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EPIDEMIOLOGY AND CARE OF INDIVIDUALS WITH CLEFT LIP AND PALATE IN SOUTH AFRICA

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A thesis completed by published work

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Doctor of Philosophy

Johannesburg, South Africa

1 April 2019

DECLARATION

This thesis is submitted in the integrating narrative format, approved by the Faculty of Health Sciences, of published work.

I, Phumzile Hlongwa, declare that this thesis is my original work. It is being submitted for the degree of Doctor of Philosophy in the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at this or any other University. I am aware that plagiarism (the use of someone else's work without their permission and/or without acknowledging the original source) is wrong. I confirm that the work submitted for assessment for the above degree is my own unaided work except where I have explicitly indicated otherwise.

I have read the sections on referencing and plagiarism in the Wits Plagiarism Policy and I have followed the required conventions in referencing the thoughts and ideas of others. I understand that the University of the Witwatersrand may take disciplinary action including suspension or permanent expulsion against me if there is a belief that this is not my own unaided work or that I have failed to acknowledge the source of the ideas or words in my writing.

Signature: PHlongwa

Phumzile Hlongwa

2019/04/01

DEDICATION

To my mother, Alzinah Busisiwe Buthelezi

15 March 1937 – 08 January 1998

"Rest in Peace"

To my husband, Stha, Vusimuzi Hlongwa

I will always love you!!!

To the girls, Khanyisile Hlongwa & Hlengiwe Hlongwa

You are my pride

To all Children born with Cleft Lip and Palate

May you all be treated with dignity

To God Almighty

"For nothing will be impossible with God" – Luk 1:37 ESV

PRESENTATIONS ARISING FROM THIS THESIS

Hlongwa P. Oral presentation: "*Mothers' perceptions of care provision and support services for a child with cleft lip and palate: A pilot study* " at the 9th International Cleft Congress 2017, 8- 11 February 2017, Chennai, India.

Hlongwa P. Oral presentation: *"People look and ask lots of questions": Caregivers' perceptions of health care provision and support services for children born with cleft lip and palate"* at the South African Society of Orthodontists Conference 2017, 7- 9 September 2017, Cape Town, South Africa.

Hlongwa P. Oral presentation: *"Psycho-social aspect of cleft lip and palate treatment"* at the Symposium of Cleft Deformities 2018, 29th August 2018, School of Oral Health Sciences, Faculty of Health Sciences, University of the Witwatersrand/International Association of Dental Research/South African Division.

PUBLICATIONS ARISING FROM THIS THESIS

Phumzile Hlongwa, Laetitia C. Rispel. "People look and ask lots of questions": caregivers' perceptions of healthcare provision and support for children born with cleft lip and palate". *BMC Public Health*. 2018; **18** (1):506. doi. 10.1186/s12889-018-5421

Phumzile Hlongwa, Dandajena Tarisai C, Laetitia C Rispel. Comparative analysis of healthcare provision to individuals with cleft lip and palate at specialised academic centres in South Africa. *South African Medical Journal (in press)*

Phumzile Hlongwa, Jonathan Levin, Laetitia C Rispel. Epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa. *PLOS ONE (under review)*

ABSTRACT

Background: Cleft lip and palate is amongst the five most common congenital anomalies reported in South Africa.

Aim: In light of insufficient knowledge on cleft lip and palate, the aim of this PhD was to determine the prevalence of, and care provision to cleft lip with or without cleft palate individuals in South Africa. The specific objectives were to: describe the epidemiology of cleft lip and palate, analyse the current approach to care provision for individuals with cleft lip and palate, measure the interprofessional collaboration (IPC) among members of cleft lip and palate care team and determine the perceptions of and the support services available to caregivers of children with cleft lip and palate.

Methods: A mixed methods, cross-sectional study was conducted at 11 specialised academic centres situated in six provinces of South Africa. The four distinct, but inter-linked components of this PhD study included:- a record review of cleft lip and palate data over a two-year period; a survey of cleft lip and palate leaders in 11 centres to determine the current approach to care provision for individuals with cleft lip and palate; a survey among cleft lip and palate team members to measure IPC; and the interviews with parents or caregivers on their perceptions of health service provision and support for their children with cleft lip and palate. STATA® 13 was used for quantitative data analysis, while the qualitative data was analysed using thematic content analysis.

Results: The estimated prevalence of cleft lip and palate in the South African public sector was 0.3 per 1 000 live births, with provincial variation of 0.1/1000 to 1.2/1000. The distribution of clefts was: 35.3% cleft palate; 34.6% cleft lip and palate; and 19.0% cleft lip, with a statistically significant difference by gender.

In the majority of centres the point of care for patients with cleft lip and palate was plastic surgery (9/11 centres = 81.8%). Surgical repair of the lip and palate (10/11 = 90.9%) and speech therapy (7/11 = 63.6%) dominated the type of treatment provided, highlighting gaps in the other types of treatment.

Regarding the IPC, the domain of care expertise obtained the highest mean score of 2.92, whereas effective group function obtained the lowest mean score of 2.55. The category of health professional was the only factor that accounted for the differences in the overall IPC mean score.

The mean age of caregivers was 33.3 years (range 17–68 years). Caregivers reported feelings of shock, anxiety, and sadness, exacerbated by the burden of care provision, and their experiences of health system deficiencies, lack of public awareness, and insufficient social support services.

Conclusion: This PhD generated new knowledge on the epidemiology and care for individuals with cleft lip and palate in South Africa. A population-based surveillance system on congenital anomalies is necessary to collect relevant information, monitor trends and inform national policy. Strategies are needed to ensure the provision of a comprehensive package of essential services to individuals with cleft lip and palate, which will be enhanced with greater IPC. There should be greater involvement of caregivers in the treatment of their children, as well as public awareness campaigns on congenital anomalies.

Key Words: Epidemiology, cleft lip and palate, multidisciplinary team, interprofessional collaboration, South Africa

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Luthuli Central Hospital, Tygerberg Hospital, Red Cross War Memorial Children's Hospital, Nelson Mandela Academic Hospital and Universitas Hospital;

- Heads of clinical departments and their nursing staff;
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My sincere thanks go to the PhD academic and administrative staff at the School of Public Health for providing me with invaluable support over the last four years. I thank my "PhD family" brought together by our supervisor Professor Rispel. Your progress and development was a source of encouragement. I knew I could just send a text or an email and help was at hand. I will miss those January meetings where we shared our milestones, supported and encouraged one another.

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"The Lord is my Shepherd and I lack nothing" – Psalm 23:1 NKJV

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LIST OF ABBREVIATIONS & ACRONYMS

ANOVA	Analysis of variance
BCL	Bilateral cleft lip
BCLP	Bilateral cleft lip and palate
BDNT	Birth Defect Notification Tool
CFA	Craniofacial anomalies
CL	Cleft lip
CLP	Cleft lip and palate
CL/P	Cleft lip with or without cleft palate
CP	Cleft palate
DALY	Disability-adjusted life-years
DST	Department of Science and Technology
EC	Eastern Cape
ESV	English Standard Version
FS	Free State
GBD	Global Burden of Disease
GP	Gauteng Province
HCP	Healthcare professional
HICs	High-income countries
HIV&ADIS	Human Immunodeficiency Virus & Acquired Immune Deficiency Syndrome
HPCSA	Health Professionals Council of South Africa
HREC	Human Research Ethics Committee
IQR	Inter-quartile range
IPC	Interprofessional collaboration

JL	Jonathan Levin
KZN	KwaZulu Natal
L	Limpopo
LR	Laetitia Rispel
LMICs	Low-and middle-income countries
NCD	Non-communicable disease
NHI	National Health Insurance
NGO	Non-governmental organizations
NRF	National Research Foundation
OFC	Orofacial clefts
PH	Phumzile Hlongwa
PhD	Doctor of Philosophy
PI	Principal Investigator
RNAO	Registered Nurses' Association of Ontario
SABDSS	South African Birth Defects Surveillance System
NDoH	National Department of Health
SARChI	South African Research Chairs Initiative
SD	Standard deviation
TB	Tuberculosis
TCD	Tarisai C Dandajena
UCL	Unilateral cleft lip
UCLP	Unilateral cleft lip and palate
WC	Western Cape
WHO	World Health Organization
YLD	Years living with disability

CHAPTER 1
Introduction

1.1 Background

Congenital anomalies are defined as abnormalities of structure, function, or metabolism that are present at birth, with some clinically noticeable at birth while others may only be diagnosed later in life [1, 2]. Worldwide, congenital anomalies are among the top 20 causes of the global burden of disease, accounting for more than 25 million disability-adjusted life-years (DALYs) [3]. These anomalies are a major cause of infant mortality and childhood morbidity and disability [4, 5]. Cleft lip and palate (CLP) is the most common congenital anomalies of the craniofacial complex, with a potentially life threatening nature or resulting in a range of physical, intellectual, visual or hearing impairment disabilities [6]. Research on CLP or orofacial clefts is important to determine the extent of the problem, identify unmet needs and contribute to health policy development [7].

In South Africa, the 2016 report of Statistics South Africa on mortality and causes of death based on notification, revealed that there were more than 2000 deaths in that year due to congenital malformations, eight of which were due to CLP [8]. However, mortality data can be misleading as they do not reveal the burden of caring for surviving individuals, quality of care received, health resource implications, and the socioeconomic well-being of affected individuals and families [7]. Importantly, there is a dearth of scholarly studies on CLP and a relative absence of a health policy focus on congenital anomalies in general, and on CLP in particular. Hence, the focus of my PhD is on the epidemiology and care provision for individuals with CLP in South Africa.

This chapter provides the background to, and context of, the research that was conducted as part of my PhD. Section 1.2 locates CLP within the global burden of disease and provides an illustrative profile of CLP. Section 1.3 provides a summary and critique of health policy on congenital anomalies, contextualises CLP within South Africa's burden of disease, and its health system. Section 1.4 presents the problem statement and the rationale for this PhD study. Section 1.5 describes the study aim and objectives. The concluding section 1.6 outlines the structure of the integrating narrative.

1.2 Global burden of disease and profile of CLP

1.2.1 Overview

Notwithstanding differences among geographical regions and population groups, the estimated worldwide average birth prevalence of all orofacial clefts is 9.92 per 10 000 live births (or 1 in 1000 live births) [9, 10]. The Global Burden of Disease (GBD) study has estimated that neural tube defects and cleft lip and palate accounted for 21 million disability-adjusted life years (DALYs) in the period from 1990 until 2010 [3, 11]. In low and middle-income countries, the burden of disease due to CLP has been estimated at 709 000 DALYs and 254 YLDs (years lived with disability) [12].

The World Health Organization (WHO) has emphasised the need for countries to address the burden of craniofacial anomalies (CFA), and the importance of research to generate new knowledge on these conditions [13-16].

The remainder of this section highlights the embryonic development of CLP, aetiology, genetics and clinical appearance of CLP.

1.2.2 The embryonic development of CLP

Development of the head and face is a series of events that occurs during the embryonic development of the fetus. In humans, the facial region develops from five facial processes, which consists of one frontonasal process (dividing into one lateral and a pair of medial prominences), two maxillary and two mandibular processes arising from the first branchial arch [17]. As shown in Figure 1.1, fusion of medial nasal and maxillary prominences gives rise to the lip and primary palate, while fusion of separate palatal processes arising from the maxillary prominence gives rise to the secondary palate and occurs later during embryogenesis. Disturbances in the fusion of these processes can result in facial clefts, depending when and where the failure of fusion occurs [17].

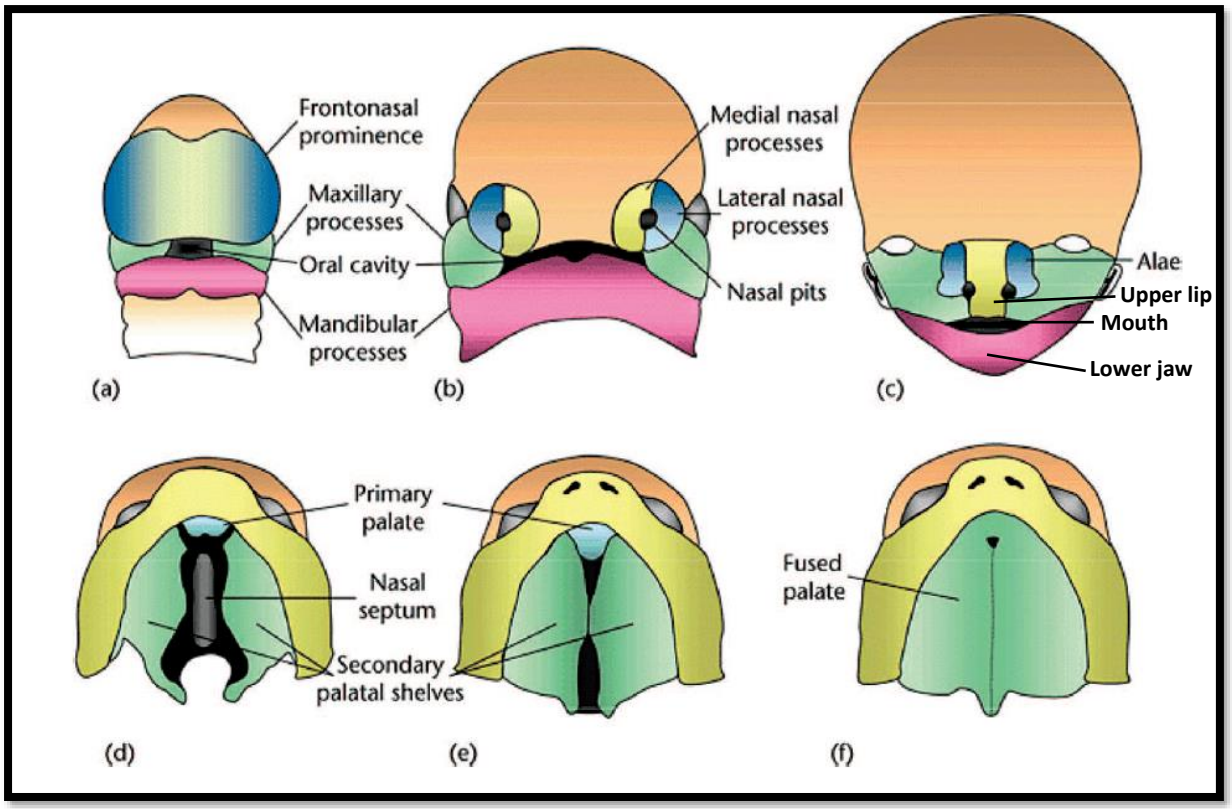


Figure 1. 1 Schematic diagrams depicting human craniofacial development and formation of the secondary palate:

(a) Formation of the frontonasal prominence, paired maxillary processes and the paired mandibular processes. (b) Formation of the nasal pits by the division of the frontonasal prominence into paired medial and lateral nasal processes. (c) The medial nasal processes have merged with one another and with the maxillary processes to form the upper lip and primary palate, whereas the lateral nasal processes form the alae of the nose. The mandibular processes fuse together to form the lower jaw. (d) The secondary palate develops from the maxillary processes as bilateral shelves that grow vertically down the side of the tongue during the sixth week of embryogenesis. (e) During the seventh week of embryonic development, the palatal shelves elevate to a horizontal position above the tongue, make contact with one another and begin to fuse. (f) Fusion of the secondary palatal shelves with one another and with the primary palate and nasal septum is completed by the tenth week of embryogenesis. [Adapted from [18] © (2009) John Wiley and Sons Ltd.]

1.2.3 Aetiology of orofacial clefts

The broad classification of orofacial clefts (OFC) has been syndromic and non-syndromic CLP or cleft palate (CP) and 70% of CLP and 50% of cases of CP are designated as non-syndromic [19], with the rest comprised of a wide range of malformation syndromes with known genetic, muscular and/or cellular aetiologies [20]. The interrupted development in utero of the orbicularis muscles has been reported as part of the phenotypic spectrum in individuals with CLP. This finding suggested the determination of subepithelial defects of this muscle in clinical setting, which would provide accurate recurrent risk estimates to relatives in CLP families [21]. The broad subdivision of OFC is associated with the distinct developmental origins of the lip, primary palate and the secondary palate (shown in Figure 1.2). Cleft palate may occur secondary to or independently from cleft lip (CL) [22, 23]. Although the embryology might be similar, cleft of the lip and that of the palate may occur as distinct entities [24]. Of the two, cleft of the palate is the more difficult to manage and has been the focus of investigations worldwide [25].

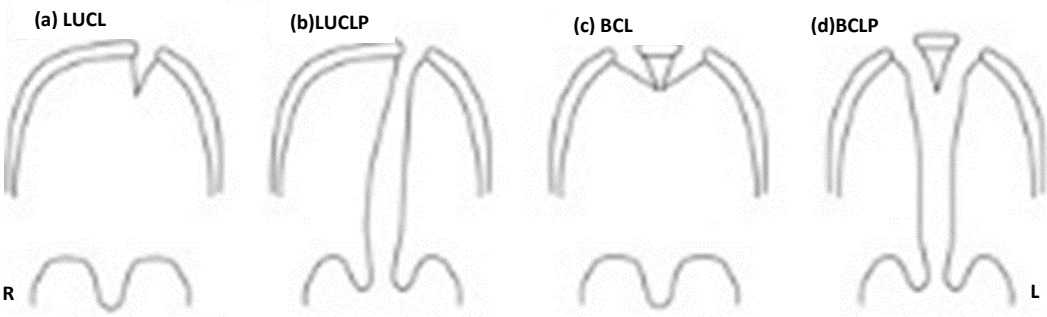


Figure 1. 2 Illustration of cleft types

(a) LUCL - unilateral cleft lip occurring on the left side. (b) LUCLP – unilateral cleft lip and palate occurring left side. (c) BCL – bilateral cleft of the lip occurring on both sides. (d) BCLP – bilateral cleft lip and palate occurring both sides.

The aetiology of CLP is multi-factorial and potential risk factors including smoking and alcohol during pregnancy, heredity, environmental and medical conditions have been reported [25-38]. Tobacco and alcohol are well reported as risk factors of clefting, with early pregnancy smoking and environmental tobacco exposure has been associated with bilateral cleft lip and palate [39]. A study in the United States has shown that orofacial clefts could be prevented with elimination of smoking in early pregnancy [32].

