grantham-McGregor (2007) who reports children's developmental delays in South Africa are most likely due to poor caregiver education. Grantham-McGregor (2007) also considers compounding factors such as poverty, malnutrition, high rates of infection which is prevalent in developing countries such as South Africa. These findings are significant as Van Belkum, and Meintjes (2013) report that health care professions often rely on caregivers' knowledge about their child's development to make correct decisions, management and referrals. Findings from the current study and literature

have implications for appropriate caregiver education and the need for co-ordinated developmental screening within the South African context, as the high-risk population may present with a range of developmental and medical concerns well into their preschool years.

5.4.4 Developmental surveillance programmes. A study conducted by Doyle, Anderson, Battin, Bowen, Brown, et al., (2014) suggested a possible model for appropriate developmental surveillance of preschool children in Australia, as it was noted that follow-up return rates for medical appointments decrease during this time period, which is similar to literature related to follow-up return rates for audiological appointments in the South African context (Kanji, 2016b; Kanji & Krabbenhoft, 2018; Atmore, van Niekerk & Ashley-Cooper, 2012).

With regards to the measures which could possibly be used at developmental surveillance programmes, Doyle, et al., (2014) lists many assessments which could be used to assess the physical, mental, learning and cognition, quality of life and family variables of the child. However, the current researcher believes that numerous measures would not be feasible to conduct within the South African overburdened health care context. The current author therefore suggests the PEDS and PEDS: DM measures to be a quick and efficient measure to address all general developmental and developmental surveillance initiatives. Since the PEDS revealed poor caregiver education regarding age appropriate developmental milestones, there is benefit in using both the PEDS and PEDS: DM in combination to adequately screen for delay. At the same time it will educate parents as to milestones their children should be presenting with during the preschool phase. The PEDS measure questions also gave them ideas as to how to encourage development. The current researcher's notion of implementing the PEDS measures within the South African context is supported by the New South Wales Ministry of Health. This Australian State introduced the PEDS as a routine measure of surveillance for all child health professionals (Eapen, Woolfenden, Williams, Jalaludin,

Dissanayake, et al., 2014). The current study's results are further supported by Van der Linde, et al., (2015) whose study revealed the PEDS measures to the PEDS and PEDS: DM were able to identify children with developmental delays or disorders and provided a referral pathway with better efficacy than the RTHB. Findings from the study by Abdoola, Swanepoel, Van Der Linde and Glascoe (2019) indicated that the PEDS tools identified 56% (n = 97), as opposed to the BSID-III that identified 35% (n = 61) of the 158 children with possible developmental delays. Although more than one measure is recommended for developmental screening, feasibility would need to be considered, particularly within an overburdened South African health care context. Therefore until more research is done to suggest a quick and efficient screening measure, the PEDS tool may be used as an interim measure to supplement the commonly used RTHB, with considerations for the inclusion of measures to monitor hearing and communication.

5.5. The Audiological Development of Participants' Children

The third sub-aim of the current study was to specifically describe the audiological development of children who were considered high-risk at birth and enrolled in a risk-based hearing screening programme. The hearing and communicative checklist was used as the PEDS does not include auditory outcomes. Findings from the hearing and communicative checklist will be discussed.

Four (6.0%) children in the current study did not meet their auditory milestones according to the hearing and communicative checklist. Hearing impairment can occur in children who are not considered high-risk, however, the prevalence is higher within the high risk population. The prevalence ranges from 0.09 to 2.3% in low-risk neonates (Prieve & Stevens, 2000; Korres, Nikolopoulos, Komkotou, Balatsouras, Kandiloros, et al., 2005), and 0.3 to 14.1% in the high-risk neonates (Roth, Hildesheimer, Maayan-Metzger, Muchnik,

Hamburger, et al., 2006; Sassada, Ceccon, Navarro & Vaz, 2005). There is a paucity of studies related to prevalence of delayed-onset hearing loss within South Africa, and other developing countries. An earlier study conducted by Fortnum, Davis, Summerfield, Marshall, Davis, et al., (2001) suggests that the prevalence of a hearing impairment is likely to increase after the neonatal period. Fortnum, et al., (2001) study revealed that a hearing impairment greater than 40dB is likely to increase from 1.06/1000 at birth to 1.65/1000 at nine years of age for children in the United Kingdom.

5.5.1 Caregiver audiological concerns. On closer inspection of the hearing and communicative checklist findings, participants presented with a broad range of concerns. The most common concern was listening difficulties especially when occupied with another task. This was closely followed by previous ear trauma, and ignoring others. Both participants who reported their children to ignore others, did in addition not meet their auditory milestones. These findings are supported by Rannard, Lyons and Glenn (2005) who found caregivers were usually the first to detect their children's speech and/ or hearing concerns. These findings justify caregiver concerns regarding hearing, speech, language and/ or developmental delay as an appropriate risk factor for delayed-onset hearing loss on the JCIH (2007) and HPCSA (2018) HRR.

Although caregiver concern has been established as reliable predictor for audiological concern, none of the participants in the current study sought further audiological assistance. The only reported reason for not returning was that participants assumed their child's hearing was normal based on the newborn hearing screening results. These results are supported by a Mann, et al., (2001) whose study revealed nine out of 10 children who *passed* their newborn hearing screening, later presented with a delayed-onset hearing loss. Caregivers were also late to address the concern as the newborn hearing screening provided with a false sense of security. These findings highlight the need for appropriate caregiver education regarding

delayed-onset hearing loss and appropriate referral pathways. Communication regarding these factors are important, despite the screening and/ or diagnostic results from the newborn hearing screening programmes. This is particularly important as the HPCSA (2018) guidelines have assigned caregivers the role of monitoring their children for delayed-onset hearing loss. If caregivers are not receiving appropriate education, this could be further delaying the identification of a delayed-onset hearing loss.

5.5.2 Audiological surveillance programmes. The hearing and communicative checklist further revealed one child who did not present with caregiver audiological concern, but did not meet her auditory milestones. These findings suggest that in addition to caregiver education and monitoring, further systems should be in place to detect possible delayed-onset hearing losses. Beswick, et al., (2012a) has suggested a protocol in Queensland consisting of a once off appointment at nine to 12 months of age, unless the child presents with family history or congenital infection as a risk factor. A high-follow up return rate would be required for this protocol to be effective. The current study revealed 120 (64.2%) caregivers contacted were not able to be followed-up on and therefore did not participate in the current study. This poor follow-up return rate therefore suggests that Beswick, et al.'s, (2012a) protocol for the Queensland population may not be effective within the South African context. Hence another possible approach would be implementing preschool hearing screening.

Preschool hearing screening according to Hall (2016) is an imperative time period to conduct a hearing screening. Particularly in contexts where UNHS is not currently being fully implemented, where follow-up return rates are poor, and where caregivers leave the hospital without completing a hearing assessment. All four of these reasons are applicable to the South African context. Furthermore, Hall (2016) suggests that continuous audiological monitoring on younger preschool children (three years of age) is not feasible due to

behavioural challenges of this age group. Wood, Stutton and Davis (2015) also suggests it is not feasible to routinely assess this age group due to limited hospital resources.

Whilst not an audiological test, the current author advocates for the implementation of the "*How Does Your Child Hear and Talk*" checklist (ASHA, 2006) to be utilised by paediatricians at NNFU clinics as a quick and effective measure. A large follow-up return rate was noted at the NNFU clinics in the current study, with only three (4.5%) participants reporting to have not attended the clinic. In addition, modifications can be made to the checklist so that it can be provided in a variety of languages to cater for South Africa's multilingual population. These findings are supported by Khoza-Shangase and Mophosho (2018) who advocate for the provision of linguistically and culturally appropriate speech-language and hearing services within the South African context. Implementing the hearing and communicative checklist will also meet the *Developmental System Model* criteria of creating effective and efficient screening programmes to ensure effective ECI.

Collaboration with health care professionals to utilize the hearing and communicative checklist will contribute towards increasing access to integrated healthcare services for the ECD population and will deepen the collaboration between paediatricians and audiologists. It could possibly also promote the optimal use of skills and expertise of health professionals. This is important as hearing affects speech and ultimately affects development as a whole. Hence, the integration of health professionals is imperative.

5.6. The Relationship between Case History Factors at Birth and the current General and Audiological Development of the Participants' Children

The last sub-aim of the current study was to determine the relationship between case history factors at birth, and the current general and audiological development of children who were considered high-risk at birth and enrolled in a risk-based newborn hearing screening

programme. The relationship between the case history factors and the PEDS pathways, as well as the milestones that were unmet from the PEDS: DM was analysed. Furthermore, the relationship between case history factors and caregiver audiological concerns and whether auditory milestones were met or unmet were analysed. The aim of this objective was to identify any contextually relevant case history factors that may highlight the need for monitoring of general development and audiological surveillance programmes.

Statistically significant relationships that were established in the current study included; a) the relationship between bilirubin treatment and PEDS pathways, b) ototoxicity and PEDS pathways, c) APGAR scores and gross motor milestones, d) length of hospital stay and receptive language milestones, and e) ventilation and social-emotional milestones.

5.6.1 The relationship between bilirubin treatment and the PEDS pathways. Firstly, the relationship between bilirubin treatment and PEDS pathways was considered statistically significant in the current study. This study associates increased levels on bilirubin requiring both PTT and EBT with a high risk of developmental delay, as both children who presented with hyperbilirubinemia and received PTT and EBT presented on path A according to the PEDS pathway. A large number of children who presented with NNJ and received PTT also presented within the 'higher risk' pathways, with 21 presenting on path B (moderate risk for developmental delays). From an observational perspective, both children who presented with hyperbilirubinemia and received PTT and EBT also did not meet their fine motor and receptive language milestones according to the PEDS: DM. There is however a conflict within the literature as to whether increased bilirubin levels are associated with developmental delays.

The current study's results are supported by Wilar, Masloman, Lestari and Tjeng (2010) who studied the outcomes of term children with and without hyperbilirubinemia at two to four years of age. Their findings suggest that there is a statistically significant

correlation between hyperbilirubinemia in term infants and developmental delay. The current study established the same conclusion within the high-risk population. However, the current study's findings and Wilar, et al.'s, (2010) study are opposed by numerous studies which established no relationship between increased bilirubin levels and developmental delays (Vandborg Hansen, Greisen, Jepsen, Ebbesen, 2012; Heimler & Sasidharan, 2010; Fallah Karimi, Bafrooee, 2013). The current author supports Wusthoff and Loe (2015) notion that the reason for this conflict in literature may be due to the level and duration of bilirubin that influences the severity of the developmental delays. Although the relationship between the exact bilirubin levels and outcomes were not explored in the current study, the current author suggests increased bilirubin levels requiring PTT and/ or EBT as a significant case history factor for developmental screening within the South African context.

Results from the hearing and communicative checklist revealed that nine participants whose children presented with NNJ and received PTT expressed a variety of other audiological concerns. Of the nine participants who expressed concerns, four children did not meet their auditory milestones. These results are supported by Amin, Prinzing and Myers (2009) who reports the auditory system to be sensitive towards increased bilirubin levels. In addition, depending on the amount and duration of increased bilirubin, this can affect the auditory system and consequently one's language skills. However, the HPCSA (2018) only considers hyperbilirubinemia requiring exchange transfusion as a risk factor for hearing loss. The current study advocates for this NJJ and hyperbilirubinemia requiring PTT and/ or EBT as a risk factor to also be considered for continued audiological surveillance.

5.6.2 The relationship between ototoxicity and PEDS pathways. The relationship between ototoxicity and PEDS pathways also proved to be statistically significant. More children who received ototoxic medication were considered at higher risk for developmental delay, that those who did not receive ototoxic medication according to the PEDS pathways.

Eight children in the current study who received ototoxic medication were also considered high-risk (path A), with 23 presenting on path B (moderate risk for developmental delay). The outcomes with regards to ototoxicity and hearing has been established in the literature. There is however a paucity of literature regarding the long term general developmental outcomes. A prospective cohort study conducted by Sukumarapilla, Dhanwadkar and Ruhman (2016) analysed the one year outcomes of 1000 newborn babies in India. Results revealed that nine babies presented with hyperbilirubinemia and received ototoxic medication, and of these presented with language delays and one presented with a gross motor delay. Concrete conclusions cannot be drawn due to the small sample size and the presence of a combination of case history factors. Nevertheless, the current researcher advocates for ototoxicity as a worthwhile case history factor to be monitored for developmental delays in children aged three to five years of age.

On analysis of the hearing and communicative checklist, 11 participants whose children received ototoxic medication presented with audiological concern. In addition, of the four children who did not meet their auditory milestones in the current study, three received ototoxic medication, more specifically gentamycin and vancomycin. With regards to these children's newborn hearing screening results; one *passed* the initial screening and *referred* on the repeat screening, one *passed* the initial and repeat hearing screening, and one *referred* the initial screening and subsequently *passed* the repeat screening. These findings reaffirm that the newborn hearing screening can be giving caregivers a false sense of security, and that the ototoxic medication could still be affecting the inner ear even after years of the drugs being administered. Literature supports this notion, as ototoxicity has been associated with a delayed-onset sensorineural hearing loss (SNHL) (Dreitht, 2000; Wroblewska-Seniuk, Wroblewska-Seniuk, Greczka, Dabrowski, Szyfter-Harris & Mazela, 2017). Frequent audiological monitoring is therefore recommended for the early detection of changes in

hearing threshold within the high-risk population receiving ototoxic medication. Currently the HPCSA (2018) only considers exposure to ototoxic medication (gentamycin and vancomycin) as a risk factor for risk-based newborn hearing screening.

5.6.3 The relationship between gross motor milestones and APGAR Scores. The relationship between APGAR scores and gross motor milestones proved to be significant. All three children who either presented with intermediate or low APGAR scores did not meet their gross motor milestones. The literature has established low APGAR scores with an increased risk of neonatal encephalopathy, and subsequent cerebral palsy (Moster, Lie, Irgens, Bjerkedal and Markestad, 2001), which can lead to delays in numerous general development domains, and predominantly motor delays. From an observational perspective, all three children who presented with low or intermediate APGAR scores in the current study were considered as moderate (path B) or elevated risk (path C) for developmental delay according to the PEDS pathways. These children also presented with delays in numerous other domains according to the PEDS: DM. These results are supported by a study conducted by Razaz, Boyce, Brownell, Jutte, Tremlett, et al., (2015) who analysed the outcomes of 33 883 children with a range of APGAR scores in Canada. Results revealed that five minute APGAR scores can be used as a marker for developmental vulnerability for children at five years of age. However, it is not certain if these children presented with a combination of other case history factors which increased the likelihood of them presenting with developmental delays. Domains assessed included physical health, social competence, emotional maturity, language and cognitive development and communication skills. These findings suggest that low and intermediate APGAR scores are associated with a higher risk of developmental delay across domains and especially within the gross motor domain. These findings suggest that low APGAR scores should be revised/ investigated as a risk factor for general developmental screening well into the preschool years within the South African context.

With regards to audiological development, none of the caregivers of children presenting with low or intermediate APGAR scores presented with audiological concern. Furthermore, all three children presenting with low or intermediate APGAR scores met their auditory milestones according to the hearing and communicative checklist. There is an overwhelming amount of evidence which suggests low APGAR scores to be an independent case history factor for congenital hearing loss (Biswas, Goswami, Baruah & Tripathy, 2012; Oliveira, Santiago, Valente, Borja, & Bernardi, 2015; Poonual, Navacharoen, Kangsanarak & Namwongprom, 2016; Coenraad, Goedegebure, Van Goudoever & Hoeve, 2010). There is however a paucity of literature suggesting low APGAR scores to be associated with a delayed-onset hearing loss, which the current study supports. These findings are in agreement with the HPCSA (2018) HRR that excludes low APGAR scores as a risk-factor for continued audiological surveillance.