1.2.4 Genetics of orofacial clefts

The genetic basis of syndromic OFC is well-described [20], with emerging evidence of the contribution of genetic factors to non-syndromic CLP [40]. Epigenetic modifications may also play a role in the development of CLP [41]. A number of syndromes have been identified, of which OFC is the primary feature resulting from the mutation of a single genetic locus, chromosomal abnormalities or teratogens [42-45]. Family, twin, and epidemiologic studies have reported that orofacial clefts include a significant genetic component [44, 46, 47]. Twin studies of orofacial clefts consistently show higher concordance rates in monozygotic or identical twins compared to dizygotic or fraternal twins [48].

1.2.5 Clinical appearance of CLP

OFC anomalies may be unilateral (U) or bilateral (B) and involve the lip, the palate, or both. They may occur on the right side or left side of the face, hence terminology used include: right unilateral cleft lip (UCL), left UCL, right unilateral cleft lip and palate (UCLP), left UCLP, bilateral cleft lip and palate (BCLP) as shown in Figure 1.3.

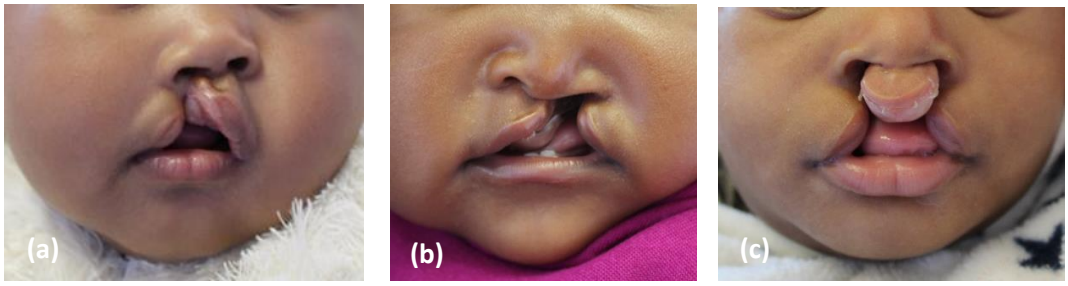


Figure 1. 3 Pictures of babies with cleft lip and palate types

(a) New-born baby boy with CL (b) A one-week-old baby girl with left CLP (c) A new-born baby boy with BCLP. (Pictures reproduced through courtesy of Professor MSP Sethusa, Department of Orthodontics, Sefako Makgatho Health Sciences University, South Africa).

The most commonly used clinical classification was presented by Kernahan and Stark, and further modified into symbolic striped 'Y' classification [49, 50]. A number of authors further modified the striped 'Y' classification [51-54]. Various classification systems for clefts have been proposed to date and recent reviews has recommended writing of cleft classification using longhand method [55] and more comprehensive description of the cleft including the severity [52]. The visual "Y" symbol classification of CLP shown in Figure 1.4, commonly used for accuracy of charting the exact embryological and clinical appearance of the defect. The Y symbol notation is a practical clinical tool to identify the type of cleft and the extent of involvement.

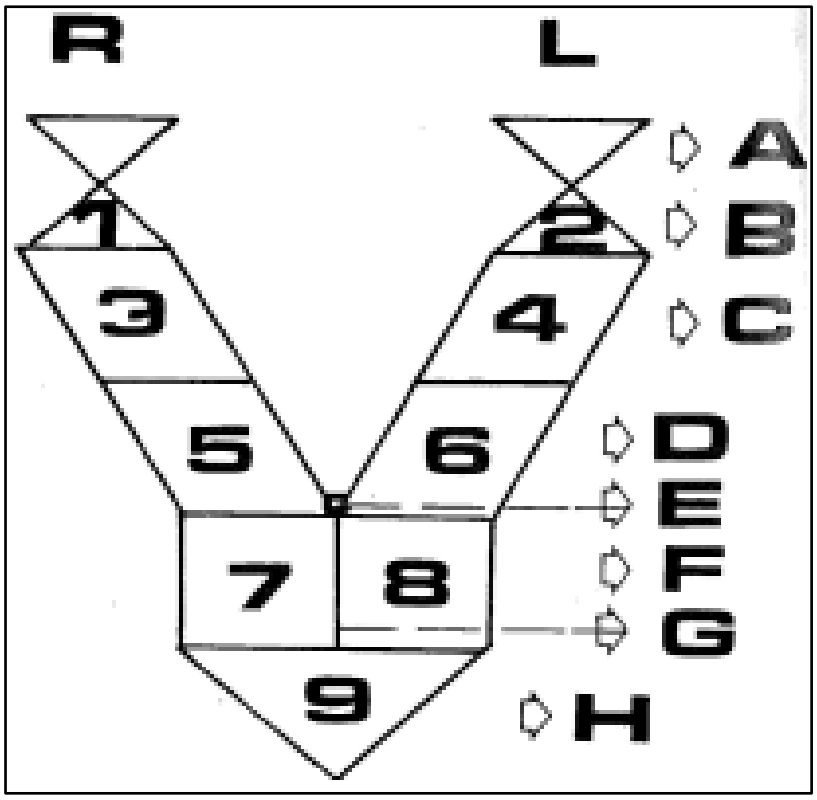


Figure 1. 4 "Y" Visual symbol for CLP

Source:[53]

R – right side; L – left side; A- nasal arch deformity; B- nasal floor (1&2); C-lip (3&4); D-pre-maxilla (5&6); E- incisive foramen; F-hard palate (7&8); G- vomer bone; H- soft palate (9)

1.3 CLP in South Africa

1.3.1 The Health Policy context

The 1997 White Paper for the Transformation of the Health System underscored the imperative of the new, democratic government to develop a unified health care system equipped to deliver quality health care to South African in an efficient and caring environment [56]. The White Paper mentions the prevention and management of genetic disorders as part of the responsibilities of the division of maternal, child and women's health [56]. However, health

policy developments on the prevention, surveillance and management of congenital anomalies have been fragmented, under-developed, and there has been insufficient investment in a coherent, national programme on congenital anomalies.

In 2001 the South African National Department of Health (NDoH) released *The Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disability* [57]. The policy consisted of a list of birth defects to be monitored, and the establishment of a national monitoring and evaluation system for genetic disorders and birth defects [57]. CLP was listed as one of the six listed, priority conditions for monitoring.

Subsequently, in 2005 NDoH published the *National Guidelines for the Care and Prevention of the Most Common Genetic Disorders, Birth Defects and Disabilities*. These guidelines outlined priority birth defects that are common in South Africa, and added talipes equinovarus, congenital infections and genetic deafness, blindness, physical handicap and mental retardation to the initial 2001 list of conditions [58].

The South African Birth Defects Surveillance System (SABDSS), developed in 1988 [59] and which collected data from 11 sentinel sites throughout the country [60], has since been stopped [61] due to a lack of funding, the untimely death of one of the champions, and the retirement of the other [62]. The Birth Defect Notification Tool (BDNT), introduced in 2006 by the NDoH to modify the SABDSS, has not been maintained and an analysis of the implementation of the tool concluded that data quality and quantity were poor [63].

Although all these developments on congenital anomalies are encouraging, there have not been any attention on congenital anomalies in the preceding 12 years (since 2006) and the extent of implementation of these policy guidelines across the country is unknown. There is a lack of

empirical information on CLP in particular, hence this PhD will begin to address this knowledge gap.

1.3.2 Burden of disease

South Africa is a constitutional democracy with an estimated population of 57.7 million in July 2018 [64]. The country has a complex, quadruple burden of disease consisting of : HIV&AIDS; a high burden of tuberculosis (TB) and other infectious diseases; high maternal and child mortality; high levels of violence and injuries; and a growing burden of non-communicable diseases (NCDs) [65].

In 2018 a special issue on the Maternal and Child Health Supplement reported on the improvements in maternal, neonatal and child quality of care in South Africa [66]. Significant progress has been made in reducing child mortality [67] whilst there is still room for improvements with neonatal deaths [68].

Notwithstanding declines in infant and child mortality rates in the preceding 10 years [69], the under-5 mortality is estimated at around 37 - 40 deaths per 1 000 live births and infant mortality rate is estimated at 27 - 33 deaths per 1 000 live births [67]. Diarrhoea, pneumonia and HIV infection remain the most important causes of death outside of the new-born period. Importantly, the proportion of deaths due to non-natural causes, congenital disorders and non-communicable diseases has increased [67].

1.3.3 Health care system

South Africa spends around 8.5% of its domestic product on health care, half of which is spent in the private health sector that covers around 17% of the population [70]. The public health care system provides care to the majority of the South African population [71]. The public

sector consists of a network of 10 central hospitals, linked to health science faculties, and a large number of regional, district and specialised hospitals [72]. The primary health care (PHC) system consists of a network of community health centres and clinics that provide mostly ambulatory care services. These PHC services, through clinics, are provided free of charge to everyone, while hospital services (secondary and tertiary levels) operate on a sliding tariff scale according to income. Children younger than six years and social grant beneficiaries are automatically exempt from paying for any public health services unless they are covered by private health insurance [73].

The South African Constitution states that: "every child has the right to basic nutrition, shelter, health care and social services" [74]. Since democracy, there have been numerous health system initiatives to improve child health, accompanied by significant investments [66, 73]. The most recent of these initiatives have been the introduction of the district clinical specialist teams (DCSTs). Evidence suggests that these clinical specialist teams have enabled innovations in care delivery, improved quality of care, achieved reductions in institutional deaths and fatality rates over time [75]. Notwithstanding these innovations and investments, implementation of these policy initiatives aimed at improving child health and care varies greatly across geographical settings [76]. A case study on the implementation of the integrated management of childhood illnesses programme found poor definition of elements of a service package for children, incompetent staff and fragmentation of service delivery mitigated against the delivery of quality of care, and successful implementation [76].

There is wide variation across the nine South African provinces in access to care, with more than one third of the population living in rural areas and served by less than 10% of doctors and less than 20% of nurses [77]. There are also a range of disability policies and programmes in South Africa, which are executed by various line departments. Programmes for children with

disability include developmental screening, free health care services, and inclusive and special education facilities [73].

Notwithstanding these encouraging developments, and progress made in improving access to health care, there is insufficient attention at both policy and practice levels on individuals with congenital anomalies in general, and CLP in particular, in part because of the predominant focus on infectious diseases.

1.4 Problem statement and study rationale

In South Africa, children are prioritised in Section 27 of the Constitution, which contains the Bill of Rights [74]. The PhD was conceived in response to several challenges. Firstly, the envisaged SABDSS was not implemented in the whole country and the sentinel surveillance system at the University of Cape Town was stopped. Hence, there is no updated epidemiological information on congenital anomalies or on orofacial clefts. Secondly, there is an outdated policy of 2005, which does not specify medical treatment and care, management of CLP in specialised health care centres, and/or human resource requirements, or interaction among members of the health care team. Thirdly, although community participation and involvement of families are implicit in legislation and policy documents, there is little information on the perspectives of parents or caregivers on having children with CLP, health care provision and social support services.

My PhD research study was premised on the following:

1. The dearth of empirical studies on:
    a) Epidemiology of CLP in South Africa
    b) Types of medical treatment provided in the public health centre

c) Health professional categories that are members of the CLP health care team, and the interprofessional collaboration among members of the team

d) Perspectives and perceptions of parents on CLP, health care provision, and support services.

2. The lack of comparative analysis of the healthcare provision for individuals with CLP in the South African public health sector.

3. Contribute to health policy and health care system interventions that will lead to improvements in the management of individuals with CLP in South Africa.

I have approached this thesis from a public health perspective, the foundation of which is epidemiology, namely the study of the distribution and determinants of diseases or disability in a population. This enables quantification of the extent of cleft lip and palate, to inform both health policy development and health service provision.

In the light of my specialty of orthodontics and research interest, I have combined the cleft lip and palate epidemiology study with the health systems research on care provision, interprofessional collaboration and parents' perceptions on treatment and care and social support services.

1.5 The study aim and objectives

The aim of this study was to determine the prevalence of, and care provision to, individuals with cleft lip and/or palate in South Africa in the public health sector.

The specific objectives of the study were to:

1. Describe the epidemiology of CLP in South Africa, specifically:
    a. Describe the demographic and clinical profile of CLP in the 11 specialised academic cleft lip and palate care centres.
    b. Estimate the prevalence of CLP in South Africa, and in each of the provinces.
2. Analyse the current approaches to care provision for CLP in the 11 specialised academic cleft lip and palate care centres:
    a. The point of entry for care of individuals with CLP;
    b. The range of care activities provided;
    c. The treatment protocols used;
    d. The internal referral system.
3. Measure the inter-professional collaboration among members of the CLP care team.
4. Explore the perspectives of parents or caregivers on:
    a. Experience of having a child with CLP;
    b. Health care provision and support services.

1.6 Structure of Integrating Narrative

Chapter 1: This chapter has introduced the global burden of congenital anomalies, provided an overview of CLP as the most common congenital anomaly of the craniofacial complex, and highlighted health policy development in South Africa on congenital anomalies, and service provision to children in general. The problem statement and study rationale were outlined, as well as the study aim and objectives.

Chapter 2: This chapter is a summary and critique of the relevant literature on CLP.

Chapter 3: This chapter describes the methods that were used in the PhD study.

Chapters 4- 7: Present the PhD study findings under the following headings:

Chapter 4: Epidemiology and clinical profile of cleft lip and palate.

Chapter 5: Healthcare provision to individuals with cleft lip and palate.

Chapter 6: Interprofessional collaboration among healthcare team members.

Chapter 7: Caregivers' perceptions of healthcare provision and support.

Chapter 8: This chapter provides an integrated discussion and conclusion that brings together all the elements of the PhD thesis, and propose key recommendations as well as areas for future research.

CHAPTER 2

Literature Review

2.1 Introduction

In this chapter, I have reviewed and critiqued the relevant literature, highlighting the knowledge gaps, especially those pertaining to the focus of my PhD thesis. Section, 2.2 contains the definition of terms used in the thesis. In section 2.3, I have reviewed the epidemiology of CLP. In section 2.4, I have reviewed the literature on the care and treatment of individuals with CLP. Section 2.5, I have reviewed the literature that focuses on parents or caregivers of children with CLP. The concluding section 2.6 summarises the gaps in the literature regarding CLP in South Africa and the knowledge contribution of my PhD thesis.

2.2 Definition of terms

Table 2.1 shows the definition of terms used in this PhD study.

Table 2. 1 Definition of terms

Terms	Description
Cleft lip with or without palate	A cleft lip (CL) or cleft palate (CP) is a fissure or opening on the lip (CL) or the roof of the mouth (CP) respectively caused by non-fusion of the body's natural structures that form before birth. These can occur as single entities or occur together as cleft lip and palate (CLP) [17].
Feeding plate	The feeding plate is an appliance made from dental acrylic which is used to obdurate the cleft and restores the separation between oral and nasal cavities. It facilitates feeding, reduces nasal regurgitation and the incidence of choking [78].
Inter-professional collaboration	Inter-professional collaboration (IPC) is defined as: "multiple health workers from different professional backgrounds working together with patients, families, caregivers, and communities to deliver the highest quality of care" [79].
Multi-disciplinary team	Sometimes referred to as "inter-disciplinary" team - A group composed of members from different healthcare professions with specialised skills and expertise to achieve specific health care objectives [80].
Orthodontic treatment	Treatment provided to align and straighten abnormal teeth which may be a result of tooth irregularity and/or abnormal jaw relationships, using removable or fixed orthodontic appliances [81].
Orthognathic treatment	This is the surgical correction of abnormal relationships of the face due to structural disorder of the jaws, abnormal teeth alignment and congenital conditions like cleft palate and other craniofacial abnormalities. Bones of the jaw are cut and re-aligned, then held in place with either screws or plates to heal [81].
Plastic surgery for CLP	Type of surgery used to correct CLP abnormal development and are meant to restore function to the lips and mouth along with producing a more normal appearance [82].
Pre-surgical orthopaedics	Also known as neonatal infant orthopaedics, presurgical infant orthopaedics or nasal-alveolar moulding; this technique is used to approximate the maxillary, alveolar, and nasal tissues of an infant with CLP [83]
Speech therapy for CLP	It refers to treatment of articulation disorders related to atypical anatomy and structural defects associated with CLP [84].

2.3 The epidemiology of CLP

2.3.1 Global prevalence of CLP

The prevalence rates for CLP are sometimes expressed per 10 000 live births or per 1000 live births. For the ease of comparisons in this section only, I have chosen to express the rates per 10 000 live births and not as they appear in the relevant literature.

Worldwide, cleft lip and palate is the most common congenital anomaly of the cranio-facial complex, with an estimated global prevalence that ranges from 1.35 to 25.31 per 10 000 live births [4, 6, 16]. Existing evidence suggests that the prevalence of orofacial clefts (OFC) is influenced by "race" or ethnicity, with the highest rates amongst Asian populations (8.2–40.4 per 10 000 live births), intermediate rates amongst Caucasians (9.0–26.9 per 10 000 live births), and the lowest rates amongst African populations (1.8–16.7 per 10 000 live births) [85].

Figure 2.1 provides an overview of the estimated CLP prevalence rates for different geographical regions [4, 86-89]. The prevalence rate in North America ranged from 3.68 to 11.77 per 10 000, with the highest found in Canada British Colombia, Canada Alberta and lowest in Canada National. In South America, the rate ranged from 7.83 to 22.94 per 10 000, with the highest in Bolivia and the lowest in Venezuela. The prevalence in Europe ranged from 4.08 to 13.94 per 10 000, with the highest in Germany and the lowest in Spain. In Asia it ranged from 3.37 to 16.04 per 10 000, with the highest in Japan and the lowest in China. In Australia the prevalence rate was 8.83 per 10 000. In Africa the rate ranged from 2.0 to 17.0 per 10 000 [4, 86-89] .