5.6.4 The relationship between length of hospital stay and receptive language milestones. The relationship between a prolonged hospital stay in the NICU, high care and/ or KMC and its effect on receptive language milestones proved to be significant in the current study. The literature has suggested that children who present with a prolonged stay in the NICU are likely to present with a variety of medical concerns, for example preterm birth and low birth weight classes. Fouché, Kritzinger & le Roux (2018) have rather associated the concomitant case history factors to be associated with receptive language impairment, and not the prolonged hospital stay as an independent factor. However, Rand and Lahav (2014) reveals contrary results when analysing the impact of the NICU environment on language in preterm infants. The absence of opportunities to recognize maternal speech sounds during prolonged NICU stay was found to inhibit language development in preterm newborns. Although further research is required on this matter, Rand and Lahav (2014) advocate for stimulating preterm newborns who have had a prolonged hospital stay using speech stimuli as

soon as possible. The current study supports both Fouché, et al.'s, (2018), and Rand and Lahav (2014) findings, and recommends a prolonged hospital stay as a worthwhile case history factor for general development screening, with focus on receptive language skills.

All children in the current study who did not meet their auditory milestones, presented with a prolonged hospital stay. There is debate within the literature as to whether a prolonged hospital stay is an independent risk factor for delayed-onset hearing loss. A study by D'agostino & Austin (2004) suggests that high-risk infants who have been in the NICU are more likely to present with auditory neuropathy. These authors do however note that the high-risk infants in their study also presented with other case history factors, for example, family history of a hearing loss and hyperbilirubinemia which are likely to increase their risk of presenting with auditory difficulties.

5.6.5 The relationship between mechanical ventilation and socio-emotional milestones. The relationship between mechanical ventilation and social-emotional milestones proved to be significant in the current study. The literature has focused on the long term outcomes of ventilation on respiratory difficulties (Gibson, Hacking, Robertson, & Doyle, 2015), and not social-emotional milestones. There is also a paucity of literature associating mechanical ventilation with general developmental delays. The current study's findings do however correspond with the literature associating ventilation with long term respiratory difficulties (Gibson, et al., 2015). Of the 18 children who received mechanical ventilation, two presented with RDS, five presented with bronchitis at the NNFU appointments, one was re-admitted for bronchitis, three returned as out-patients for upper respiratory tract infections and two returned for bronchopneumonia.

The hearing and communicative checklist revealed that 17 participants whose children received mechanical ventilation expressed audiological concern, and one of them did not

meet his auditory milestones. These results are supported by Lü, et al., (2011) who found NICU stay with mechanical ventilation to be a risk-factor for delayed-onset hearing loss.

5.6.6 The association between preterm birth, low birth weight classes and general development and audiological delays. Many of the case history factors present did not present with a statistically significant relationship in terms of general development delays. However, it must be noted that some children who presented with case history factors which did not yield a statistically significant relationship did present with developmental delays. For example, children who were born preterm birth and LBW/ VLBW presented within the 'higher risk' groups on the PEDS pathways. The PEDS: DM also revealed significant delays in terms of motor and language development within the preterm and LBW population in the current study. The current study is therefore in agreement with the literature that suggests that the low birth weight population are likely to present with motor delays (Tavasoli, Aliabadi & Eftekhari, 2014; Nazi & Aliabadi, 2015; Sampaio, Nogueira, Pontes & Toledo, 2015). This is significant as motor delays in the LBW population can affect the child's capability to learn and restrict activities at school and home (Goyen & Lui, 2002). Furthermore, a study conducted by Zimmerman (2018) on language outcomes of very preterm and low birth weight children during the school age period, revealed these children do not catch up to their peers at school age, and present with total language delays. This included; receptive and expressive language, phonological awareness and grammar. Although the children in the current study were considered preterm and not very preterm, the current author advocates for the same recommendation. Early developmental screening must be conducted to provide intervention to ensure this high-risk population are reaching their developmental potential.

As mentioned previously, two recent South African based studies, one analysing the long term outcomes of late preterm infants (Ramdin, et al., 2018) and another analysing the outcomes of VLBW infants (Ballot, et al., 2017) were conducted. The late preterm population

presented with evidence of developmental disability of 7.1%, and within the VLBW population, 7.0% presented with developmental delay. Although these values may not seem significant, both authors suggest that these high-risk populations are at risk for developmental delays and require long term follow-up, which the current researcher supports.

In terms of preterm birth and delayed-onset hearing loss, all four children who did not meet their audiological milestones in the current study presented with preterm birth. There is a paucity of literature analysing the long term audiological outcomes of previously considered preterm babies. A recent study conducted in Poland, analysed the hearing outcomes of 11438 preterm neonates, of which 53.3% were considered to have a low birth weight of less than 2500 g (Wroblewska-Seniuk, et al., 2017). However, the mean time of the final diagnosis and intervention of all infants was only until day 89 of life. Nevertheless, results did not suggest preterm birth as an independent risk factor for hearing loss. The authors rather suggested that preterm infants have numerous concomitant risk factors which may increase the occurrence of a hearing impairment. These include; low APGAR scores, mechanical ventilation, hypoxia, ototoxic medication and hyperbilirubinemia. The current study supports the results obtained from Wroblewska-Seniuk, et al., (2017) study, as all four children who did not meet their audiological milestones also presented with other possible contributing case history factors, for example, exposed HIV status, ototoxic medication and RDS. These findings suggest that preterm birth should not be considered as an independent risk factor for continuous audiological surveillance.

Furthermore, of the four children who did not meet their auditory milestones, three (75.0%) presented with VLBW, and one presented with a LBW. However, as mentioned previously they also presented with other case history. Both Beswick, et al., (2012) and Vos et al., (2015) found no or weak levels of association between low birth weight and delayed-onset hearing loss. These findings may explain why the JCIH (1990) was the only guideline

which considered birth weight lower than 1500 grams as a risk factor for hearing loss. None of the recent guidelines have stipulated this case history factor to be monitored for delayed-onset hearing loss. These findings suggest that preterm birth and/or low birth weight classes (in isolation) may not necessarily be suggestive for continuous audiological surveillance. The presence of co-occurring risk factors should rather be considered.

5.6.7 The association between HIV and general developmental and audiological delays. On analysis of the relationship between HIV status and general development, the current study revealed children presenting with an exposed or PCR (negative) status were more likely to not meet their milestones according to the PEDS: DM. In addition, Results from the current study revealed that of the four children who did not meet their auditory milestones, one presented with an HIV status of exposed and one presented with an HIV status (PCR negative). These results correlate with the literature which have well established an exposed HEI status with adverse child development in multiple domains (Knight, Mellins, Levenson Jr, Arpadi & Kairam, 2000; Walker, Pierre, Christie & Chang, 2013; Sherr, et al., 2014; Mwaba, Ngoma, Kusanthan & Menon, 2015; Sherr, et al., 2018). On further analysis of the systemic review by Sherr, et al., (2014) 21 studies were analysed from developed and developing countries including America, Kenya and South Africa. The studies analysed children with HIV with an ages ranging from six months to 18 years of age. Results revealed numerous developmental domains being affected including neurocognitive, language, cognitive, global (mental and motor) and behavioural domains. Unlike the current study, Sherr, et al., (2014) study emphasise the delay in cognitive delays, and suggest monitoring of these children who are at a higher risk for special education needs.

A recent study conducted by Hrapcak, Kuper, Bartlett, Devendra, Makawa, et al., (2016) revealed that of 380 HIV infected children, 82.0% presented with a conductive hearing loss (most commonly caused by frequent ear infections), 14.0% presented with a

SNHL and 4.0% presented with a mixed hearing loss. Results revealed a history of ear infections was the most common cause resulting in hearing loss. Considering the nature of HIV and the compelling literature associating HIV with audiological difficulties, the author agrees that maternal and/ or child HIV to be a risk-factor for on the HPCSA (2018) HRR for delayed-onset hearing loss. The author further suggests continuous audiological monitoring for this risk factor especially during the preschool period and beyond.

5.6.8 The association between neurological conditions and general and audiological developmental delays. With regards to neurological conditions, as discussed previously the most common neurological conditions in the current study sample was IVH and perinatal asphyxia. The current study results revealed two children who presented with neurological conditions to present on path A on the PEDS (high-risk for developmental delay), and four were considered moderate risk for developmental delay (path B) and one was considered low risk (path E). In addition, a significant number of these children did not meet their developmental milestones according to the PEDS: DM, especially in terms of fine motor milestones (71.4%) and receptive language milestones (57.1%). These findings are consistent with Adhikari and Rao (2017) study who studied the neurodevelopmental outcomes of term infants with perinatal asphyxia with hypoxic ischemic encephalopathy stage II. Results revealed that 19.2% of children presented with language delays, 29.4% of children presented with gross motor delays and 18.2% of children presented with fine motor delays. The current study therefore advocates for general developmental screening for children presenting with neurological conditions within the South African context, with focus on motor and language milestones.

The hearing and communicative checklist revealed that none of the participants whose children presented with neurological conditions voiced audiological concern, and all of the children met their auditory milestones. These findings are consistent with O'Shea,

Allred, Kuban, Hirtz, Specter, et al., (2012) findings who observed the developmental outcomes of extremely preterm infants with IVH at 24 months of age in North Carolina.

5.6.9 The association between syndromes and general developmental and audiological delays. Syndromes were considered one of the least frequently occurring case history factors in the current study with one child presenting with Isotretinoin syndrome, and one child presenting with low set ears and micrognathia. The child who presented with Isotretinoin syndrome was considered high risk for developmental delay according to the PEDS (path A). The PEDS: DM further revealed that this child did not meet fine motor, gross motor, expressive language, self-help and social-emotional milestones. The hearing and communicative checklist further revealed this child to present with caregiver auditory concern (questionable grommets still present in one ear), and that she did meet her auditory milestones. A study conducted by Kritzinger and Steenkamp (2006) analysed the communication development of a child with FRS from six months to seven years of age in South Africa. Results revealed that this child presented with anomalies of the pinna and external auditory meati, which resulted in otitis externa at the beginning of the longitudinal study. At seven years of age, the child presented with a mild hearing loss in the right ear, which could have affected the child's expressive language delay. Both Kritzinger and Steenkamp (2006) and the current study suggest that children with FRS present with ear infections in their early years and expressive language delays. The current study further suggests that this population are also likely to present with fine motor, gross motor, self-help and social-emotional delays. Since both studies present with only a single case example, results cannot be generalised.

The second child who presented with a syndrome, presented with low set ears and micrognathia. This child presented on path E (low risk for developmental delay) on the PEDS, and did not meet social-emotional milestones according to the PEDS: DM. The

hearing and communicative checklist further revealed that the caregiver did not present with audiological concern for the child presenting with low set ears and micrognathia, and that the child met her auditory milestones. There is a paucity of literature associating low-set ears with a delayed-onset hearing loss. Stach (2003) has however associated low-set ears with possible conductive hearing losses. With regards to micrognathia, this feature can present in numerous syndromes which the HPCSA (2018) has identified for continued audiological surveillance. This includes; Treacher Collins syndrome, CHARGE syndrome and Alport syndrome. The literature has well established these syndromes to be associated with delayed-onset hearing loss (Dumanch, et al., 2017; Koffler, Ushakov & Avraham, 2015). These findings suggest that micrognathia is associated with syndromes where a relationship with delayed-onset hearing losses have been established. These findings suggest that even when further genetic testing is not conducted, low-set ears and micrognathia should be features which should indicate reason for audiological surveillance.

5.6.10 The association between congenital infections and general developmental and audiological delays. Analysis of the child presenting with congenital syphilis in the current study revealed that she presented on path E on the PEDS (low risk of developmental delay), and did not meet expressive language milestones according to the PEDS: DM. There is a paucity of literature regarding the long term outcomes of children exposed to syphilis in-utero. However, a recent study by Verghese, Hendson, Singh, Guenette, Gratrix, et al., (2018) studied children who presented with congenital syphilis into their early childhood and results revealed that four of 11 (36%) children presented with speech language delays.

The hearing and communicative checklist further revealed that the caregiver did not present with any audiological concern and the audiological milestones were met for this child. Congenital syphilis is however a known aetiology of a progressive sensorineural hearing loss at birth (Chau, Atashband, Chang, Westerberg & Kozak, 2009). Numerous

studies within the African context have also confirmed a delayed and/ or progressive-onset hearing loss within the childhood period (Sellars & Beighton, 1983; Obiako, 1987; McPherson & Swart, 1997). The lack of recent studies associating delayed/ progressive-onset hearing loss with congenital syphilis could be associated with possible advances in antenatal care and consequently lowering the incidence of associated hearing loss with congenital syphilis.

5.6.11 The association between a family history of hearing loss and general developmental and audiological delays. Lastly, analysis of the child who presented with a family history of a hearing loss in the current study revealed that she presented in path C (elevated risk for developmental delay) on the PEDS measure, and all milestones were met on the PEDS: DM measure. According to the hearing and communicative checklist, the caregiver of the child who presented with a family history of a hearing loss did not present with audiological concern and the child met the auditory milestones for her age. Findings from a study by Ramma and Sebothoma (2016) revealed that those with a family history of a hearing loss were 3.02 (CI - 1.93, 4.73) more likely to present with a hearing impairment. However, on further analysis of Ramma and Ben (2016) results, the majority of the participants presented with a hearing loss at age 30 – 60 years and older. These findings highlight the need to distinguish between a family history of congenital versus acquired hearing loss.

5.7. Summary of Main Findings

- Preterm birth (100%), prolonged hospital stay (95.5%), NNJ (83.6%), exposure to ototoxic medication (79.1%) and VLBW (71.6%) were amongst the most common case history factors present in the current study sample. Amongst the least frequently occurring case history factors present included; low and intermediate APGAR scores,

hyperbilirubinemia, syndromes, in-utero infections and a family history of a hearing loss.

- All 67 children in the current study attended their initial and repeat newborn hearing screening tests as part of Kanji (2016b) study. Forty one (61.2%) of the children attended the diagnostic assessment, of which all of them presented with hearing within normal limits.
- The Parents' Evaluation of Developmental Status (PEDS) revealed that caregivers most commonly reported concerns within the 'other' (46.3%) domain, behavioural (25.4%), expressive language (20.9%) and social-emotional (11.9%) domains.
- 'Other' caregiver concerns were most frequently related to visual/ eye and ENT concerns. The hospital file review revealed that visual/ eye and ENT concerns were also most frequently reported at NNFU appointments, out-patient visits and re-admissions.
- A large proportion of children (34.3%) presented on path B (moderate risk for developmental delay) according to the PEDS. A small proportion of children (16.4%) presented on path A, indicating a high-risk for developmental delay.
- The Parents' Evaluation of Developmental Status: Developmental Milestones (PEDS: DM) revealed that a large proportion of children in the current study, (68.7%) did not meet their fine motor milestones, followed by receptive language milestones (44.8%), expressive language milestones (40.3%) and social-emotional milestones (38.8%). Amongst the least common domains where milestones were unmet included; self-help (23.9%) and gross motor milestones (15.8%).
- Four (6.0%) children in the current study did not meet their auditory milestones according to the hearing and communicative checklist.

- The most common caregiver audiological concerns was listening difficulties especially when occupied with another task (n=3). This was followed by previous ear trauma (n=2) and ignoring others (n=2). Amongst the least frequently occurring audiological concerns included; delayed speech and complaints of itchy ears, lump behind ear, questionable ear tag, questionable grommets still in situ and excessive cerumen.
- Statistically significant relationships were established between bilirubin treatment, ototoxicity, and PEDS pathways. In addition, the relationship between APGAR scores and gross motor milestones, length of hospital stay and receptive language milestones and ventilation and social-emotional milestones.
- Observed results revealed a large proportion of children to not meet their milestones within all general development domains.
- Children who did not meet their auditory milestones presented with one or more of the following case history factors; preterm birth, low birth weight classes, prolonged hospital stay, NNJ most commonly requiring PTT, HIV status of exposed or exposed uninfected mechanical ventilation (CPAP), ototoxic medication (gentamycin and vancomycin) and RDS.
- The most common reason as to why caregivers contacted in the current study were not able to participate included a change in contact details.