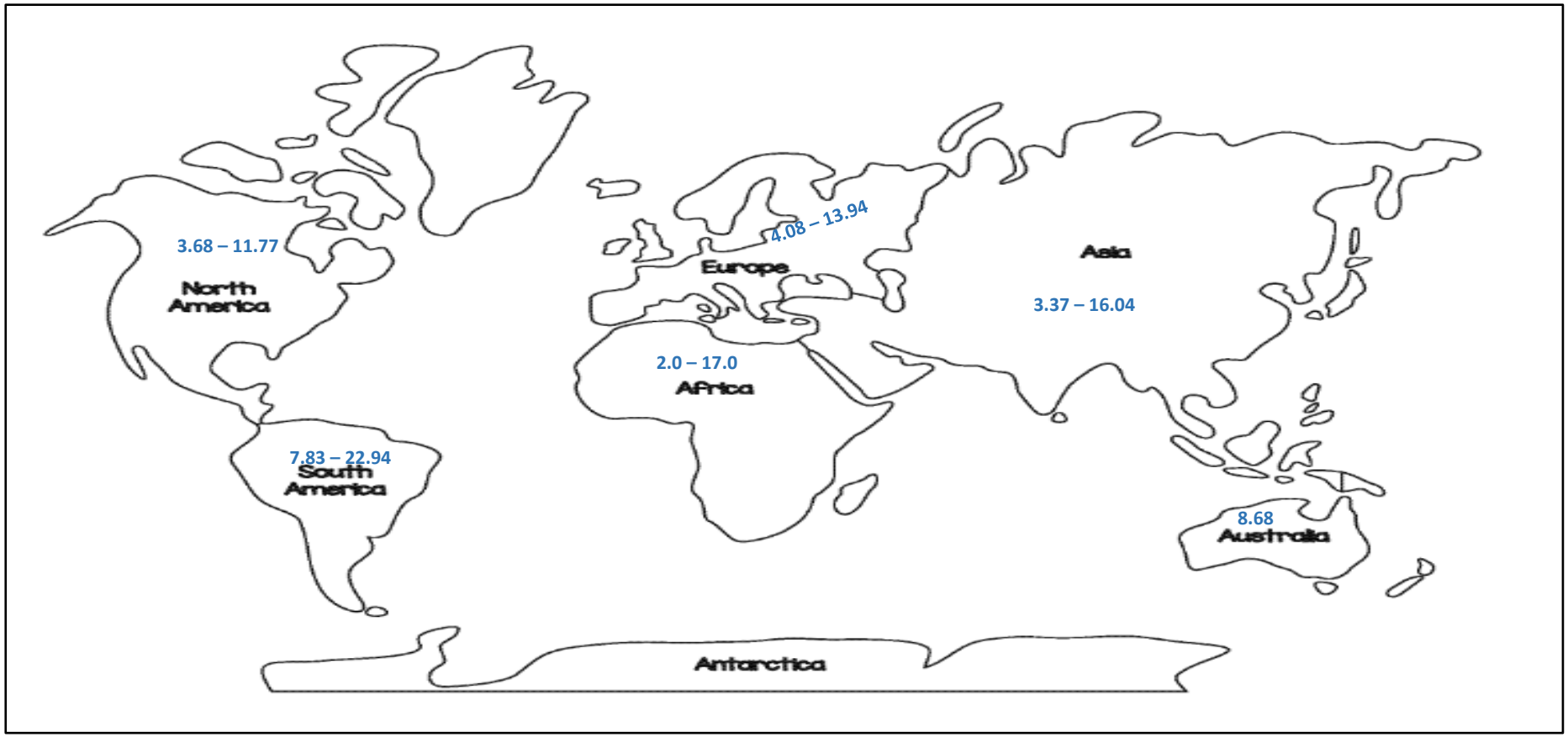


Figure 2. 1 Estimated CLP prevalence rates in different geographical regions of the world per 10 000 live births

Source for map: http://getdrawings.com/world-map-for-drawing [90]

Sources of prevalence rates: [4, 86-89].

In African, prevalence rates ranged from 0.2 to 1.7 per 1000 live births, and are estimated from hospital-based data, because of lack of congenital anomalies surveillance system, and it. The lowest prevalence was reported in Ethiopia at 0.2 per 1000 live births [86], followed by Nigeria at 0.5 per 1000 [87], Uganda at 0.8 per 1000 [88] and the highest in Kenya at 1.7 per 1000 [89]. A 2011 community household survey in South East Ghana reported a prevalence of 6.3 per 1000 [91], which was much higher than the above reports in other parts of Africa. However, a major limitation was that they used community self-reporting rather than clinical examination based on recognized cleft classification system [49, 51-54] and description of orofacial cleft phenotypes [44].

In South Africa, CLP is one of the five most common congenital anomalies [92]. Three decades ago, studies on CLP prevalence were conducted in Cape Town [93, 94], Johannesburg [95] and Pretoria [96-98]. These studies found estimated prevalence rates that ranged from 0.1 to 0.4 per 1000 live births. In 2018, a hospital in Pretoria analysed the profile of patients with CLP [99], and found that 47% of patients presented with unilateral cleft lip and palate (UCLP), 23% presented with bilateral cleft lip and palate (BCLP), and 30% presented with cleft palate (CP). A major critique is that the prevalence estimates were done 30 years ago, and only focused on three cities in South Africa. This PhD is one of the first studies to focus on all the CLP specialised care centres in South Africa, thereby obtaining a more representative sample of individuals with CLP.

2.3.2 CLP Gender differences

Various studies have found that clefts, especially cleft lip and palate are more common in boys [100-103], because there is a higher intra-uterine mortality rates for girls with cleft lip and palate [4]. CP is more common among girls [41, 87, 104]. Isolated cleft lip (CL) has also been

reported to have a female predominance, but with good survival rates. However, isolated CL may be associated with other anomalies [4].

2.3.3 CLP Laterality

Clefts occurring on one side of the mouth, left side or right side, are called unilateral clefts and those occurring both sides, bilateral. The proportion of bilateral cases is 10.3% for CL and 30.2% for CLP. Amongst unilateral cases, 36.9% of CL and 41.1% of CLP occur on the right side, suggesting that unilateral cases of CLP occur more frequently on the left [10, 105-108]. This left side predominance has been linked to developmental interruption of the left palatal shelf taking longer to rotate into the horizontal position in utero [109], suggesting lower arterial pressure on the left side compared to the internal carotid artery on the right side [110].

2.4 Treatment of individuals with CLP

Children born with CLP require treatment from different health care professionals [111-113], because of the complexity of the congenital anomaly. Such treatment begins at birth and extends to early adulthood. A multidisciplinary team of experts consisting of geneticists, maxillo-facial surgeons, otolaryngologists, orthodontists, paediatricians, plastic surgeons, paediatric dentists, psychologists, registered nurses, social workers, speech and language therapists has been proposed [114-116]. These teams are available in high-income countries (HICs) [114, 117] where clearly defined team members provide evidence-based treatment including monitoring and continuous evaluation of outcomes in patients with CLP.

It has been reported that only 20% of children with CLP in the world have access to comprehensive CLP treatment that involve all relevant members of the team, the majority of whom are in low and middle-income countries (LMICs) in Africa, Asia and South America

[118, 119]. In LMICs, outreach programmes operated by non-governmental organisations (NGOs) have assisted in improving the services for individuals with CLP [120-122]. Together with support groups that include parents of the affected children, these NGOs [123] have played a role in the ongoing management of OFC. However, the majority of LMICs, including South Africa, lag behind in terms of epidemiological data on CLP, defined treatment protocols, access to care, multi-disciplinary team approach, inter-centre collaborations and evaluation of treatment outcomes [13, 114, 124].

In South Africa, there is a dearth of studies on the modalities of treatment at the various CLP specialised academic care centres, the composition of health care teams, and the extent to which these teams work collaboratively. This is one of the first studies to conduct a comparative analysis of CLP treatment in the public health sector, and to measure interprofessional collaboration (IPC), using a validated framework.

2.5 Parents' perspectives of CLP care and treatment

A birth of a child with CLP is accompanied with mixed emotions for the parents, in particular the mother who has been carrying the child for nine months. Most studies have shown that the majority of mothers did not know that their child would be born with a cleft. Although all the parents in studies indicated that they would not have chosen a termination of pregnancy [125-127], pre-natal diagnosis or psychological counselling soon after birth would provide assistance for parents in dealing with the shock of having a baby with a congenital anomaly [128, 129].

The face is the most conspicuous part of the body, therefore seeing a child's face with a cleft condition tend to evoke strong parental reactions and emotions. Parents of children with CLP

have different beliefs regarding the causes of the deformity [130], and they seek treatment from all possible sources including traditional healers [131]. Having a child with CLP is a life-changing event, with the parents in great need of support from health professionals. In the first instance, parents need basic information which includes practical information on feeding, the need for long-term treatment and care, and reassurance that the defect is not a disability and it can and will be managed [47]. Information pamphlets on CLP can be a useful tool for disseminating information to parents [132]. The parents also have to be counselled and prepared for possible reactions from relatives, friends and the general public, and how to manage such incidents [133]. Parents also need to be involved in the ongoing and long-term care of their children.

I considered it important in this PhD to obtain a comprehensive picture on CLP. There is little information available on the perspectives of parents having children with CLP and their experiences of health care provision and/or social support services. These perspectives are critical in developing health policies and/or programmes that are responsive to their needs and those of their children with CLP.

2.6 Summary of Literature Review

Table 2.2 summarises the identified gaps in the literature, and the contribution of the PhD in addressing some of the gaps.

Table 2. 2 Summary of literature review

Study Objective	What is known	Knowledge Gaps	Scholarly contribution of the PhD
Describe the epidemiology of CLP in South Africa	• Global prevalence estimates • Prevalence of CLP in cities of Cape Town, Johannesburg and Pretoria	• Dearth of studies on CLP epidemiology in LMICs • Studies in South Africa conducted more than 30 years ago and predate democracy	• One of the first studies on epidemiology of CLP in all specialised centres in public health sector in South Africa • Analysis of clinical profile of CLP in public health sector
Analyse the current approach to care provision for CLP in 11 care centres	• Children with CLP require immediate medical treatment to ensure survival and avoid complications • Treatment and care enhanced by availability of a multidisciplinary team of health professionals • Good practice to have clinical protocols for the treatment of CLP • Globally, only 20% of children with CLP have access to comprehensive treatment	There is dearth of studies on: • Modalities of treatment and care provision for CLP • Availability of treatment protocols • Description of CLP team	Description and comparative analysis of health care provision for CLP in South Africa Types of treatment provided at each CLP centre Analysis of treatment protocols
Describe the collaboration of the CLP care team in the 11 care centres	• Management of individuals with CLP is enhanced by inter-professional collaboration (IPC) amongst different health professionals • Various advantages of IPC in clinical care, and positive patient and health professional outcomes	• Dearth in studies that assess IPC in LMICs in general, including South Africa • Lack of empirical assessment of IPC in CLP treatment and care	One of the first studies to: • Measure IPC in CLP treatment and care • Use a validated instrument on IPC
Describe the parents or caregivers' views on having a child with CLP and their perceptions of healthcare provision and support services	• Parents experience mixed emotions of shock, anxiety, etc., which are universal regardless of level of income of the country • Parents should be involved in the ongoing treatment and care of their children with CLP	Dearth of studies on parents' perceptions of care provision for their children with CLP, especially in LMICs	One of the first studies that explored the perceptions of caregivers on having children with CLP in South Africa, experiences of health care provision, and of social support services

CHAPTER 3
Methodology

3.1 Introduction

This mixed methods PhD study has combined quantitative and qualitative research methods, which are summarised in this chapter. It is divided into nine sections: Section 3.2 describes the conceptual framework that I have used in my study. This is followed by the cross-cutting methodological elements in Section 3.3: the study setting, the study population, and the ethical considerations and approvals. Section 3.4 presents the four distinct, but inter-linked components of this study conducted to achieve the different study objectives. In sections 3.5 to 3.8, I have summarised the methods of each of the four studies. Section 3.9 describes the study limitations and how these were addressed, and the steps taken to avoid potential bias. The concluding section (3.10) highlights the strengths of my PhD study.

3.2 The conceptual framework

3.2.1 Brief overview of frameworks

There are several models that focus on the management of chronic diseases (now called non-communicable diseases), but these models were not designed for the management of children with cleft lip and/or palate [134-136]. In 2016, the WHO reviewed integrated care models, ranging from individual care, disease-specific and community-based models. The review highlighted that any integrated care model development should be context-specific, and it should take into account the unique needs and characteristics of the population it aimed to serve [137].

The Chronic Care Model (CCM) was developed in the mid-1990s by the MacColl Centre to improve chronic illness management, and consists of the community, the health system, self-management support, delivery system design, decision support and clinical information systems. These elements foster productive interactions between informed patients who take an active part in their care and providers with resources and expertise [134]. The elements identified in the CCM are applicable in CLP integrated care. Furthermore, the interactions between the informed patients, a child with CLP and their parents in this PhD, and care provider (the multidisciplinary team in the case of CLP) are also essential for CLP care to produce evidence-based outcomes [114, 138, 139].

The Model of Child and Family Centred Care (C&FCC), was developed by the Hospital for Sick Children (SickKids), a paediatric academic health sciences centre in Canada. The model consist of systems, elements and outcomes, and includes the child, the family and the community and health system. The child is at the core, a priority in everything that is done. The family, adjacent to the child, illustrating the centrality of the family to the child's life. The community and health system is where SickKids interacts beyond the organisation, locally, nationally, internationally, to inform, shape, and support health-care service delivery. The elements included respect, communication and partnership as three essentials in the process of delivering for C&FCC. The model outcomes includes promoting optimal health, ensuring patient safety, achieving health equity and maximizing the patient experience [135]. This model is applicable for CLP since it places the child in the centre of care coordination.

The House of Care (HoC) is a framework developed for primary care in the United Kingdom. It uses the metaphor of a house to describe the components that need to be in place to make coordinated personalised care planning a reality, especially for individuals with long-term conditions. It highlights the importance of clinicians working together to determine and shape

the support needed to make the patients live well with their condition. The house with its walls, roof and foundations is used as a metaphor and a checklist, highlighting the importance and inter-dependence of each element – if one element is weak or missing the service is not fit for purpose [136]. Similarly, individuals with CLP require coordinated multidisciplinary collaboration, even though this condition is not chronic but a congenital anomaly. Because the treatment is long, as it start from birth to adulthood, it also requires inter-dependence of all elements for its success. This model, together with aspects of the other models, was the most relevant for the care of children with CLP, and was hence adapted for use in this PhD.

3.2.2 Description of my conceptual framework

In this PhD study, I have used the internationally accepted principles and concepts of the WHO on integrated models of care [137], and adapted analytical framework drawn from the CCM [134], the C&FCC model [135] and the HoC model [136]. My conceptual framework, called Cleft Lip and Palate Ekhaya Lethu (House of Care) is shown in Figure 3.1.

CLP integrated management should be initiated as soon as a child born with OFC presents at the appropriate level of care. The point of entry for care could be through a specialised CLP centre or any other health facility. A health care provider (HCP) at that level should identify and classify the cleft, notify on clinical grounds and then send specimens to the laboratory for genetic confirmation of the diagnosis if necessary. The immediate need following the birth of a baby with CLP is appropriate care, treatment and psychosocial support to the parents and/or family. This is followed by the feeding needs for the baby, which should be established due to lack of suckling and the cleft causing communication between the mouth and nose. The Ekhaya Lethu shows the analogy of a secure house with seven components and attributes, that should interact in providing the age-specific treatment and care for the comprehensive management of children with CLP.

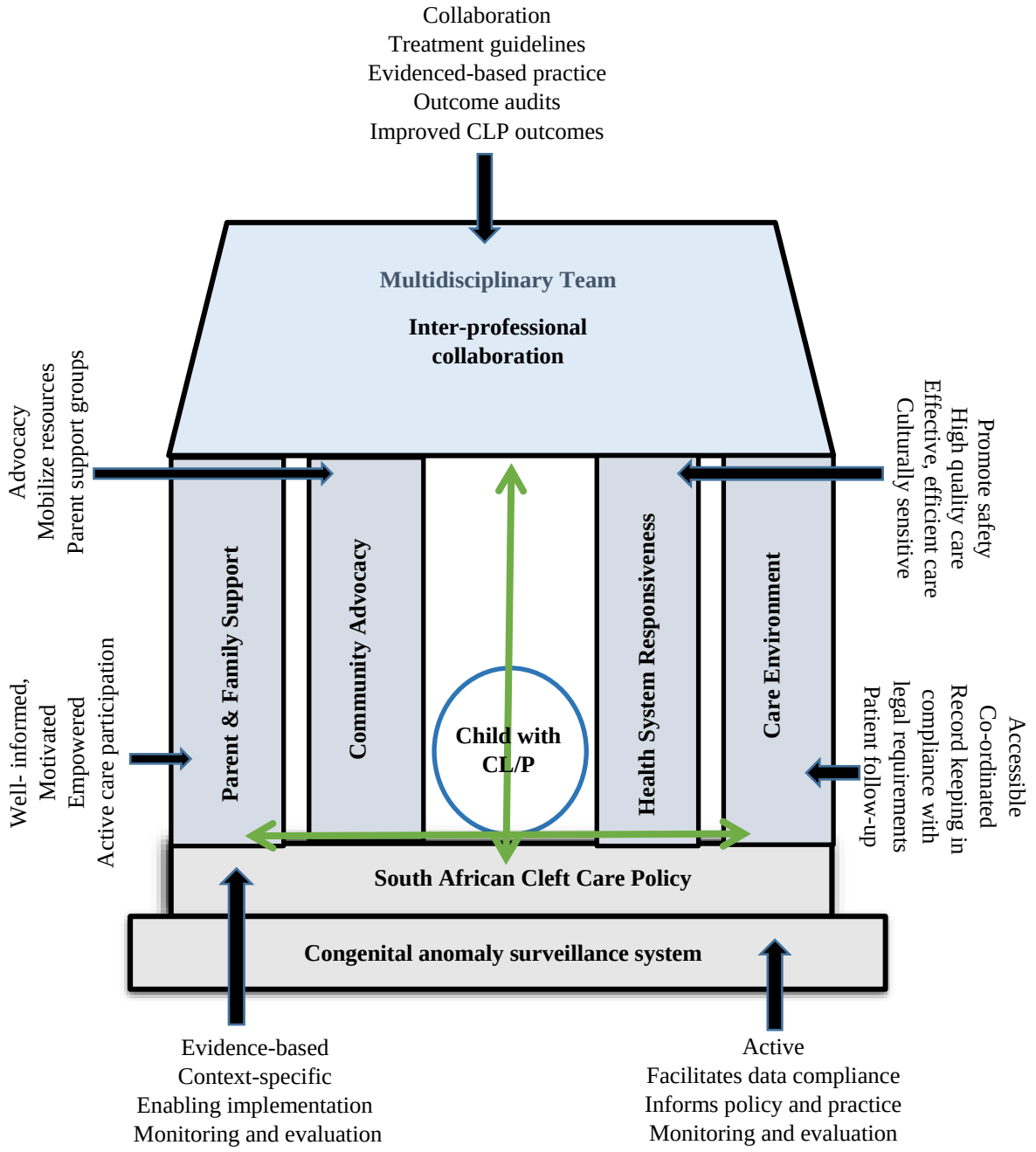


Figure 3. 1 Cleft Lip and Palate Ekhaya Lethu Conceptual Framework

Source: Adapted from: House of Care Model [136]

My Ekhaya Lethu Conceptual Framework uses the analogy of a secure house with seven components and attributes, described below.

The ***foundation*** consists of a population-based congenital anomaly surveillance system and national policy framework– together these two components will be context specific to South Africa, and inform health policy and practice on congenital anomalies

The ***four walls*** give structural support to the house, and each wall is represented by the four pillars:

- *Parent and family support* – Information about CLP, screening and genetic counselling, treatment information as the treatment starts from birth to 18 years (and beyond), active participation in care, reassurance and emotional support.
- *Community advocacy*– environment within which a child with CLP is born, public awareness to reduce stigma and improve acceptance, mobilization of resources and parent support groups.
- *Health system responsiveness* – this includes safe, high quality care that is effective and efficient, while being culturally sensitive at the same time.
- *Care environment* – availability of resources, appropriate access, training of healthcare professionals that are members of the CLP team, information, supportive treatment.

The ***roof*** it is protective and consists of a multidisciplinary team of specialised healthcare professionals (HCPs), who work together in a coordinated manner, collaborate with one another to ensure the provision of the necessary treatment to a child with CLP as well as support and counselling to parents.

The ***contents of the house*** – the child with CLP together with the parents or caregivers – they occupy this space with the assurance that the house is well secured by the roof, the pillars and standing on a strong foundation.

3.3 Cross-cutting methodological elements

3.3.1 Study setting

The study was restricted to the public health sector in South Africa, as it provides health care services to the majority of the population [140]. The study setting consisted of all 11 CLP care centres situated in six of South Africa's nine provinces (Fig 3.2). These centres are specialised academic centres located at central hospitals, which in turn are attached to health science faculties. Multidisciplinary teams of health professionals are available at these centres to provide specialised treatment and care.

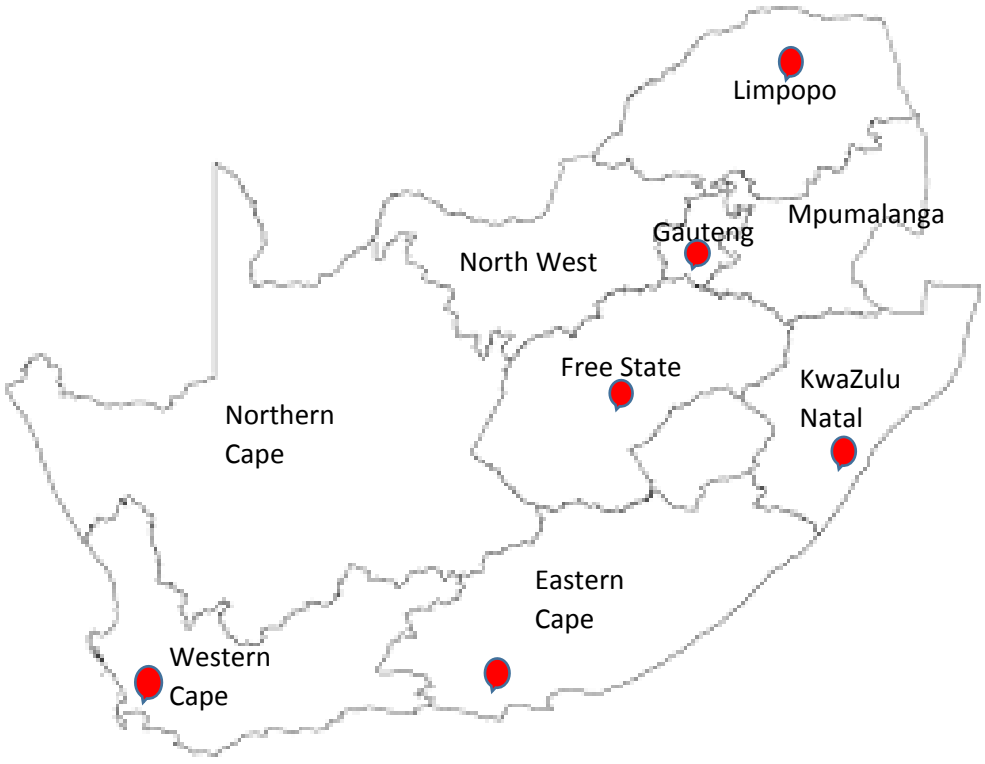


Figure 3. 2 Map of South Africa showing the study provinces

Source: Getdrawings.com [141]

In Gauteng Province, five CLP care centres were included in the study: Charlotte Maxeke Johannesburg Academic Hospital, Chris Hani Baragwanath Hospital, Dr George Mukhari Hospital, Pretoria Dental Hospital and Steve Biko Academic Hospital. In the Western Cape Province, Tygerberg and Red Cross War Memorial Children's Hospitals were included. The one centre in each of the following provinces was included in the study: KwaZulu Natal Province - Inkosi Albert Luthuli Central Hospital; Eastern Cape Province - Nelson Mandela Academic Hospital; Free State Province - Universitas Hospital and Limpopo Province – Polokwane/Mankweng Hospital Complex.

3.3.2 Study population

The study population consisted of all individuals born with CLP in South Africa who sought treatment in the 11 CLP care centres from 1 January 2013 to 31 December 2014. In light of the potentially life threatening nature of CLP, I assumed that all individuals with the condition would seek care or be referred to these specialised centres.

3.3.3 Ethical considerations

Ethical approval (M1501536) was obtained from the Human Research Ethics Committee (HREC) (Medical) of the University of Witwatersrand, Johannesburg (Appendix 1) to conduct this PhD study. Approval for the research was also obtained from the Provincial Departments of Health, Chief Executive Officers of the academic hospital centres and the heads of hospital clinical departments. Informed consent was sought from all participants who completed the survey. There was no coercion and participants were given the opportunity to withdraw from the study at any time without prejudice. Confidentiality and anonymity were maintained throughout the study.

Ethical matters considered and how they were addressed, are outlined below:

(i) Obtaining free, informed and voluntary participation of potential participants

All participants received an information sheet (Appendices 2 and 3), and informed consent was obtained. I stressed that there are no direct benefits of participation. I emphasized that participation is voluntary and that there would be no negative consequences for refusal to participate.

(ii) Potential of my position as a Specialist Orthodontist and Adjunct Professor to influence participation in the survey

At the time of my PhD, I was a Specialist Orthodontist and an Adjunct Professor in the Department of Orthodontics, School of Oral Health Sciences at the University of the Witwatersrand. To ensure that this did not have an influence on the participation of HCPs and vulnerability of the parents of children with CLP, I drew a clear distinction between my position and the research undertaken by me as a PhD student, and emphasised that it was not part of my official work. The information sheet (Appendices 2 and 3) only referred to my status as a PhD student and did not refer to my official position.

(iii) Privacy and confidentiality for the medical record review

The Principal Investigator (PH) is registered with the Health Professions Council of South Africa (HPCSA), and understands the importance of confidentiality. The patients' records were given unique codes and the individual details that could be used to identify them were not recorded in the record review form to ensure confidentiality and anonymity in the management of data and reporting of study findings.

(iv) Privacy and confidentiality for study participants

The survey questionnaires were completed anonymously. Questionnaires were coded and no personal identifiers were obtained to ensure confidentiality.

The parents of children with CLP could be regarded as the most vulnerable group of study participants. Each interview was conducted in a private room. I maintained professionalism

and courtesy in obtaining information from them. They were only interviewed after informed consent was obtained.

All captured data were given unique codes, and personal details were not recorded to ensure anonymity, and could not be used to identify participants in the management of data and reporting of study findings.

(v) *Securing the information after data collection and maintaining confidentiality of records*

Responses from the records review (study 1) and three questionnaires (studies 2 -4) were stored on a password secure, protected computer, with only the researcher who had access to this password.

3.4 Overall methodology approach

This PhD thesis consisted of four distinct, but inter-linked components, based on the objectives of this PhD described in Chapter 1.

- **Study 1** – Description of the demographic and clinical profile of CLP in the 11 care centres and estimated prevalence of CLP in South Africa, and each of the provinces.
- **Study 2** – Analysis of the current approach to care provision for CLP in the 11 centres.
- **Study 3** – Measurement of the inter-professional collaboration among the members of the CLP care team at the 11 centres.
- **Study 4** - Perspectives of, and support services available to parents/caregivers of children with CLP.

Table 3.1 shows the linkages of the study objectives, the study methods and data collection tools to the research outputs. Table 3.2 describes the four study components.

Table 3. 1 Overview of the study objectives, methods, data collection tool and research outputs

Objective	Methods	Data collection tool	Research papers
1. Describe the epidemiology of CLP in South Africa and estimate the prevalence	Retrospective review of two years (2013-2014) CLP data from the 11 care centres	A specific designed data collection form was used The form was piloted prior to implementation	Epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa (Chapter 4)
2. Analyse the current approach to care for CLP in the 11 care centres	A survey of CLP leaders in the 11 care centres to evaluate the point of entry for individuals with CLP, treatment protocols, continuum of care and the internal referral systems	A structured interview schedule was specifically designed for the survey The questionnaire was piloted prior to implementation.	Comparative analysis of healthcare provision to individuals with cleft lip and palate at the specialized academic centres in South Africa (Chapter 5)
3. Measure the inter-professional collaboration of the members of CLP care team in each of the 11 care centres	A survey of the CLP team members in the 11 care centres was conducted to assess how they work together as a team	A structured questionnaire obtained from the Registered Nurses' Association of Ontario (RNAO) was used in its original form, with added questions on background and demographic characteristics of providers The questionnaire was piloted prior to implementation	Inter-professional collaboration among cleft lip and palate healthcare team members in South Africa (Chapter 6)
4. Determine views of, support services available to parents of children with CLP	Interviews with parents and/or caregivers of children with CLP attending the 11 care centres	A structured interview schedule was specifically designed for the study The interview schedule was piloted prior to implementation	**"**People look and ask lots of questions": caregivers' perceptions of healthcare provision and support for children born with cleft lip and palate (Chapter 7)

Table 3. 2 Overview of the study components

Component	Study 1	Study 2	Study 3	Study 4
Study Population	All individuals with CLP who accessed services from 1 January 2013 to 31 December 2014	CLP Team leaders	HCPs at the CLP care centres	Parents of children with CLP who were present at the care centres on the fieldwork day
Sampling	Records from 1 Jan 2013 to 31 Dec 2014	Team Leader or Senior Consultant in CLP clinic	All HCPs who were members of the CLP team, and present on the day of survey	5 – 10 parents of children with CLP, older than 3 months, present on survey day, and selected purposively
Sample Size	699 Records	11 Team Leaders	52 HCPs	79 caregivers
Data collection	Record review form	Self-administered questionnaire	Self-administered questionnaire	Structured interview schedule
Data Analysis	Quantitative analysis with STATA® 13	Quantitative analysis with STATA® 13 and descriptive content analysis	Descriptive analysis and demographics with STATA ® 13. The explanatory variables analysed with ANOVA	Thematic content analysis for interviews and demographics with STATA ® 13
Integrated Narrative	Triangulation and integration of the findings of the various studies			

3.5 Study 1

The methods used in this study are summarised below with further details provided in Chapter 4.

3.5.1 Study population

The study population consisted of all individuals with CLP that visited the 11 care centres from 1 January 2013 to 31 December 2014.

3.5.2 Sampling

The sample size included medical/clinical records of all individuals with CLP who attended the care centre during the two- year study period.

3.5.3 Record review form

I developed a record review form (Appendix 4) to extract socio-demographic, clinical information and treatment history from the hospital clinical records of individuals with CLP.

3.5.4 Piloting record review form

The record review form was piloted using 10 CLP records in one care centre for a year outside the study. During the pilot study, I found that due to the high level of incomplete information in the records, some of the variables including the place of birth of a child with CLP (home delivery/ primary health centre, hospital); method of delivery at birth (caesarean/vaginal/ suction assisted); delivery at term (i.e. 9 months of pregnancy); history of alcohol during pregnancy and history of smoking during pregnancy were missing. The Y-symbol classification [53] was also not used for clinical classification of clefts since the pilot records did not utilize it. Therefore, the record review form was revised to exclude these variables. Figure 3.3 shows the steps in developing the record review form and approach to data collection:-

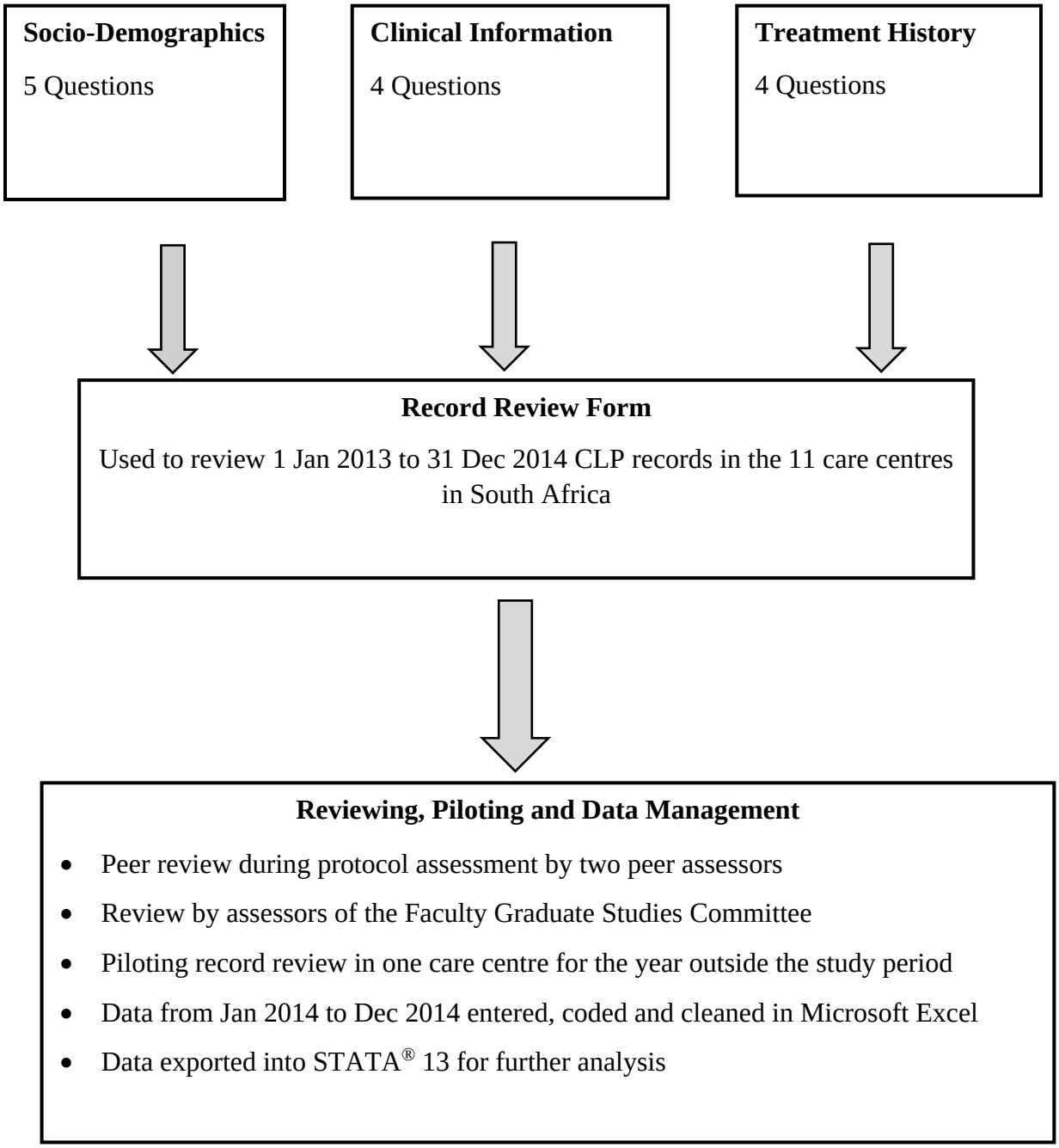


Figure 3. 3 Development of record review, data collection, capturing and data management for Study 1

3.5.5 Data collection, capturing and data management

I reviewed and extracted all the data from the clinical records of all patients with CLP for the period, 1 January 2013 to 31 December 2014. I cleaned and coded the data in Microsoft Excel. The data were password protected on a computer that only I had access, thereby ensuring confidentiality. Data were exported into STATA® 13 for further analysis.

3.6 Study 2

The methods used in this study are summarized below with further details provided in Chapter 5.

3.6.1 Study population

The study population consisted of all CLP team leaders in the 11 care centres.

3.6.2 Sampling

The sample size included 11 CLP team leaders.

3.6.3 Development of data collection tool

I could not find a standardised tool to determine the current approach to care provision in the 11 care centres. I therefore designed an information sheet (Appendix 2) and a structured interview questionnaire in English (Appendix 5) for use in the survey. English is one of the 11 official languages of South Africa and is the main language used for official business. I assumed that all of the CLP team leaders had a good working knowledge of English and no translations were made of the questionnaire. The different components of the questionnaire and approach to this study component are shown in Figure 3.4.