This chapter provided a discussion of the results from the current study in relation to the aims of the study. The discussion provided an integration of the results to existing literature. The discussion emphasised the limitations and implications which arose from the examination of the data. The implications may impact the South African health care context where risk-based general development and audiological surveillance is being conducted, but

no systematic programme in place. These implications can possibly support further research in this area, which elaborated on in the following chapter.

Chapter 6: Conclusions, Limitations and Recommendations

The main aim of the current study was to determine the outcomes of children who were considered high-risk at birth and previously enrolled in a risk-based newborn hearing screening programme, of which the summary of main findings is as follows.

6.1. Conclusions

According to the *Developmental Systems Model* (Guralnick, 2005) an effective ECI programme must include effective and efficient screening programmes and referral components. In addition, children who are considered at risk should be enrolled in a risk-based monitoring/ surveillance programme, and points of access must be established and available for children to access ECI. The American Academy of Paediatrics (AAP) (2001) recommends the use of monitoring/ surveillance and screening measures for the identification of children who would require developmental assessment and evaluation. Currently this is not being implemented within the South African context.

The Health Professionals Council of South Africa (HPCSA) (2018) has recommended that for early identification of hearing loss, caregivers must be educated regarding risk factors associated with hearing loss. Caregivers are given the responsibility of monitoring their children's hearing ability and communicative development, and to return if concern arises. Using a risk factor registry combined with caregiver monitoring has however proven to be ineffective (Wood & Stutton, 2012). Caregivers often present with poor risk factor knowledge associated with hearing impairment and targeted surveillance appointments were not viewed as important (Wood & Stutton, 2012). Exploring the long term outcomes of high-risk neonates can help inform developmental screening and audiological surveillance programmes in South Africa, highlighting the importance of the current study.

Results from the current study revealed that caregivers most commonly report concerns that are most noticeable, such as behavioural, expressive language and social-emotional concerns. Children however most commonly presented with developmental delays within the fine motor, receptive and expressive language and self-help domains. These findings suggest that caregivers must be informed of appropriate developmental milestones for children aged three to five years, with specific reference to domains that aren't as noticeable. This can be implemented by all health care professionals, for example, paediatricians at NNFU appointments, or speech therapists at outpatient visits. These findings also encourage all health care professionals to monitor the mentioned developmental domains with care.

Findings further suggest that the high-risk population may be prone to a range of long term developmental and medical difficulties, for example visual/ eye related and ENT concerns. Health care professionals who manage this population therefore have the responsibility to consider the child holistically and assess domains beyond their area of expertise. Utilization of the PEDS measures has been proven in the current study to assist health care professionals in this process of detecting general developmental delays across domains. These findings advocate for the PEDS to not only be used by paediatricians at the NNFU appointments, but all professionals, for example, speech therapists, physiotherapists, occupational therapists and dieticians. This will give all child health care professionals further potential and opportunity to promote ECD within the South African population.

Findings suggest that continuous auditory surveillance is needed within the high-risk population. Caregiver audiological concern was proven as a reliable indicator of concern. However, caregivers did not seek further audiological assistance when concern arose. Caregivers should be educated regarding appropriate referral pathways so that they seek help

when concern arises. This will enhance the efficiency of the HPCSA (2018) principle of caregivers being assigned the responsibility for monitoring for delayed-onset hearing loss.

In order to reduce the threat of further delaying the confirmation of a delayed-onset hearing loss, the current study advocates for preschool hearing screening. Until further research establishing the efficiency of this method within the South African context is established, a fast and effective measure to detect the presence of a hearing loss is recommended. The current study revealed the "*How Does Your Child Hear and Talk*" developmental milestones provided by the American Speech-Language-Hearing Association (ASHA, 2006) as appropriate measures to be implemented by health care professionals. This measure can be used practically within public health care sector hospitals to inform and improve audiological surveillance protocols. Collaboration with paediatricians can be made to implement the checklist at NNFU appointments, which can assist in EHDI.

6.2. Limitations of the Study

It is important to note that the generalisability of the results are subject to the following limitations in the current study:

- The data was collected from one public sector hospital in Johannesburg, Gauteng and the sample size was small. The current study sample may therefore not be representative of the development of high-risk children in the rest of Johannesburg, Gauteng or South Africa.
- The Parents' Evaluation of Developmental Status: Developmental Milestones (PEDS: DM) did not assess the gross motor milestones for 10 of the children in the current study, which could have affected the results.
- Follow-up diagnostic audiometry was not conducted on all participants in the current study, and therefore the presence of a delayed-onset hearing loss cannot be confirmed.

Findings from the hearing and communicative checklist could not be compare to any current audiological assessment results.

- A few questions on the PEDS: DM (particularly from forms N, P and Q) require the child to be present, which wasn't always possible in the current study. These answers were therefore parent self-report and not directly observed.
- The different methods/ sources of extracting information for the file review checklist can introduce a bias.

6.3. Recommendations in terms of Future Directions

Exploring the general and audiological development of the high-risk population has given insight into providing effective developmental screening and audiological surveillance within the South African context. It has also highlighted the importance of health care professional and caregiver education regarding risk factors for delayed-onset general development delays and hearing loss. Therefore although the current study is subject to limitations, clinical and educational implications can still be drawn, which has the prospect of informing policy and future research studies. Future research may involve validation of the PEDS and PEDS:DM as measures in the South African context.

6.3.1 Clinical recommendations for the implementation of monitoring developmental outcomes.

- Findings suggest that all children considered high-risk should be continuously monitored across all developmental domains. However, emphasis should be placed on children who received bilirubin treatment and ototoxic medication as they were found to present with a higher risk of developmental delays. In addition, children with low APGAR should have their gross motor milestones monitored closely, and children

with a prolonged hospital stay should have their receptive language milestones monitored thoroughly.

- The case history factors of the four children who did not meet their auditory milestones may encourage audiologists to monitor certain risk factors more closely than others in an overburdened healthcare context. The current study suggests NJJ and hyperbilirubinemia requiring PTT and/ or EBT, ototoxic medication (gentamycin and vancomycin), mechanical ventilation (CPAP), and exposed/ exposed (PCR negative) status as possible risk indicators for delayed-onset hearing loss.
- The current study advocates for multidisciplinary (MDT) screening of the high-risk population. This may encourage early detection and intervention for children presenting with delays. It may also encourage ongoing support and skill sharing between child health care professionals, which can inform intervention and ensure appropriate referrals.
- The most common reason caregivers were not able to participate in the current study, was due to incorrect contact details. This calls for a national database for high-risk children, and for caregivers to inform professionals when details change. This can enhance developmental screening and audiological surveillance programmes.

6.3.2 Recommendations for education of team members.

- The results of this study highlight the important role audiologists, and all health care professionals managing the high-risk population have within both general development screening and audiological surveillance programmes. All health care professionals have the responsibility to educate themselves of case history factors that place a child at risk of developing both general and hearing development delays.

6.3.3 Recommendations for policy formation.

- Findings from the current study can be used practically at the research site, RMMCH, to inform and improve developmental screening and audiological surveillance protocols. Continuous monitoring and feedback of the effectiveness of these recommendations can indicate whether these cost-effective measures can be implemented nationally and/ or provincially.
- The current study may encourage policy makers to consider developmental screening as a requirement by law for the high-risk population in both the public and private sector. The Parents' Evaluation of Developmental Status (PEDS) can be viewed as a coordinated and cost-effective measure for developmental screening. Findings may also encourage policy makers to recommend certain risk factors for developmental screening.
- Findings may inform the HPCSA of contextually relevant risk factors associated with delayed-onset hearing loss.

6.3.4 Recommendations for future research. This study indicated a number of possible directions and opportunities for future research.

- Further large scale studies establishing the relationship between case history factors and general and hearing development delays are needed to establish contextually relevant risk factors. This will inform developmental screening and audiological surveillance programmes.
- Since only the relationship between individual case history factors and outcomes were established in the current study, there is a need for addressing a combination of case history factors and outcomes on a larger sample size.