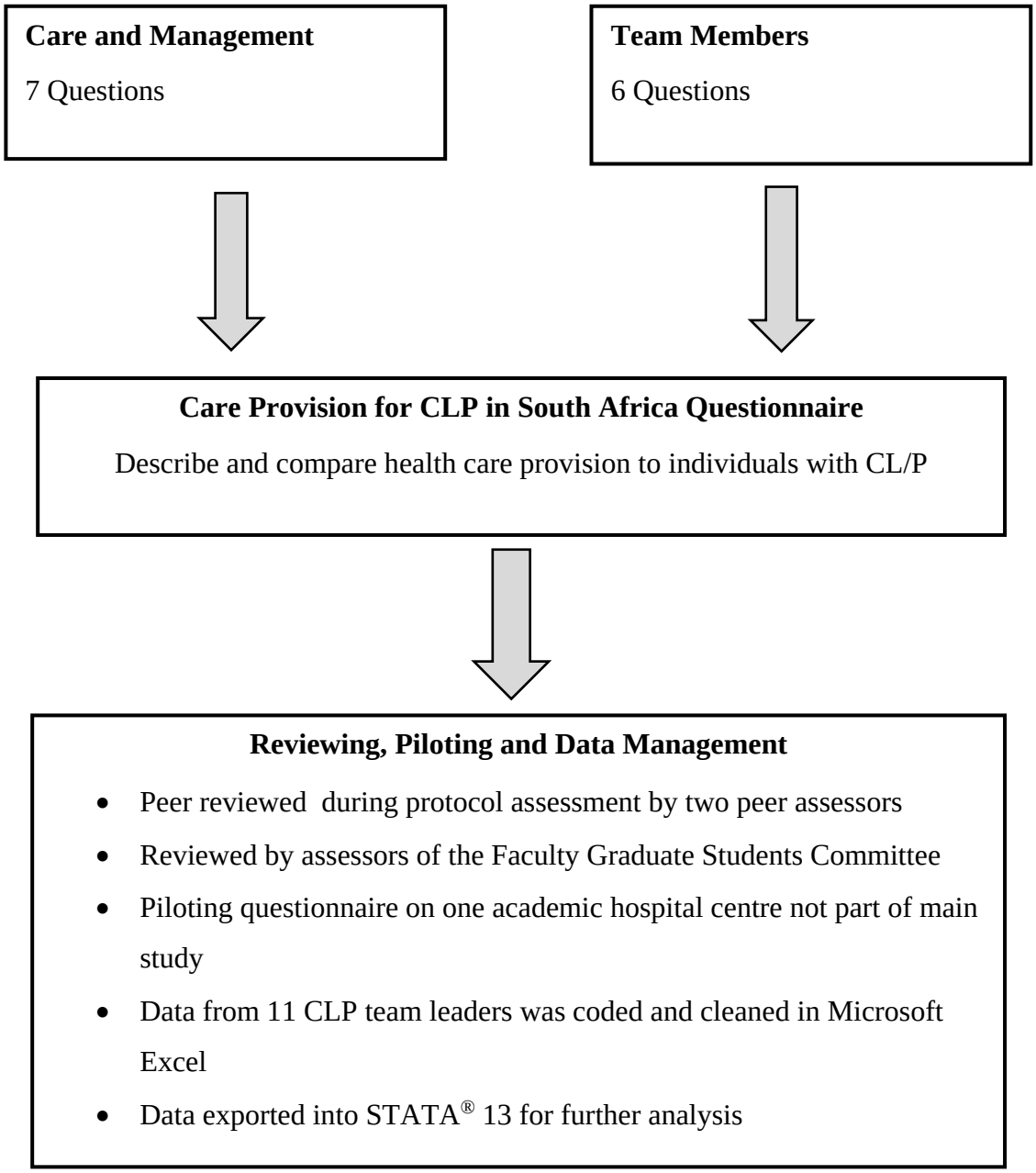


Figure 3. 4 Development of care provision questionnaire, data collection, capturing and data management for Study 2

3.6.4 Piloting of data collecting instrument

The questionnaire was piloted in one care centre not included in the main study population to determine the clarity of questions and time taken for completion. This centre was excluded from the main PhD study because it re-commenced treatment and care for individuals with CLP in 2014. The questionnaire took an average of 20 minutes to complete and no changes were deemed necessary after the pilot study. The data collected in the pilot study were excluded from the main study.

3.6.5 Determining reliability and inter-item correlation

Cronbach's alpha coefficients were calculated on the care provision questionnaire to determine reliability and coherence between items. They were found to range from 0.80 to 0.90, which indicated a high reliability and inter-item correlation.

3.6.6 Data capturing and management

The participants completed the self-administered survey on paper. Upon the completion of the questionnaire, I checked each one for quality, cleaned, coded and stored the questionnaires on a secure computer. The computer was password protected and I was the only person with access to this password. Data were labelled and exported into STATA® 13 for analysis.

3.7 Study 3

The methods used in study 3 are summarized below and discussed further in Chapter 6.

3.7.1 Study population

The study population consisted of all HCPs who were members of the CLP team in the 11 care centres. These HCPs included maxillo-facial surgeons, registered nurses, orthodontists, plastic surgeons, social workers, speech therapists, geneticists, psychologists, dentists, and paediatricians.

3.7.2 Sampling

The sample size consisted of all HCPs who were members of the CLP team and were present on the day of the survey. Undergraduate students and student nurses were not included as they did not form part of the CLP team.

3.7.3 Development of data collection tool

I developed the information sheet (Appendix 2) and used the inter-professional collaboration framework (Appendix 6) developed by the Registered Nurses' Association of Ontario (RNAO) [142] to assess the collaboration of HCPs with each other within the team. The background and socio-demographic information was included in the questionnaire, which was in English, the official business language of South Africa. Figure 3.5 outlines the development of the questionnaire for Study 3, and approach to data collection.

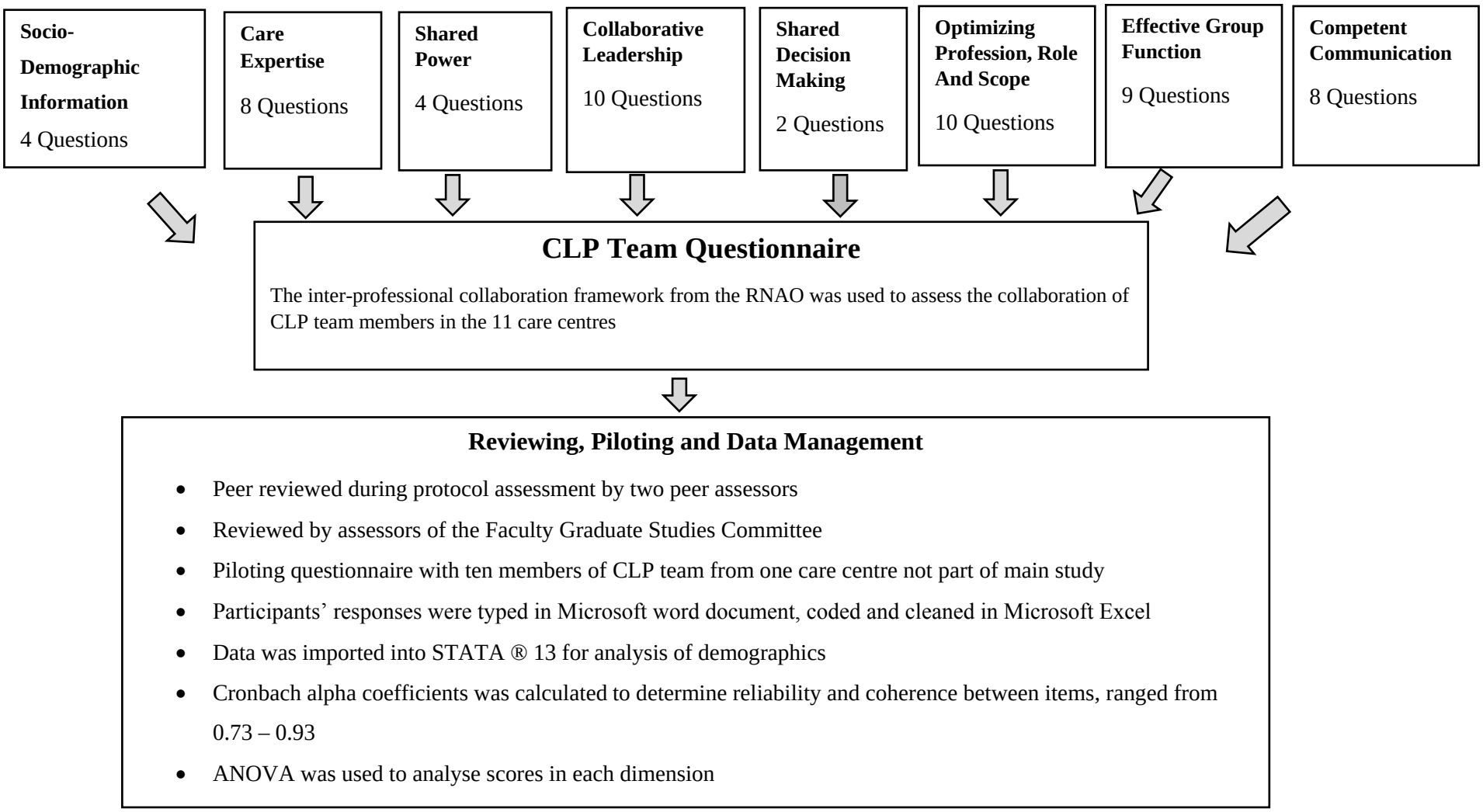


Figure 3. 5 CLP team questionnaire adapted from RNAO, data capturing and data management of Study 3

3.7.4 Piloting questionnaire

The questionnaire was piloted amongst 10 HCPs from one care centre not included in the PhD study prior to data collection, to determine clarity of questions and time taken for administration. It took an average of 30 minutes to complete and no changes were deemed necessary upon completion of the pilot study. The questionnaires completed in the pilot studies were excluded from the main study.

3.7.5 Determining reliability and inter-item correlation

Cronbach's alpha coefficients were calculated on the HCP questionnaire to determine reliability and coherence between items. These ranged from 0.73 to 0.93, indicating high reliability and inter-item correlation.

3.7.6 Data management and capturing

I checked the responses from the HCPs questionnaire for quality and completeness. I cleaned the data, coded and entered the questionnaires into Excel spreadsheet and saved the information on a password-protected computer.

3.8 Study 4

The methods used in this study are summarized below with further details provided in Chapter 7.

3.8.1 Study population

The study population consisted of parents or/and caregivers of children with CLP attending the 11 care centres.

3.8.2 Sampling

The sample size included 5-10 parents of children with CLP in each of the 11 care centres who were present on the fieldwork day. Parents of children with CLP older than three months were invited to participate in the study to determine their perceptions on having a child with CLP, health care provision and support services available for their children.

3.8.3 Development of data collection tool

I could not find a standardised tool to determine the perceptions of parents/ caregivers on care provision and support services available for their children born with CLP. I designed an information sheet (Appendix 3) and an interview schedule in English (Appendix 7) for use in the study with parents. English is one of the 11 official languages of South Africa and is the main language used to conduct official business. The interviews were conducted in English. I am fluent in all South African languages and clarification of terms was made in one of South Africa's local languages, where relevant. The different components of the interview schedule and approach to data collection are shown in Figure 3.6

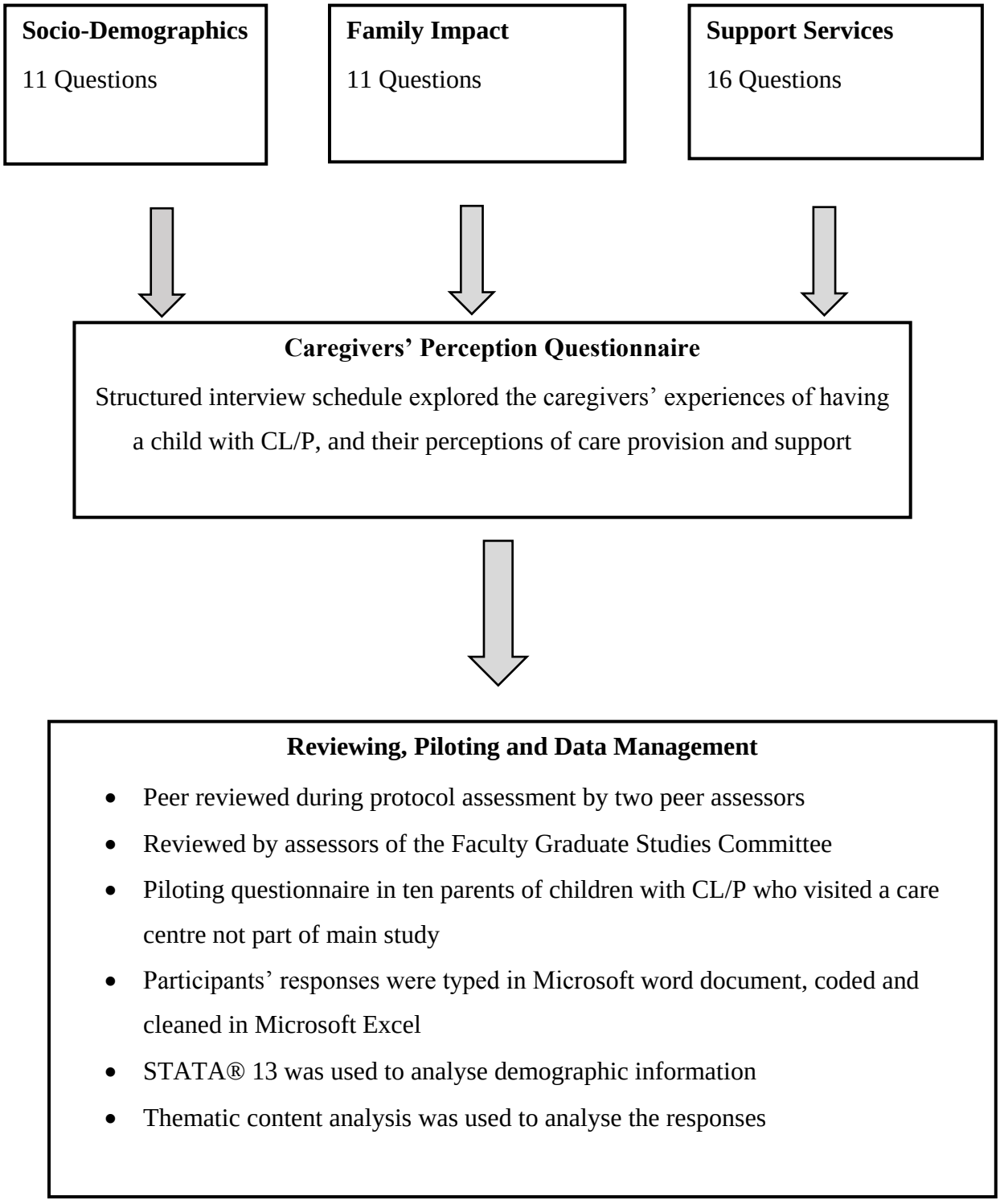


Figure 3. 6 Development of parents' questionnaire, data collection, capturing and data management for Study 4

3.8.4 Piloting interview schedule

The interview schedule was piloted amongst 10 parents of children with CLP visiting a care centre, not part of the main study, prior to data collection. Piloting assisted to determine the clarity of questions and time taken for administration. It took an average of 30 minutes to complete and no changes were deemed necessary upon completion of the pilot study. The interviews completed in the pilot studies were excluded from the main study.

3.8.5 Determining reliability and inter-item correlation

Cronbach's alpha coefficients were calculated on the parents' interview schedule to determine reliability and coherence between items. The results ranged from 0.75 to 0.90, indicating high reliability and inter-item correlation.

3.8.6 Data management and capturing

The responses of parents were written down verbatim, typed up and saved as individual Microsoft Word © documents soon after the interview. The data were cleaned and coded and entered into Excel spreadsheet and saved in a password-protected computer. The data were password protected and only I had access to this password for confidentiality. The interviews were analysed using thematic content analysis.

3.9 Addressing potential source of bias and study limitations

3.9.1 Potential biases and remedies

This section outlines potential sources of bias, and steps taken to address it. Bias results from systematic errors in the research methodology [143, 144]. In this study, the potential sources of bias were: selection bias, observation bias, social desirability bias, and prevalence-incidence bias.

The possible sources of *selection bias* included the selection of study sites and the selection of clinical records of individuals with CLP. I made every effort to prevent selection bias and included all the CLP centres in the public health sector in the study. These centres included a mix of urban and rural settings. Although the public sector provides care to the majority, 17% of the South African population have access to private sector [71]. The centres in the private sector were excluded, as well as the annual CLP campaigns by non-governmental organisations such as the Smile Train. Hence, the study findings may not be generalisable to the whole of South Africa.

The potential selection bias of clinical records was minimised by reviewing all the records for the two-year period from 1 January 2013 until 31 December 2014.

Prevalence-incidence bias, also called Neyman Bias is a selection bias where the very sick or very well (or both) are erroneously excluded from a study [144]. In the case of CLP, individuals do not recover spontaneously, hence this was not a possibility. However, excluding patients who have died prior to referral to the specialised centres will lower the prevalence of CLP. Although the number of deaths due to CLP were small in 2016 [8], I do not know how many cases were part of the deaths with unspecified causes. Hence it is impossible to adjust for this bias in my results. Nonetheless, CLP is a severe condition that requires specialised care, there have been major improvements in health care access since democracy [66] and I did a meticulous recording of all cases of CLP in the public health sector.

Observation bias results when different interviewers ask or record the questions in a slightly different way, or posing different questions to members of the study population [144]. This was not an issue in this study as I did all the interviews with parents and CLP team leaders, and I used standardised instruments.

The structured interview schedule obtained information on the perceptions of parents or caregivers of children with CLP, whilst the two surveys obtained information on the reporting of CLP team leaders and HCPs who are members of the CLP team. These were subject to *social desirability bias* and may have resulted in over- or under-reporting [145, 146]. However, the questions were phrased in such a manner as to get truthful responses. In the case of the interviews with parents, the questions were open-ended and I stressed that there was no wrong or right answer. My experience during fieldwork was that the parents or caregivers found it therapeutic to talk about their experiences, as no one had asked them questions before. Hence, they spoke from the heart on their experiences and perceptions of care and support services.

The HCPs survey used a 4-point Likert scale to allow a wide range of responses, whilst the CLP team leaders' survey was reporting on their daily experiences. There was no reason for the study participants to provide answers that they thought I wanted to hear. Importantly, these were self-administered questionnaires, and this reduced the potential of social desirability bias.

3.9.2 Addressing study limitations

A limitation of the study was the review of patient records, which were not designed for an epidemiological study. Many of the records contained missing and incomplete information. This limitation was overcame by piloting the record review form and excluding information that was not available (e.g. smoking or drinking during pregnancy). In addition, a two-year period for individuals with CLP was selected, which overcame the problems of seasonal variation in births and health care utilisation. Finally, in the absence of an active surveillance system on congenital anomalies, the record review provided a relatively cost-effective approach to data collection on CL/P prevalence.

One major specialised care centre in Gauteng Province did not maintain any records. During analysis, I adjusted the estimated prevalence rate to take account of the live births in the centre with missing data. I also adjusted the prevalence rate for the private health sector utilisation. Further details are provided in Chapter 4.

I did not record the interviews of CLP parents and this is a limitation of the study. As a clinician, I considered the recording of the interviews inappropriate, in light of the vulnerability of parents of children with cleft lip and/or palate, as it could have served as a barrier to the conversation with them, or make them worried about what could be done with their information. I maintained eye contact, wrote down responses, and verified their responses with them.

3.10 Strengths of the PhD study

3.10.1 Scholarly contribution

This is one of the first comprehensive studies on CLP conducted at all specialised care centres since the dawn of democracy in 1994. The scholarly contribution of the PhD is at two levels: the generation of new knowledge, and methodological.

The study has provided updated information on the estimated prevalence of CLP in the South Africa's public sector, as well as the clinical profile of orofacial clefts.

Studies 2 (survey of CLP team leaders) and 3 (survey of HCPs) had a national coverage of all care centres and are therefore representative of public sector treatment and care of individuals with CLP. There is little, if any information on the types of treatment provided at the different care centres. The study has highlighted the treatment gaps, which would need to be addressed (Chapter 5).

Although there have been calls for inter-professional collaboration (IPC) in education and teaching [147], to my knowledge, no study in South Africa has used the framework of the Registered Nurses' Association of Ontario to measure IPC in the clinical setting and in the case of highly specialised teams that manage individuals with CLP, which is a common congenital anomaly.

Study 4 was one of the first comprehensive studies that provided unique parental perspectives of the effects on the parents and family of children born with OFC, in an African setting. The study has provided important information on the issues that need to be taken into account in the provision of health services that are responsive to the needs of caregivers of children with CLP.

The study has also made an important methodological contribution. The record review form, questionnaires and interview schedule developed for this PhD could be adapted and/or used for future research on CLP and other congenital anomalies in South Africa, other African countries, and other LMICs settings.

3.10.2 Policy and practice relevance

Notwithstanding policy pronouncements on a South African birth defect surveillance system, this has not been implemented. The study provides information that could be used to establish the surveillance system, which in turn could assist with prevention efforts and/or resource allocation decisions [148]. The study findings could also be used to develop a more realistic national policy on congenital anomalies.

The study provides information on the variations in the treatment and care across the 11 care centres. The gaps identified can be used to assist with the comprehensive and integrated management of individuals with CLP. Such management should include addressing the information needs of parents and caregivers; the education and training of HCP on CLP; raising public awareness of all birth defects, including CLP and enhancing social support programmes.

CHAPTER 4

Epidemiology and Clinical Profile of Cleft Lip and Palate

4.1 Introduction

Congenital anomalies, defined as abnormalities of structure, function, or metabolism that are present at birth, are a major public health concern due to their life threatening nature or the potential to result in disability or death. Worldwide, it is estimated that 303 000 new-born infants die within four weeks of birth every year, due to congenital anomalies [11]. Clefting of the lip with or without palate (CLP) is the most common congenital craniofacial anomaly with the global prevalence estimated at 1 in 700 live births [16]. Orofacial clefts can occur on the lip only (CL), alveolar (CA), involve both lip and alveolar, affect the palate (CP) or involve both lip and palate (CLP). A cleft of the lip and/ or palate is serious, as it also affects negatively an individual's self-esteem, social skills, and behaviour [149-151].

The prevalence of CLP differs according to gender, ethnicity, and socio-economic status [152]. Boys are more affected than girls with a reported ratio of 2:1 with cleft lip and/or cleft lip and palate, whilst females have a slightly greater risk for cleft palate only [104].

In many high-income countries (HICs), active surveillance systems are in place, and several CLP studies have been conducted that provide epidemiological trends and prevalence estimates [152-156]. Several studies have reported on access and utilization of treatment and health care services for CLP [157-161]; standards and quality of care and long term health outcomes [162]; the clinical profile of cases, and the composition and interaction among healthcare team members in the treatment of CLP [13, 163-165].

There is an emerging body of literature on CLP in low-and-middle-income countries (LMICs), focusing on the epidemiology of CLP [88, 93, 94, 96, 97, 166-168], treatment and care of individuals with CLP, health care access, service challenges, and resource constraints [95, 98, 119, 169]. The City of Bauru in Brazil has developed a centre of excellence for the comprehensive management of individuals with CLP more than 40 years ago [170]. A review of challenges in CLP care in Africa [171] underscored the lack of reliable data on the prevalence of CLP because most of the reported studies are hospital-based [102, 166, 172].

In many African countries, active population based surveillance systems are not available. Prevalence is estimated from hospital-based data, and ranges from 0.2/1000 live births in Ethiopia [86], 0.5/1000 in Nigeria [87], 0.8/1000 in Uganda [88] and 1.7/1000 reported in Kenya [89]. A community household survey in South East Ghana found an estimated prevalence of 6.3/1000 people with CLP [173], however CLP was measured through community self-reporting rather than clinical examination. Community reporting of orofacial cleft is influenced by context and the community's description of the cleft which may be contrary to the scientific description of orofacial cleft phenotypes [44]. A recent study conducted in Democratic Republic of Congo reported an incidence of 0.8 per 1000 live births for non-syndromic CLP [174].

In South Africa, earlier studies on prevalence of CLP were conducted in Cape Town [93, 94], Johannesburg [95] and Pretoria [96, 97] in the late 1980s. The reported prevalence ranged from 0.1 to 0.4 per 1000 live births. However, these studies were conducted more than 30 years ago and they predate democracy in 1994. Furthermore, the studies focused on three major South Africa's cities, and not all the specialised academic treatment centres were included. Importantly, a study that examined the causes of under-five mortality rates found that the proportion of deaths due to non-natural causes, congenital disorders and non-communicable

diseases has increased [67]. In light of the dearth of scholarly studies on the epidemiology of CLP in South Africa, we conducted this study to determine the epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa.

4.2 Methods

4.2.1 Ethical considerations

The Human Research Ethics Committee (Medical) of the University of the Witwatersrand in Johannesburg provided ethical approval for the study to review patient medical records. All personal identifiers were removed from the records, hence no informed patient consent was required. Each specialised academic treatment centre also provided approval to access and review the patient medical records.

The principal investigator (PH) is registered with the Health Professions Council of South Africa as an orthodontist, and is familiar with all the principles of patient confidentiality in medical records. Only PH had access to the relevant records for the study period. The medical records at each centre were assessed in a private area and never left unattended. The principal investigator allocated each patient record a unique identifier and no patient name or any other form of identification was recorded on the data collection form. The data containing unique numbers were kept on a password-protected computer.

4.2.2 Study sites and setting

South Africa's public health sector provides health care to an estimated 83% of the population, while the private health sector provides care to a minority (17%) of the population with private health insurance [71]. Public sector hospitals are categorised into five types, namely: - district hospitals; regional hospitals; tertiary hospitals; central hospitals and specialised hospitals [72].

There are ten central hospitals situated in six of South Africa's nine provinces. These central hospitals are attached to Health Science Faculties, and serve as teaching centres for the training of health professionals. These central hospitals also provide tertiary hospital services and serve as referral facilities for primary and secondary health facilities, and in some cases as specialised centres for referral of complicated medical conditions from neighbouring provinces [72].

The study setting consisted of 11 specialised academic centres (nine central hospitals and two specialised dental hospitals) with multi-disciplinary teams of health professionals who provide care to individuals with CLP. These 11 centres are situated in six of South Africa's nine provinces as shown in Figure 3.2, which included three mixed urban-rural provinces. We selected these centres because they cover all those individuals who obtain care for CLP in South Africa's public health sector (83% of the population), and to generate new knowledge that will contribute to improvements in health care in the public health sector of South Africa.

4.2.3 Study population and sampling

All clinical records for the selected study period, 1 January 2013 to 31 December 2014, constituted the study sample. At each of the CLP specialised care centres, we selected the clinical records of all cleft individuals that visited these academic centres during the study period. Because individuals with CLP make numerous visits to these centres over a prolonged period, care was taken to record each individual only once, in order to avoid duplications.

A structured, pre-tested record review form (Appendix 4), was used to obtain demographic, clinical and treatment information on each CLP individual. We recorded the "race" of individuals with CLP from a list of pre-defined categories: Black African, Coloured, Indian or Asian, White and other. Although we do not want to give credibility to the apartheid roots of these group classifications, years of systematic, legislated racism continue to shape socio-

economic circumstances and access to care in post-apartheid South Africa [175]. The principal investigator (PH) extracted the information from the CLP medical records.

Each academic centre was allocated a unique identifier to ensure anonymity. The cleft types were classified into the various broad categories: Bilateral cleft lip and palate (BCLP), left cleft lip and palate (LCLP), right cleft lip and palate (RCLP), bilateral cleft lip only (BCL), left cleft lip only (LCL), right cleft lip only (RCL), cleft palate only (CP), cleft of alveolar only (CA), cleft lip and alveolar (CLA).

4.2.4 Data management and analysis

The data was imported into STATA® 13 for descriptive and inferential statistical analyses. All statistical tests were conducted at 5% significance level.

The total number of live births for the two year period (January 2013-December 2014) was obtained from Statistics South Africa [176].

We estimated three different CLP prevalence rates: overall prevalence rate; prevalence rate adjusted for the private health sector utilisation and missing data in one specialised centre; and provincial prevalence rates.

Overall prevalence rate: The total number of individuals with orofacial clefts at the specialised academic care centres, (numerator data) was divided by the total number of live births during the study period (denominator data) and the value multiplied by 1000.

Prevalence rate adjusted for the private health sector utilisation and missing data in one centre: In order to adjust for the 17% of the South African population covered by private health insurance [71], we assumed that 17% of all live births occur in the private health sector. We further calculated that 2% of births occurred in the specialised centre with missing data. During

the study period, 43 240 live births [177] were recorded at that centre. The denominator was adjusted by subtracting 19% (private sector and missing data from one centre) from the total number of live births. Therefore, the prevalence of CLP was estimated by dividing the numerator with the adjusted denominator and multiplied by 1000.

Provincial prevalence estimates: We also calculated the prevalence of CLP in each province. The denominator (number of live births) was adjusted for each province to take account of private sector utilization. The proportion of provincial population with private health insurance ranged from 9% to 28% [71]. In the case of Gauteng Province, the denominator was also adjusted to take account of the centre with missing data. Following adjustments, the prevalence was calculated by dividing the number of CLP individuals in each province (numerator) by the number of live births (adjusted denominator) in that province during the study period, multiplied by 1000.

4.3 Results

We reviewed 717 CLP records from 10 specialised academic centres, 18 were excluded from the study because of incomplete data, and the final sample was 699 records.

The majority of CLP cases (45.6 %) were treated in four centres located in Gauteng Province.

4.3.1 Profile of individuals utilising specialised academic centres

Table 4.1 shows the demographic characteristics of the individuals utilising the specialised academic centres. Cleft distribution by population groups showed majority for Black African, followed by White, Coloured and Indians respectively. More females presented with CLP compared to males. The majority of individuals treated in these centres (97%) were South Africans with a small percentage from neighbouring countries.

Table 4.1 Demographic characteristics of the CLP individuals

Characteristics	Sample size
CLP median age at consultation in months (IQR)	3 (0.75-13)
Gender	**n=694**
Male	330 (47.5%)
Female	364 (52.5%)
Race	**n=687**
Black African	448 (65.1%)
Coloured	94 (13.6%)
Indian	37 (5.4%)
White	109 (15.9%)
Nationality	**n = 690**
South African	669 (97%)
Non- South African	21 (3%)
CLP Birth by Province	**n=694**
Eastern Cape province	18 (2.6%)
Free State Province	108 (15.6%)
Gauteng Province	202 (29.1%)
KwaZulu Natal Province	66 (9.5%)
Limpopo Province	60 (8.7%)
Mpumalanga Province	52 (7.5%)
North West Province	22 (3.1%)
Northern Cape Province	2 (0.3%)
Western Cape Province	146 (21%)
Non- South Africans	18 (2.6%)
Number of CLP per Academic Centre	**n= 699**
SITE 2	93 (13.3%)
SITE 3	33 (4.7%)
SITE 4	52 (7.4%)
SITE 5	141 (20.2%)
SITE 6	79 (11.3%)
SITE 7	70 (10%)
SITE 8	62 (8.9%)
SITE 9	16 (2.3%)
SITE 10	122 (17.5%)
SITE 11	31 (4.4 %)
Accounting for missing data: - From 699 records, only 694 records had gender and province of birth indicated in them, 690 records had nationality shown, and race was recorded in 687 records. One centre, SITE 1, was excluded because there were no records available.	

4.3.2 Estimated CLP prevalence

The estimated overall prevalence rate of CLP was 0.3 per 1000 live birth calculated from 2,300 897 live births during the study period [176]. Prevalence rate adjusted for the private health sector utilisation and missing data in one centre was 0.4 per 1000 live births.

The prevalence of CLP per province shown in Table 2, ranged from 0.1 to 1.2 per 1000 live births. The highest prevalence was in the Free State Province and the lowest in the Eastern and Northern Cape Provinces. Although the majority of CLP were recorded in Gauteng Province, the estimated prevalence rate in this province was 0.5 per 1000 live births.

Table 4.2 Prevalence of CLP in each Province

Provinces	**Number of CLP**	**Proportion on private health care***	**Adjusted denominator****	**Prevalence per 1000 live births**
Eastern Cape Province	18	11%	235 247	0.1
Free State Province	108	18%	93 134	1.2
Gauteng Province***	202	28%	384 406	0.5
KwaZulu Natal Province	66	13%	412 034	0.2
Limpopo Province	60	9%	245 075	0.2
Mpumalanga Province	52	15%	150 813	0.3
North West Province	22	15%	130 298	0.2
Northern Cape Province	2	20%	43 374	0.1
Western Cape Province	146	26%	158 081	1.0
Adjusted denominator=number of live births adjusted by subtracting percentage to account for private sector utilisation. For Gauteng Province, the denominator was also adjusted by 7% to account for missing data in one centre).				

*Source [176] ; **Source [71]; ***Source [177]

Figure 4.1 shows the profile of clefting at academic treatment centres according to cleft type, cleft description, cleft laterality and cleft position. Cleft lip and palate (CLP) was the most predominant type of cleft followed by cleft palate (CP) and the cleft lip (CL) Figure 4.1(a). Other cleft abnormalities included midline facial cleft (2), lateral facial cleft (6), and syndromes (6). The majority of cleft were unilateral as shown in Figure 4.1(b) and occurred predominant on the left side compare to the right side, Figure 4.1(c).

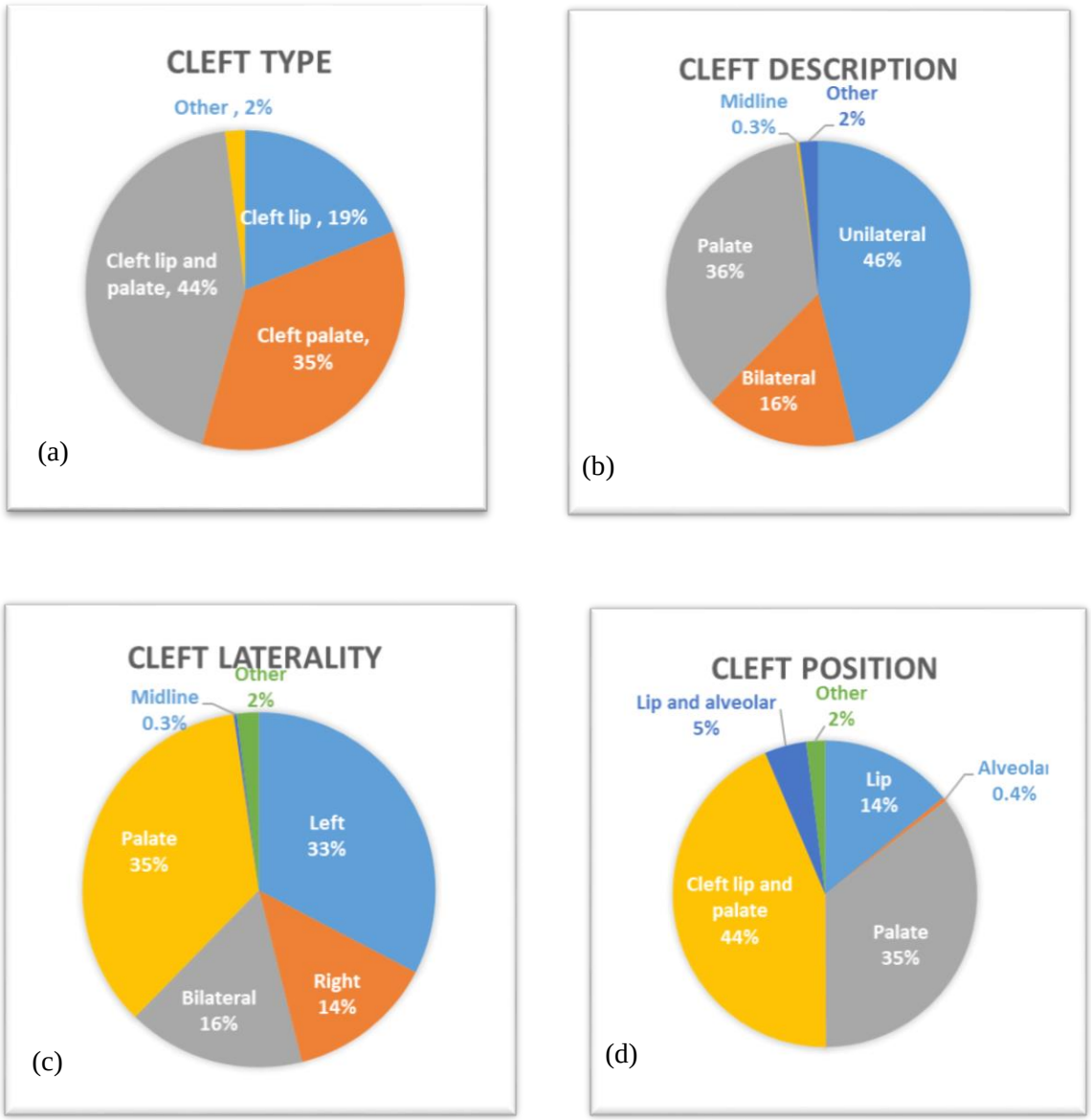


Figure 4.1 Profile of clefting at academic treatment centres

Cleft distribution by gender and types is shown in Table 4.3. From 694 clefts, there were more CP in females than males whilst CLP was predominant in males. Unilateral clefts occurred most frequently in males compared to females. The left side dominated the occurrence of clefts compared to the right side in both CL and unilateral CLP for both genders. Statistically significant differences ($p<0.001$) were observed for cleft type, distribution and location between males and females.