- Reduplication of this study with both younger and older children is considered high-risk is recommended. This can give further insight as to time periods to conduct developmental screening.
- Further research regarding the inclusion of auditory outcomes on the PEDS measures is also recommended.
- Translating the "*How Does Your Child Hear and Talk*" developmental milestones provided by The American Speech-Language-Hearing Association (ASHA, 2006) so that it is more linguistically appropriate for South Africa.

The current study reveals that there is great potential and opportunity to improve general development screening and audiological surveillance within the South African context. With appropriate measures used, education of team members, renewed policy and continuous research within the ECI context can reduce the gaps within the current ECI system. Consequently, this may reduce developmental challenges to the high-risk population in South Africa.

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Appendices

Appendix A – Ethical Clearance from the Human Research Ethics Committee of the University of the Witwatersrand (medical)

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

R14/49 Miss Rumaana Bham

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

CLEARANCE CERTIFICATE NO. M180468

NAME: (Principal Investigator)	Miss Rumaana Bham
DEPARTMENT:	Speech Pathology and Audiology Rahima Moosa Mother and Child Hospital
PROJECT TITLE:	Developmental outcomes of children previously enrolled in a risk-based hearing screening programme
DATE CONSIDERED:	04/05/2018
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Dr Amisha Kanji
APPROVED BY:	Professor CB Penny, Chairperson, HREC (Medical)
DATE OF APPROVAL:	11/07/2018

This clearance certificate is valid for 5 years from date of approval. Extension may be applied for.

DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Research Office Secretary on the Third Floor, Faculty of Health Sciences, Phillip Tobias Building, 29 Princess of Wales Terrace, Parktown, 2193, University of the Witwatersrand. I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated, from the research protocol as approved, I/we undertake to resubmit the application to the Committee. **I agree to submit a yearly progress report**. The date for annual re-certification will be one year after the date of convened meeting where the study was initially reviewed. In this case, the study was initially reviewed in **April** and will therefore be due in the month of **April** each year. Unreported changes to the application may invalidate the clearance given by the HREC (Medical).

Principal Investigator Signature

Date 12/07/2018

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

Appendix B – Permission from Rahima Moosa Mother and Child Hospital (RMMCH)

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

RAHIMA MOOSA MOTHER AND CHILD HOSPITAL

Enquiries : Karen Marshall
Tel : (011) 470 9284
Fax : 086 553 4623
Email : Karen.Marshall@wits.ac.za

Rumaana Bham
School of Human Community Development
Faculty of Humanities
University of the Witwatersrand

Dear Ms. Bham,

RE: DEVELOPMENTAL OUTCOMES OF CHILDREN PREVIOUSLY ENROLLED IN A RISK-BASED HEARING SCREENING PROGRAMME

Permission is granted for you to conduct the research as indicated in the title above.

The terms under which this permission is granted is contained in the Researcher Declaration form that you have signed. Failure to comply with these conditions will result in the withdrawal of such permission.

It is crucial for you to inform the Research Coordinator, Karen Marshall of the actual start and end dates of your study. This could be done by e-mail.

Should the study commence more than 12 months after receipt of this approval letter you will have to go through the process of applying again.

You are strongly advised to keep a signed copy of the declaration form so as to ensure that the terms of this agreement are complied with at all times.

Yours sincerely,

ADJUNCT PROFESSOR ASHRAF COOVADIA
Acting Chief Executive Officer
2018:05:18

ADDRESS: Cnr. FUEL & OUDSTHOORN STREET CORONATIONVILLE 2093 / PRIVATE BAG X20 NEWCLARE 2112 JHB

Appendix C – The Participant Information Sheet

Information Letter and Consent Form for Caregivers

Dear Caregivers

My name is Rumaana Bham, and I am a speech and hearing therapist. I am studying towards my postgraduate degree through the University of the Witwatersrand. As part of my degree I must do a research study. I am therefore following-up on children who were part of another research study done at *Rahima Moosa Mother and Child Hospital* from 2013 - 2015.

What is a Research Study?

Research is done when we want to find out new information, so that we can do things in a better way.

In my study I want to find out how your child is developing - walking, behaving, hearing and talking etc. I would therefore like to invite you to take part in my study, which will be explained below.

Why am I asking about your Child's Development?

There are certain skills that a child should be doing at certain ages. If the child is not doing these certain skills e.g. walking at 1 year, it tells us that we must help him/ her to learn these skills. It is therefore important to keep an eye on children to see if they are developing properly.

What Is the Process?

I am going to ask you about your child's general development and hearing development. The sets of questions will take about 15 minutes to complete.

General Development Screening: I am going to ask you questions about what is worrying you about your child's development. I am also going to ask you if your child can perform certain skills (how he/ she walks, behaves, speaks).

It is okay if your child cannot do some of the skills, as I am trying to see where your child is in terms of his/ her development, and if we can help him/ her.

Audiological monitoring: I will ask you questions regarding your child's hearing. This is to see if your child has any difficulty hearing.

At the end I will give you feedback regarding the results from the sets of questions I asked you about. If need be referrals will be made to appropriate professionals, unless you are already seeing these professionals.

File Review: I will look through your child's hospital file with your permission and take down any important information.

Privacy

Your child's name or hospital number from the information obtained from the general developmental screening, audiological monitoring and file review, will not be used in my research study and won't be made public.

Benefits

Results from this study may advise professionals to keep an eye on some children more than others. It could also help in improving follow-up clinics and help professionals work together to provide better care for children.

Reimbursements

You will be provided with R50 for transport if you attend the general development screening and audiological monitoring appointment.

What are your Rights?

- ✓ Participation is voluntary and you have the right to withdraw from my study at any time, with no negative consequences to you or your child's future treatment.
- ✓ You have the right to know the results of the questions I ask you, and the results of my study.
- ✓ You have the right to contact me at any time with any questions or worries.
- ✓ You have the right to complain or share any concerns about my study, or me as a researcher by contacting the Human Research Ethics Committee (Medical) on 011 717 1234.

If you agree to take part in my study, please fill in the consent form below.

If you have any questions about my study you are welcome to ask me now, or later by contacting me on 0829661315.

Kind Regards,

Rumaana Bham

Appendix D – The Participant Consent Form

I, _____ caregiver of

_____ (child's name)

wish to participate in this study which, is authorized by the University of the Witwatersrand and the Superintendent of *Rahima Moosa Mother and Child Hospital*.

I agree that I will be asked questions relating to my child's general and hearing development, for approximately 15 minutes. I agree that the researcher will access my child's hospital file. I agree that I will be reimbursed for transport costs worth R50. This study has been explained to me, and I understand all the information that has been explained. I am aware of my rights as a caregiver, and that I can withdraw from this study at any point with no negative consequence. Results from this study will be confidential and my identity, as well as my child's identity will remain anonymous.

_____ _____

Signature of caregiver Date

I, the undersigned have explained the study to the participant, and have answered any questions asked by the participant.

_____ _____

Audiologist Date

Appendix E – Permission from the PEDS Author to Adapt the Tool

Rumaana Bham <rumaanabham@gmail.com> Thu, Mar 22, 2018, 5:50 PM

to Frances

Dear Prof. Glascoe

I hope you're well.

Thank you so much for your valuable advice and assistance, I really appreciate it.

In my study I will be using the PEDS and PEDS:DM to ask caregivers about their child's general development. The age range of the children will vary from about 17 months to 4 years, 5 months. However, when conducting the PEDS only caregivers will be present and not the child.

I have however noticed that certain questions, particularly on form N, P and Q require the child to be present. For example, asking the child to draw a triangle or to count. Would it be possible to adapt these questions so that they are phrased in a hypothetical case situation, for example, instead of asking a child to draw a triangle, the caregiver will be asked if his/ her child can draw a triangle. Will this influence scoring, and would it be better if these questions are rather eliminated?

Kind Regards

Rumaana Bham

Frances Glascoe <[redacted]> Mon, Mar 26, 2018, 12:05 PM

to me

Hey Rumaana!

Yes you can adapt those questions. We've done this for telephone-based screening and just preface hands-on items with "If you asked your child……"

In terms of effects on scoring, parent-report versus hands-on administration have excellent inter-rater reliability. But if parents state "I don't know", "Never tried", "I'm not sure" (or similar answers), you'll want to mark the answer options as "No".

A parent not knowing what their child can do is not a good thing and also means their child is likely not exposed to those kinds of tasks - also not a good thing.

If the above isn't clear and you want to go over the rewording of the items/response options in question, let me know.

Thanks,
Frances

Appendix F – The Hearing and Communicative Development Checklist

Questions to ask prior to the administration of the checklist:

1. Do you have any concerns regarding your child's hearing? ❑ Yes ❑ No

2. If yes, did you come back to the audiology department at RMMCH to address the concern? ❑ Yes ❑ No

3. If yes, did your child have another hearing test done since being discharged from the newborn hearing screening? ❑ Yes ❑ No

Birth to 3 Months	**Yes**	**No**
Reacts to loud sounds		
Quiets or smiles when spoken to		
Recognize your voice and quiets if crying		
Has a special way of crying for different needs		
Makes pleasure sounds; cooing		
Smiles when he/she sees you		

4 to 6 Months		
Moves his/ her eyes towards the direction of sounds		
Responds to changes in tone of your voice		
Looks at toys that make sounds		
Pays attention to music		
Babbling sounds in a speech-like manner, with many different sounds; including sounds that begin with /p/, /b/ and /m/		

7 Months to 1 Year		
Enjoys games like peek-a-boo and pat-a-cake		
Turns and looks in direction of sounds		
Listens when spoken to		
Recognizes words for common items like "cup", "shoe", or "book".		
Respond to requests, for example, "come here" or "want more?		
Babbles using long and short groups of sounds; "tata, upup, bibibi"		
Babbles to get and keep attention		
Imitates different speech sounds		
Has one or two words, for example, "hi", "dog", "dada", "mama", by his/ her first birthday		

1 to 2 Years		
Points to a few body parts when asked		
Follows simple commands, for example, "roll the ball", and understands simple questions, for example, "kiss the baby," and "where's your shoe?		
Listens to simple stories, songs, and rhymes		

Points to pictures in a book when named		
Says more words every month		
Uses some one- or two- word questions; “where kitty?" "go bye-bye?", "What's that?").		

2 to 3 Years

Has a word for almost anything		
Uses two or three word phrases to talk about and ask for things		
Uses /k/, /g/, /f/, /t/, /d/ and /n/ sounds		
Follows two requests ("Get the book and put it on the table		
Speaks in a way that is understood by family members		
Names objects when he/ she wants them or to direct attention towards them		

3 to 4 Years

Hears you when you call from another room.		
Hears television/ radio at the same loudness level as other family members		
Answers simple “Who?” “What?” “Where”, and “Why” questions		
Uses sentences that have 4 or more words		
Talks about activities at preschool, or friends’ homes		

4 to 5 Years

Pays attention to a short story and answers simple questions about it		
Hears and understands most of what is said at home and in school		
Uses sentences that give many details		
Tells stories that stay on topic		
Communicates easily with other children and adults		
Says most sounds correctly except for a few (l, s, r, v, z, ch, sh, and th)		
Names some letters and numbers		

The *How Does Your Child Hear and Talk* checklist retrieved from the American Speech–Language–Hearing Association (ASHA, 2006).

Appendix G – The Referral Letter

To whom it may concern

My name is Rumaana, and I am a qualified speech-language therapist and audiologist, and currently registered for a Master's degree (Audiology) at the University of the Witwatersrand. I am conducting a research study as part of completing the degree requirements. I am interested in researching developmental monitoring of children who were previously considered high-risk and enrolled in a risk-based hearing screening programme.

_____ caregiver of _____
has been asked a set of questions using the Parents Evaluation of Developmental Status (PEDS) and a hearing and communicative development checklist relating to his/ her child's general and hearing development. Analysis of the results indicate that
_____is falling below his/ her age level for:

_____

_____

_____

_____

Since the questions in my study are a screening tool and not an in-depth assessment, the caregiver has been given feedback, and asked to contact you for continued assessment and/ or monitoring of the above mentioned developmental areas.

Kind Regards,

Rumaana Bham

082 966 1315

Appendix H – The Hospital File Review Checklist

Section 1: General Information

Participant No.: _____

Date of Birth: _____

Gender: _____

Contact Number: _____

Home Language: _____

Date of discharge: _____

Date of file review: _____

Section 2: Significant Medical History at Birth

Birth Weight: _____

❑ ELBW: 999g or <
❑ VLBW: 1000 – 1499g
❑ LBW: 1500g – 2499g

APGAR Scores:_____
❑ Inappropriate
❑ Appropriate

Gestational Age: /40
❑ Premature

❑ HIV exposed
❑ Unknown HIV Status

❑ HIE Grade I/ II/ III
❑ IVH I/ II/ III/ IV
❑ Syndrome/ Craniofacial Anomalies: _____
❑ Other significant illnesses:
_____
_____
_____
❑ Significant medication:
_____
_____
_____

❑ CMV
❑ Herpes
❑ Rubella
❑ Toxoplasmosis
❑ Malaria

❑ Prolonged NICU Stay
❑ NNJ
❑ Birth Asphyxia
❑ Mechanical/ Assisted Ventilation

❑ CPAP
❑ ECMO

Date of discharge: _____

Section 3: Follow-up Appointments

Follow-up Appointment 1:

Date: _____

Period of time since discharge/ last follow-up appointment: _____

Age (Years and Months)	
Corrected Age (Years and Months)	
Weight (Kg)	

Length (cm)	
Head Circumference (cm)	
Diet (Type and Mode)	
HIV Status	
Ophthalmology Results (Refer/ Pass)	
Hearing Test Results (Refer/ Pass)	
Diagnosis (symptoms, onset and duration)	
Medication	
Developmental Milestones	
Other	

Follow-up Appointment 2:

Date: _____

Period of time since discharge/ last follow-up appointment: _____

Age (Years and Months)	
Corrected Age (Years and Months)	
Weight (Kg)	
Length (cm)	
Head Circumference (cm)	
Diagnosis (symptoms, onset and duration)	
Medication	

Developmental Milestones	
Other	

Follow-up Appointment 3:

Date: _____

Period of time since discharge/ last follow-up appointment: _____

Age (Years and Months)	
Corrected Age (Years and Months)	
Weight (Kg)	
Length (cm)	
Head Circumference (cm)	
Diagnosis (symptoms, onset and duration)	
Medication	
Developmental Milestones	
Other	

Follow-up Appointment 4:

Date: _____

Period of time since discharge/ last follow-up appointment: _____

Age (Years and Months)	
Corrected Age (Years and Months)	
Weight (Kg)	

Length (cm)	
Head Circumference (cm)	
Diagnosis (symptoms, onset and duration)	
Medication	
Developmental Milestones	
Other	

Follow-up Appointment 5:

Date: _____

Period of time since discharge/ last follow-up appointment: _____

Age (Years and Months)	
Corrected Age (Years and Months)	
Weight (Kg)	
Length (cm)	
Head Circumference (cm)	
Diagnosis (symptoms, onset and duration)	
Medication	
Developmental Milestones	
Other	

Immunisations

	Date	**Age**
BCG		
OPV0		
OPC1		
RV1		
DTaP-IPV-Hib1		
Hep B1		
PCV 1		
DTaP-IPV-Hib2		
Hep B2		
DTaP-IPV-Hib3		
Hep B3		
PCV2		
RV2		
Measels 1		
PCV3		
DTaP-IPV-Hib4		
Measles 2		
Td		

Appendix I – The Hospital File Review for Audiological Concerns

Section 4: Audiology

Early Audiological History

Type of Hearing Screen	Pass/ Refer and elaborate on measures used and results
Initial Screen	
Repeat Screen	
Re-Screen	
Diagnostic Assessment	

Current Audiological Status

If the child returned to the audiology department after being discharged from the newborn hearing screening, classify the caregivers concern and outcome below.