Table 4.3 Distribution of types of clefts by gender

n=694	**Male** n (Col %)	**Female** n (Col %)
Cleft type		
Cleft lip	65 (19.7%)	66 (18.1%)
Cleft palate	88 (26.7%)	158(43.4%)
Cleft lip and palate	174 (52.7%)	129 (35.4%)
Other	3 (0.9%	11 (3%)
Total	330 (100%)	364 (100%)
Cleft description		
Unilateral	176 (53.3%)	142 (39%)
Bilateral	63 (19.1%)	53 (14.6%)
Palate	88 (26.7%	158 (43.4%)
Other	3 (0.9%)	11 (3%)
Total	330 (100%)	364 (100%)
Cleft laterality		
Left	117 (35.5%)	108 (29.7%)
Right	59 (18.5%)	34 (9.3%)
Left and Right (Bilateral)	63 (18.2%)	53 (14.6%)
Palate	88 (26.7%)	158 (43.4%)
Other	3 (0.9%)	11 (3%)
Total	330 (100%)	364 (100%)
Cleft position		
Lip	47 (14.2%)	50 (13.7%)
Alveolar	1 (0.3%)	2 (0.5%)
Palate	88 (26.7%)	158 (43.4%)
Cleft lip and palate	174 (52.7%)	129 (35.4%)
Lip and alveolar	17 (5.2%)	14 (3.8%)
Other	3 (0.9%)	11 (3%)
Total	330 (100%)	364 (100%)

P value < 0.001***

4.4 Discussion

The prevalence rate for CLP in individuals utilising the specialised academic treatment centres in South Africa's public sector was estimated to be 0.3 per 1000 live births and 0.4 per 1000 when the denominator was adjusted. This prevalence rate could be underestimated, because it excludes stillbirths, abortions and those children who might have died within the first three months of birth, or before seeking care. Nonetheless, this rate is also comparable to those found in Nigeria of 0.5 per 1000 live births [87] and higher than that of Ethiopia at 0.2 per 1000 live births [86]. In contrast, the prevalence rate reported in our study is lower than the rates reported for a population base birth defects registries from 30 countries from 54 international craniofacial registries during the period 2000 to 2005, where the overall prevalence of CLP was 1.0 per 1000 [10].

We also estimated the prevalence of orofacial clefts in each province. The prevalence rate ranged from a low of 0.1 per 1000 in the Eastern and Northern Cape Provinces to a high of 1.2 per 1000 in Free State Province. The geographic location of the different provinces within South Africa, and their different cultural and ethnic factors could have contributed to the variability of orofacial cleft prevalence found in this study. Other studies have also found that geographic factors are associated with orofacial cleft predisposition, occurrence and treatment methods [178]. A study performed in Colorado, USA, showed that country of residence and place of birth, whether metropolitan or non-metropolitan, had greater range of difference for OFC occurrence [154]. Nonetheless, our study did not determine the reasons for the geographic difference in the prevalence rates. This area requires future research.

In many LMICs including South Africa, infectious diseases dominate the causes of infant and child mortality, and congenital anomalies account for a relatively small proportion of under-five mortality [179]. Nonetheless, children born with CLP require treatment in the public sector that will start at infancy, and extend into late adolescence, or early adulthood. The affected individuals, their families, and the public health care system feel the burden of care as reported by Bamford et al [67] that the proportion of under-five mortality due to congenital anomalies has increased. Our study findings could inform the proposed national surveillance system on congenital anomalies.

Clefts of the lip have been reported to be predominant in blacks [180]. A retrospective study in Tanzania reported individuals with higher proportions of cleft lip only [181] and similar reports from a study in Kenya [182] and Zimbabwe [183]. Our study also found a predominance of black Africans with orofacial clefts (OFC). However, it could be a reflection of the South Africa's population demographic profile where the majority of the population are black Africans who utilise the public health sector [140], rather than ethnic differences, as found in studies in other countries [104]. Further research is needed to determine whether ethnic differences play a role in South Africa.

Orofacial clefts have been reported to be more predominant in males compared to females [87]. Our study found a female predominance, with the majority of females presenting with CP. This proportion of cleft type from our study was similar to that found in a Nigerian study, which reported more females with CP than males [87]. Our study findings could be because parents perceive CP as the mildest form of cleft since it is not visible on the outside and hence the parent does not delay seeking care [184]. However, general societal neglect for cleft palate can lead to decreased access to palatal surgery [181, 185], and furthermore, CP has implications

for feeding, speech and jaw development and is usually associated with syndromes. This type of cleft is more likely to increase morbidity and mortality from poor feeding leading to child malnutrition and vulnerability to infectious diseases [186, 187].

Treatment and management of individuals with orofacial clefts vary depending on the type and severity of the cleft, the presence of associated syndromes, other birth defects and the child's age. Cleft lip repair is proposed to be done at three months after birth [114]. The median age at consultation for our study was about three months with a relatively high interquartile range of 3 weeks to 13 months. A retrospective study of the epidemiology, clinical aspect and management of clefts in Burkina Faso reported that more than 60% of children presented for consultation when they were older than one year [188]. However, delays in first consultation will delay treatment and the individual may suffer physical impairment and societal relationships with potential long-term psychological effects, including behavioural problems and lack of social integration [189-191].

Our study used clinical records to review previously recorded data to update the prevalence of clefting in the public sector hospitals in South Africa. Clinical records have advantages as they enable a relatively easy and less resource intensive research approach to answering specific clinical questions. However, they have certain disadvantages including variation in the manner in which data has been gathered and recorded in thus limiting the extraction and interpretation of the variables, as well as records may be incomplete or lost in the course of time, leading to missing data [192]. Furthermore, the estimation of prevalence from hospital records of CLP exclude all stillbirths and miscarriages that would be possible to obtain from an active birth defect surveillance system, thus leading to a possible underestimation of the prevalence rate. Although some of these patients might have been referred from the private sector to the

academic centres, none of the records indicated this referral. Nevertheless, we analysed CLP records to compute an estimated prevalence of OFC in South Africa's public health sector and to provide a detailed description of cleft types from the specialised academic centres. Therefore, our study has provided updated information on the epidemiology of CLP in South Africa's public sector, especially since the end of apartheid in 1994. The study findings provides baseline data that should inform the implementation of the planned active birth surveillance system.

4.5 Conclusions

The study has generated new knowledge on the epidemiology and clinical profile of individuals with CLP in the South African public health sector. It is imperative for South Africa to establish an active birth surveillance system on congenital anomalies to enable comprehensive management of CLP individuals and to inform health service planning and policy.

4.6 Authors' contributions

PH and LR conceptualized and designed the PhD study. PH collected all the data, and conducted the record review, as part of her PhD. LR is the primary supervisor of the PhD. JL is a biostatistician and together with PH performed the statistical analysis, with inputs of LR. All authors contributed to the writing and editing of the manuscript, and read and approved the final version of the manuscript.

CHAPTER 5

Healthcare Provision to Individuals with Cleft Lip and Palate

5.1 Introduction

Cleft lip and palate (CLP) is the most common congenital anomaly of the craniofacial complex, with an estimated worldwide prevalence of 1 in 500 to 700 live births [16]. In South Africa, CLP is amongst the five most common birth defects reported, [92] with an estimated prevalence that ranges from 0.1 to 0.4 per 1000 live births [92, 96, 193].

Orofacial cleft (OFC) phenotypes can present as cleft lip only (CL), clef palate (CP), cleft lip and palate (CLP) and cleft lip with or without palate (CLP) [44]. Affected children present with a number of medical problems and potential complications that include feeding difficulties, hearing loss, speech problems, disfigured appearance and dental malformation [194-197]. Hence, individuals with CLP require co-ordinated and specialised treatment offered by a multidisciplinary team [111-113], that includes geneticists, maxillo-facial surgeons, otolaryngologists, orthodontists, paediatricians, plastic surgeons, paediatric dentists, psychologists, professional nurses, social workers, speech & language therapists [114-116]. These teams are available in high-income countries (HICs).

There are various clinical protocols that have been proposed for the treatment of CLP [198]. Many HICs have adopted national protocols and guidelines and have centralised services and/or established cleft centres for the management and follow up of affected individuals with CLP [13, 199-201]. The advantages of centralisation and/or concentration of specialised services in a country includes: coordination of treatment, standardised data collection, availability of a critical mass of experts, clinical audit and review to enhance quality of care;

responsiveness to patient and family needs, and ongoing monitoring and evaluation [114, 138, 139, 202-206].

Globally, it is estimated that only 20% of individuals with CLP have access to comprehensive treatment that involves a coordinated team approach [118]. The treatment and care gaps includes: lack or shortages of health care professionals, lack of infrastructure, and delays in surgical repairs of the clefts [118, 119]. These gaps are most pronounced in LMICs in Africa, Asia and South America [119, 169]. In many LMICs, outreach programmes by non-governmental organisations (NGOs) have assisted in improving the services for individuals with CLP [119-123, 169]. These NGOs, together with support groups that include parents of the affected children, have played an important role in the ongoing management of patients with CLP. However, the majority of LMICs, including South Africa, lag behind in terms of national treatment protocols, standardised data collection, access to care by a multi-disciplinary team and evaluation of treatment outcomes[13, 114, 124].

Notwithstanding improvements in the treatment of individuals with CLP in the preceding decades [207-209], there is still no universal protocol for repair and ongoing clinical management. Nonetheless, treatment modalities in the management of CLP are often based on chronologic age and dento-facial development[209]. The common elements of clinical standards and the sequence of treatment of CLP are shown in Table 5.1[209]. At birth, genetic counselling is given to parents and feeding of the baby is evaluated. Depending on the size of the cleft, feeding plates are recommended within the first week after birth to assist with feeding. Pre-surgical infant orthopaedics for the approximation of the segments is also performed prior to cleft lip repair. As the child grows, several follow-up consultations for management of the cleft are done until treatment is completed during adulthood [114].

Table 5.1 Treatment sequence in CLP management

Chronologic age/dento-facial development	CLP Treatment
At birth	• Genetic counselling • Feeding plate • Pre-surgical infants orthopaedics • Psycho-social counselling
3-months	• Surgical lip repair
6-12 months	• Surgical palate repair • Grommets • Speech therapy • Routine dental treatment
5-7years	• Alveolar bone graft • Speech therapy • Dental treatment/maxillary expansion/bone graft/dental arch alignment • Speech therapy
10-14 years	• Orthodontic treatment/maxillary expansion/bone graft/maxillary protraction
16-18 years	• Orthodontics treatment/orthognathic surgery/maxillary advancement • Psychology counselling
18-20 years	• Prosthodontic replacement of missing teeth • Nose revision

Source: de Ladeira & Alonso [209]

There is a dearth of studies on the management of CLP in South Africa, except for a 1953 report in a Johannesburg private clinic which found that the clinical management of patients with CLP was uncoordinated [95]. The aim of this study was to compare the treatment and care of individuals with CLP at all the CLP care centres in South Africa.

5.2 Materials and methods

This cross-sectional study was conducted during 2015 and 2016 in all the 11 specialised CLP academic care centres situated in six of South Africa's nine provinces.

The Human Research Ethics Committee (Medical) of the University of the Witwatersrand in Johannesburg provided ethical approval for the study. Approval was also obtained from the CLP care centres. Confidentiality was maintained throughout the study. Each centre and CLP team leader were allocated the same unique identification number to ensure anonymity. The data containing unique numbers and centre information were kept on a password protected computer.

The CLP team leaders were interviewed using a semi-structured questionnaire, which contained 29 questions divided into three parts: centre information, treatment and management of CLP and team members. The questions focused on the entry point of care for individuals with CLP, the type of services and treatment provided at each centre, the treatment protocols followed in the clinical management , the continuum of care, internal referral systems as well as the healthcare professionals who were members of the team. The interviews were complemented with a record review of all individuals with CLP that visited these academic centres for the period from 1 January 2013 until 31 December 2014.

At each centre, the principal investigator (PH) conducted the interviews in a private room. Each interview lasted an average of 30 minutes. The responses of participants were written down verbatim in the space provided in the questionnaire, typed up and saved as individual Microsoft Word © documents. The data was imported into STATA® 13 for descriptive analyses.

5.3 Results

A 100% response rate was obtained and all 11 team leaders from CLP care centres participated in the study: five in Gauteng Province, two in Western Cape Province and one in each of the provinces of the Eastern Cape, Free State, Kwa-Zulu Natal, and Limpopo. Most of the centres

have been in existence more than five years. The descriptive characteristics of the CLP centres are shown in Table 5.2.

One centre in the Gauteng Province had no records for the 2-year review period. The number of individuals with CLP at the 10 centres totalled 699 CLP, ranging from 16 to 144. The team leaders reported that individuals with CLP were treated mostly by plastic surgeons. Most centres reported that they use team approach for the provision of CLP treatment.

Table 5. 2 Descriptive characteristics of the CLP Centres

Characteristics	Sample size
CLP Team Leader	**n=11**
Plastic surgery	9 (81.8%)
Maxillo-facial surgery	1 (9.1%)
Orthodontics	1 (9.1%)
Type of care provided	**n=11**
Individual specialist	1 (9.1%)
Team approach	6 (54.6%)
Hybrid approach	3 (27.3%)
Other	1(9.1%)
Number of individuals with CLP per Academic Centre	**n= 699**
SITE 2	93 (13.3%)
SITE 3	33 (4.7%)
SITE 4	52 (7.4%)
SITE 5	141 (20.2%)
SITE 6	79 (11.3%)
SITE 7	70 (10.0%)
SITE 8	62 (8.9%)
SITE 9	16 (2.3%)
SITE 10	122 (17.5%)
SITE 11	31 (4.4 %)

The treatment provided by each centre is shown in Table 5.3. The dominant modes of treatment were surgical repair of CLP and speech therapy. Other treatment modalities were less common at the CLP care centres. Although site 9 is listed as a CLP specialised academic centre, it does not provide any active treatment but serves a booking and follow-up centre for surgical repairs performed by a non-governmental organisation.

Table 5. 3 Types of CLP Treatment provided by each academic centre

Site	Treatment									
	*FP	*PS0	*ST	*GC	*LPR	*GDT	*ORT	OGT	*ABG	*ENT
1	No	No	Yes	Yes	Yes	No	No	No	No	No
2	No	No	Yes	No	Yes	No	No	No	No	No
3	No	No	No	No	Yes	No	No	No	No	No
4	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	No
5	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
6	No	No	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
7	No	No	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes
8	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
9	No	No	No	No	No	No	No	No	No	No
10	No	No	Yes	Yes	Yes	No	No	Yes	No	No
11	No	No	No	No	Yes	No	No	No	No	No
Total	2/11	2/11	8/11	6/11	10/11	4/11	5/11	5/11	5/11	2/11

*FP - feeding plate; PSO – presurgical orthopaedics; ST- speech therapy; GC – genetic counselling; LPR – lip and palate repair; GDP – general dental treatment; ORT – orthodontic treatment; OGT – orthognathic treatment; ABG – alveolar bone graft; ENT – ear, nose and throat treatment; Yes - treatment offered; No – treatment not offered

Six centres provided their treatment protocols for analysis (Table 5.4). These protocols showed that two centres utilised feeding plates and pre-surgical orthopaedics. All six centres followed a set protocol for the timing of the surgical procedure of the lip and palate. The sequencing of lip surgery was similar in five centres. Palate surgery and alveolar bone graft were performed at similar times by all six centres. Whilst centre 4 and 5 reported availability of orthodontic treatment, the age of treatment was not indicated. Similarly, orthognathic treatment was performed by four centres and speech therapy offered by all six centres, no specific age of delivery was indicated. Other treatments were not provided according to the standard protocol (Table 5.1) and were neither provided whenever needed nor available in most centres.

Table 5. 4 Comparison of CLP treatment protocols across six centres*

Centre	Feeding plate	Pre-surgical orthopaedics	Lip repair	Palate repair	Alveolar bone graft
2	No	No	3m	6-12m	6-8y
4	birth	Birth	3-6m	12-18m	8-10y
5	birth	birth	12-18m	6-12m	8-10y
6	No	No	3m	6-12m	8-10y
7	No	No	3m	12-18m	8-10y
8	No	No	3m	9m	7-11y

*These centres provided written protocols for analysis

Table 5.5 shows the categories of healthcare professionals (HCP) in each CLP care centre. Most centres had plastic surgeons (91%) and speech therapists (72.7%) as part of CLP team. Other HCPs such as ENT surgeon, paediatric dentist, paediatric surgeon, psychologist and social worker were not available in most of the centres. Although professional nurses were present in seven centres (63.6%), only two centres responded that these nurses were actively involved with CLP clinical treatment and they played primarily administrative roles in the other five centres. Only two centres reported that parents' support groups are incorporated in CLP management.