- **Initial concern:**

❑ Uni/bilateral otorrhea ❑ Otalgia/ frequently tugging on ears

❑ Delayed speech e.g. not speaking at 1 year ❑ Does not respond when called/ needs repetition

❑ Speaks loudly, or turns up the volume of the television

❑ Other:

_____

_____

- **Outcome**

❑ Cerumen occlusion. Elaborate on outcome:

_____

❑ Outer ear pathology. Specify and elaborate on outcome:

_____

❑ Middle ear pathology. Specify tympanometry results, duration e.g. acute/ chronic otitis media, and treatment:

_____

_____

❑ If AABR performed, specify results:

_____

_____

❑ If OAEs performed, specify results:

_____

_____

❑ If behavioural/ conditioned audiometry performed, specify severity and type of hearing loss:

_____
_____

❑ If ABR performed, specify results:

_____
_____

❑ Inconclusive results. Specify:

_____
_____

❑ Other:

_____
_____

Appendix J: Summary of Findings Checklist

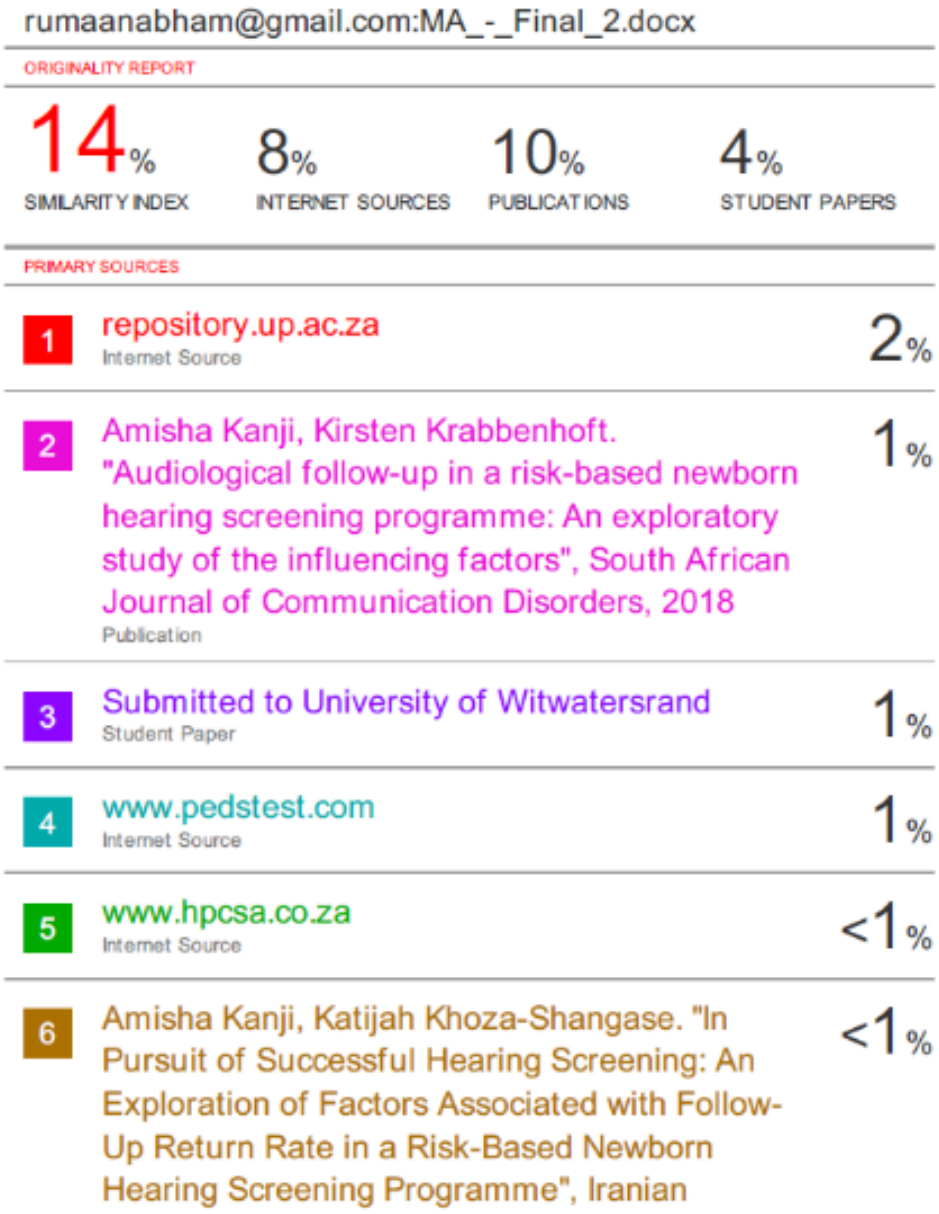
Participant No.: _____

Tick the appropriate boxes:

PEDS & PEDS:DM Screening Measures	Hearing and Communicative Development Checklist	Paediatric File Review	Audiology File Review
PEDS Path ❑ A ❑ B ❑ C ❑ D ❑ E Predictive Concerns ❑ Expressive language ❑ Receptive language ❑ Fine motor ❑ Gross motor ❑ Behaviour ❑ Socio-emotional ❑ Self-help	**Prior to completing the checklist:** Did the caregiver have any concerns regarding the child's hearing? ❑ Yes ❑ No Did the caregiver and child return to the audiology department at RMMCH to address any audiological concern? ❑ Yes ❑ No Did the child have another hearing test done since being discharged from the newborn hearing screening? ❑ Yes ❑ No	**Significant medical case history at birth:** ❑ ELBW: 999g or < ❑ VLBW: 1000 – 1499g ❑ LBW: 1500g – 2499g ❑ Inappropriate APGAR Scores ❑ Premature ❑ Prolonged NICU Stay ❑ NNJ ❑ Birth Asphyxia ❑ HIE Grade I/ II/ III ❑ IVH I/ II/ III/ IV ❑ Syndrome/ Craniofacial Anomalies ❑ CMV ❑ Herpes ❑ Rubella ❑ Toxoplasmosis ❑ Malaria ❑ HIV exposed ❑ HIV unknown ❑ Mechanical/ Assisted Ventilation ❑ CPAP ❑ ECMO ❑ Significant medication. Specify: ❑ Other significant illnesses. Specify:	**Early Audiological History** ❑ Pass initial hearing screen ❑ Refer initial hearing screen ❑ Pass repeat hearing screen ❑ Refer repeat hearing screen ❑ Pass re-screen hearing screen ❑ Refer re-screen hearing screen ❑ Pass diagnostic assessment ❑ Inconclusive results from diagnostic assessment
PEDS:DM Milestones met ❑ Expressive language ❑ Receptive language ❑ Fine motor ❑ Gross motor ❑ Behaviour ❑ Social-emotional ❑ Self-help Milestones unmet ❑ Expressive language ❑ Receptive language ❑ Fine motor ❑ Gross motor ❑ Behaviour	**After completing the checklist:** Is the hearing and communicative development of concern? ❑ Yes ❑ No Has appropriate referrals been made? ❑ Yes ❑ No Must an audiology file review be done for this child?	**Follow-up appointment information** Weight 1. 2. 3. 4. Length 1. 2. 3. 4. Head Circumference 1. 2.	**Current Audiological Status** Initial Concern: ❑ Uni/bilateral otorrhea ❑ Otalgia/ frequently tugging on ears ❑ Delayed speech ❑ Does not respond when called/ needs repetition ❑ Speaks loudly, or turns up the volume of the television ❑ Other: Outcome ❑ Cerumen occlusion

❑ Social-emotional ❑ Self-help	❑ Yes ❑ No	3. 4. Diet 1. 2. 3. 4. HIV status 1. 2. 3. 4. Ophthalmology results 1. Developmental milestones 1. 2. 3. 4. Immunisations 1. 2. 3. 4.	❑ Outer ear pathology ❑ Middle ear pathology ❑ If AABR performed, specify results: ❑ If OAEs performed, specify results: ❑ If behavioural/ conditioned audiometry performed, specify severity and type of hearing loss: ❑ If ABR performed, specify results: ❑ Inconclusive results. Specify: ❑ Other:

Appendix K: Turnitin Report



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