Table 5. 5 Members of CLP team in each academic centre

CLP Team Members												
Centre	**§Gen**	**§PS**	**§Orth**	**§ENT**	**§MFS**	**§PD**	**§Paed**	**§Psych**	**§PN**	**§SW**	**§ST**	**§PSG**
1	0	1	0	0	0	0	0	0	1	1	1	1
2	0	1	0	0	1	0	0	0	1	1	1	0
3	0	1	0	0	0	0	0	0	1	0	0	0
4	1	1	1	0	1	1	0	0	0	0	1	0
5	1	0	1	0	1	1	0	0	1	0	1	1
6	1	1	1	1	1	0	0	0	1	0	1	0
7	1	1	1	1	0	1	0	1	0	0	1	0
8	1	1	1	0	1	1	1	0	1	0	1	0
9	0	1	0	0	0	0	0	0	1	0	0	0
10	0	1	0	0	0	0	0	1	0	0	1	0
11	0	1	0	0	0	0	1	0	0	0	0	0
Total	6(54.5%)	10(91%)	5(45.5%)	2(18.2%)	5(45.5%)	4(36.4%)	2(18,1)	2(18.2%)	7(63.6%)	2(18.2%)	8(72.7%)	2(18.2%)

§Gen = geneticist; PS =plastic surgeon; Orth = orthodontist; ENT = ear-nose -throat surgeon; MFS= maxillo-facial surgeon; PD = paediatric dentist; Paed = paediatric surgeon; Psych = psychologist; PN = professional nurse; SW = social worker; ST = speech therapist; PSG= parent support group; 1 = member of team; 0 = not member of team.

5.4 Discussion

This is one of the first comparative studies to analyse treatment and care of individuals with CLP in the South African public sector. Our study found variations across the 11 care centres in: the number of individuals with CLP treated, the type treatment provided to these individuals, availability of written protocols and the composition of the health care teams.

The number of individuals with CLP treated during the two-year period of the review ranged from 16 to 141. Site 9 does not meet the criteria of a specialised care centre, since it only provided booking and follow-up services to individuals with CLP. Experts have pointed to the importance of sufficient case-loads to ensure competent clinical care and to secure adequate resources for comprehensive care[14]. A 2001 review in the United Kingdom led to government directive to the regions to provide care from a single regional centre, with a comprehensive specialist team and a guideline that one surgeon should be responsible for 40 to 50 new cases requiring primary surgery per year [13, 210]. Similarly, cleft care has also been centralised in Brazil [170, 211] and in the Scandinavian countries of Denmark, Finland, Norway, and Sweden [138]. However, the concept of regionalisation or concentration of CLP services is under-developed in South Africa, exacerbated by the relatively low priority of congenital anomalies within the overall context of the country's quadruple disease burden.

This study found that the dominant treatment modalities were surgical repair of the lip and palate (10/11 or 90.9% of centres) and speech therapy (7/11 or 63.6% of centres). Two of the centres without speech therapy are located in the rural provinces of the country, one (Site 9) does not provide any active treatment for CLP. This underscores the inequities in access to specialised services in the South African public health sector [212]. Speech therapy is an essential service for CLP, and this gap would need to be addressed, either through efforts to

recruit speech therapists to these centres, formal referral to, and/or partnerships with the urban CLP centres, or public/private partnerships.

Our study found that feeding plates were under-utilised since two centres (2/11 or 18.2%) reported their use, whilst the other nine centres (9/ 11or 81.8%) did not use them for children with CLP. Notwithstanding the controversies regarding the use of feeding plates [213-215], feeding difficulties following birth of babies with CLP are common and the feeding plate can assist with the closure of the cleft to facilitate sucking and to prevent choking. A study among caregivers of children with CLP found that feeding difficulties were one of the most distressing aspects for in caring for their children [184], highlighting the importance of adequate support and assistance to caregivers regarding feeding methods.

The comparison of treatment protocols was only possible for six centres of the 11 centres. Five of the centres did not have a written protocol, which means that care might not be standardised in these centres, and this could influence the quality of care provided. CLP care centres should be encouraged to develop standard treatment protocols. The findings of this study could be used to develop standard treatment guidelines, with the involvement of all 11 CLP centres, drawing on the experiences of those centres with protocols, and international best practice [138, 170, 211].

The timing for lip repair was 3-6 months in most centres. However, the median age at consultation for our sample was about three months with a high interquartile range of three weeks to 13 months, which means that a sizable portion of these children came after six months when ideally their first operation should have been done. This delayed age of consultation has also been reported in other LMICs [86, 183, 188]. Lip surgery provides comfort to the parents

of children with CLP, as they are able to show their children in public and avoid stigmatisation. Strategies would need to be developed to ensure that the majority of children with CLP get access to lip surgery within the first six months of life. This will assist with the reduction of possible physical impairment and societal courtesy stigma and discrimination [184, 216].

The CLP centres had variations in the number of the team members, ranging from two to eight. In most centres, there was scarcity of certain specialists. There are existing guidelines on the health professionals that should constitute members of CLP teams [114]. Although the South African context would need to be taken into account, these guidelines should inform the constitution of the multidisciplinary team. Our study found that professional nurses were under-utilised as members of the CLP team. It has been reported in other settings that professional nurses play an important role, especially in assisting mothers with feeding [85, 114, 217].

Our study findings revealed that only two centres reported incorporating parent support group in their management of individuals with CLP. Other studies have found that parents' involvement and participation in care decisions are important in CLP care outcomes, as their knowledge and support enhance compliance with treatment, reduction of complications [184, 218, 219].

Although our study generated new knowledge on the types of treatment available at CLP centres, it was mainly descriptive. We did not evaluate: the types of surgery used for treatment, the quality of care, the outcomes of treatment, the cost of treatment, and the follow-up treatment provided to individuals with CLP. These are study limitations, but point to areas for further research. Nonetheless, the knowledge generated by this study can be used to develop standard treatment guidelines for individuals with CLP, enhance the discourse on appropriate treatment

modalities for these individuals, and contribute to the development of collaborative partnerships across the 11 centres.

5.5 Conclusions

Although the treatment of children with CLP and other types of craniofacial anomalies in South Africa has been ongoing for more than six decades [95], comprehensive care is still lacking. The study has shown that there are gaps in the types of treatment provided across the 11 centres, lack of some of the members of the multi-disciplinary team, and absence of standardised treatment protocols. These gaps are more pronounced in those centres located in the rural provinces of South Africa.

5.6 Recommendations

The study findings underscore the need for the National Department of Health (NDoH) to review and update the national policy on congenital anomalies, which has not been revised since 2005. The NDoH should also ensure equitable access to appropriate treatment and care for individuals with CLP in South Africa, regardless of geographical location. This might be achieved through regionalisation of centres, developing a standard treatment protocol for CLP, in consultation and with the involvement of health professionals at these 11 centres. Such regionalisation will ensure: the existence of a critical mass of health professionals at each specialist facility, foster collaboration amongst the centres, enhance the education and training of health care providers on congenital anomalies, enable sharing of experiences and mutual learning, optimise resource utilisation for CLP care, and facilitate research on treatment models and outcomes. Ultimately, these initiatives will contribute to quality of care improvements for all individuals with CLP.

5.7 Authors' contributions

PH conducted the study as part of PhD. LR is the primary supervisor of the PhD. TCD is the co-supervisor of the PhD. PH and LR conceptualized and designed the PhD study. PH collected all the data, PH and LR analysed the data. All authors contributed to the writing and editing of the manuscript, and read and approved the final version of the manuscript

CHAPTER 6

Interprofessional Collaboration among Healthcare Team Members

6.1 Introduction

The global discourse on inter-professional collaboration (IPC) or the ability of health-care professionals to work together as a team has intensified [79, 220-225]. IPC is defined as: "*multiple health workers from different professional backgrounds working together with patients, families, caregivers, and communities to deliver the highest quality of care*" [79]. The envisaged benefits of IPC include identifying and drawing on the strengths of each member of the health-care team and using those strengths to prevent and manage complex diseases, provide quality of care, and improve both patient and health worker outcomes [79, 224]. This is because IPC improves communication and teamwork and promotes coordination of care across the continuum of health care [79, 224]. IPC is also seen as facilitating egalitarian relationships among health professionals [226], and assisting with the amelioration of health workforce shortages [79]. The converse also applies with some scholars suggesting that lack of or suboptimal IPC among members of healthcare teams is the root cause of poor health care quality [227].

There is a considerable body of research on IPC that shows that patient outcomes and quality of care are enhanced and costs are reduced when health care team members work together towards shared patient-centred goals [227-234]. IPC has been reported to benefit those patients with chronic disorders, mental illnesses, and social conditions [227]. A study that evaluated the effect of including a pharmacist as a team member during rounds in the intensive care unit demonstrated a two-thirds reduction in the number of potential adverse drug events [235]. A

systematic review of 36 randomised controlled trials involving IPC demonstrated the risk for hospital readmission was reduced by 19% [236]. Older adults were more than 30% less likely to visit the emergency department with implementation of IPC [236]. A study found that patients treated by IPC teams were more satisfied with the care they received and improved their confidence in the health system [237]. Another study in an acute care setting found that IPC resulted in a decrease in readmissions and an overall decrease in catheter-associated urinary tract infections over time [222].

IPC has also been found to benefit professionals in primary health care settings [238], and IPC also benefits members of the health care team and patients in studies on palliative care and care for the elderly [237, 239].

In concert with the increasing attention on IPC, a number of competency frameworks have been developed to assess interactions among health professionals in different settings [142, 239-243]. In general, the frameworks assess power imbalances among team members, communication, shared responsibility, cooperation, and coordination [142, 239-243]. These competency frameworks have been criticised for limiting innovation and interfering with inter-professional practice [244], variations in quality since they were designed for specific populations and purposes, insufficient validation in different geographical settings and are validated in various settings [245]. Nonetheless, the frameworks are useful in generating empirical information on IPC and in pointing to areas of improvement. These competency frameworks are discussed briefly below:

The Perception of Interprofessional Collaboration Model-Questionnaire (PINCOM-Q) measures perceptions and behaviour between professionals in the interprofessional collaboration process on an individual, group and organisational level [243], but was still in development stage at the time of the study.

The Collaboration and Satisfaction About Care Decisions (CSACD) is a framework developed to measure physician/nurse collaboration in critical care units and assess power imbalances between team members. It focused on planning, communication, shared responsibility, cooperation, and coordination [240]. However, it was designed for critical care units that deal with life and death situations, and is therefore inappropriate for measuring IPC among CLP team members. The Survey of Attitudes Toward Physician-Nurse Collaboration, has 4 subscales: teamwork, caring as opposed to curing, nurse's autonomy, and physician's dominance [241]. It is limited in its assessment of collaborative elements among team members.

The Interdisciplinary Team Performance Survey (ITPS) is reported to measure team process and predictors of team performance. These include leadership, coordination, communication, conflict management, team cohesion and team effectiveness [239]. It addresses some components of collaboration but focuses on team meetings more than team functioning or on IPC.

The Assessment of Interprofessional Team Collaboration Scale (AITCS) was developed to assess how health care teams form and function in their collaborative relationship. AITCS consist of 47 items within 4 subscales (partnership, cooperation, coordination, and shared decision making) [242]. However, the framework has not been validated [246].

The Registered Nurses' Association of Ontario (RNAO) competency framework describes the competencies required for effective interprofessional collaboration. It proposes seven competency domains that highlight the knowledge, skills, attitudes and values that are essential for interprofessional collaborative practice. The seven domains has 51 items shown in Table 6.1 [142], nonetheless this framework has also not been validated.

WHO has pointed out that IPC by itself will not achieve the desired outcomes, but that it requires a set of enablers, and overcoming constraints and challenges [79]. The factors that enable IPC include: visionary leadership; institutional support; mentorship and learning; and enabling practice environments [79, 224, 226]. A multi-country case study on IPC in primary health care in Brazil, Canada, India, South Africa and the USA recommended key success factors for inter-professional education and collaborative practice [224]. These include: shared vision, shared governance, government infrastructure, supportive legislation for health and education sectors, dedicated funding and resources, and strong linkages between academia and clinical sites [224].

Research studies have also identified various barriers to IPC. Professional cultures and stereotypes, often created by the process of professional training and socialisation, and silo practice in many health care settings, hinder IPC [224]. Other barriers include curricula and accreditation requirements of health professions regulatory authorities, inadequate knowledge of the roles and scope of other health professions [224, 226, 247].

Individuals born with CLP require treatment and care from birth until early adulthood by a multi-disciplinary team of health professionals [114]. Studies have found that IPC among health professionals is a major contributor to success in cleft treatment [248, 249]. However, these IPC studies have been done in high-income countries. Furthermore, these studies have

not measured IPC across a number of domains of care expertise, collaborative leadership, shared power, effective group function, optimising professional role and scope, effective communication. In many African countries, including South Africa, IPC remains a fairly new concept [147, 250]. We could not find any studies in South Africa that have focused on IPC in the context of CLP or that have measured IPC using a validated competency framework.

There has been an increasing calls for inter-professional education in South Africa to facilitate IPC [147] and universal health coverage reforms [251]. In light of dearth of literature on the ideal IPC framework, the RNAO's framework [142] was chosen for this study because it has good face validity, the questionnaire is available free-of-charge, and it includes elements that examine the relationships between the doctor and the patient. Therefore, the aim of this study was to measure IPC among CLP health care teams in the public health sector using the RNAO's framework [142].

6.2 Methodology

6.2.1 Ethical considerations

The Human Research Ethics Committee (HREC) (Medical) of the University of the Witwatersrand in Johannesburg provided ethical approval (#M1501536) to conduct this research. Permission was also obtained from the relevant health care authorities. We adhered to standard ethical procedures, which included study information sheets, voluntary participation, informed consent, anonymity of responses, and confidentiality.

6.2.2 Study setting

The study setting consisted of all 11 CLP care centres situated in six of South Africa's nine provinces that provide specialised care to individuals with CLP.

6.2.3 Study population and selection

The study population consisted of all health professionals (HCPs) that are members of CLP teams at the 11 centres. At each of the centres, the principal investigator (PH) approached all the HCPs who were present on the day of the cleft clinic, explained the study verbally, handed each person an information sheet and invited these health professionals to participate in the survey.

6.2.4 Development of the self-administered questionnaire

The study used the IPC framework developed by RNAO [142]. The IPC framework consists of seven domains and 51 items shown in Table 6.1 below. Each item was assessed on a 4-point Likert scale as: (4 = always, 3 = sometimes, 2 = rarely, and 1 = never). There is also an option of "does not apply".

The framework was used in its original form, but a section on background and demographic information was added to obtain information on gender, age, professional category, continuous development and cleft qualification.

Table 6. 1 RNAO's IPC framework summarised

Domain	Description
Care expertise (8 items)	Collaboration among health –care professionals, as well as between them and patients and their families and circle of care in order to identify and take advantage of each person's care expertise
Shared power (4 items)	Willingness to share power as a commitment to create balanced relationship through democratic practices of leadership, decision making, authority and responsibility
Collaborative leadership (10 items)	Collaborative leadership (also called reciprocal or shared leadership) is a people – and relationship – focused approach based on the premise that answers should be found in the collective (the team)
Shared decision making (2 items)	Shared decision-making gives all team members, including patients, the opportunity to contribute their knowledge and expertise, to arrive collaboratively at an optimal goal
Optimizing professional role and scope (10 items)	Exemplary inter-disciplinary care let all team members work to their full scope of practice and takes advantage of the synergies professionals working together can create
Effective group function (9 items)	A health-care system that supports effective teamwork can improve the quality of patient care, enhance patient safety, and reduce workload issues that cause burnout among professionals
Competent communication (8 items)	Competent communication – openness, honesty, respect for each other's opinions and effective communication skills – is a part of all domains of inter-disciplinary practice

Source: RNAO[142]

6.2.5 Data collection

During 2016, a cross sectional survey was conducted at all 11 CLP care centres. Following informed consent, each HCP on duty at the CLP care centre, completed a self- administered

questionnaire. Each questionnaire took about 15 minutes to complete. The principal researcher conducted quality checks to confirm the completeness of the questionnaires. Data were cleaned and checked for inconsistencies before importing into STATA®13 for analysis. A "does not apply" response was set as a 'missing' value in STATA.

6.2.6 Data Analysis

Cronbach's alpha coefficients were calculated to determine reliability and coherence between items in the seven domains. These ranged from 0.73 to 0.93, indicating high reliability and inter-item correlation.

An overall mean IPC score was computed as well as a mean score for each domain. The responses on each domain were summarised using means, standard deviations, and ranges. In order to examine differences in mean scores, we classified the respondents according to gender, professional category, and CLP treatment centre. The professional categories were grouped into doctors (plastic surgeons, maxillo-facial surgeons, orthodontists, paediatricians and dentists) and therapists (speech therapists, geneticists, nurses, psychologists and social workers). We used ANOVA to analyse the differences in scores across the domains. All statistical tests were conducted at 5% significance level.

6.3 Results

6.3.1 Participants' characteristics

The participants' characteristics are shown in Table 6.2. A total of 52 participants completed the questionnaire. The mean age of participants was 41.9 years (range 22– 72) and the median was 40 years (inter-quartile range (IQR) 31.5 – 53). The majority were women (52%). The majority of participants were plastic surgeons (38.5%) followed by speech therapists (23.1%), nurses (9.6%), geneticists (7.7%), orthodontists (5.8%), maxillofacial surgeons (3.9%).

psychologists (3.9%), paediatricians (3.9%), dentist (1.9%) and social worker (1.9%). The HCP members of each centre ranged from two to eight. Three participants (5.8%) reported to have a specific CLP qualification and all reported participation in continuous professional development.

Table 6. 2 Descriptive characteristics of the participants

Characteristics	n (%)
Mean age in years (SD)	41.9 (1.8)
Median age in years (IQR)	40 (31.5 - 53)
Age range in years	22 - 72
Gender	
Male	25 (48.1%)
Female	27 (51.9%)
Professional category	**n=52**
Doctor	
Plastic surgeon	20 (38.5%)
Maxillo-facial surgeon	2 (3.9%)
Orthodontist	3 (5.8%)
Paediatrician	2 (3.9%)
Dentist	1 (1.9%)
Therapists	
Speech therapist	12 (23.1%)
Geneticist	4 (7.7%)
Nurse	5 (9.6%)
Psychologist	2 (3.9%)
Social worker	1 (1.9%)
Number of HCP per CLP Centre	**n= 52**
SITE 1 (GP)	4 (7.7%)
SITE 2 (GP)	5 (9.6%)
SITE 3 (GP)	3 (5.8%)
SITE 4 (GP)	5 (9.6%)
SITE 5 (GP)	8 (15.4%)
SITE 6 (WC)	6 (11.5%)
SITE 7 (WC)	6 (11.5%)
SITE 8 (KZN)	5 (9.6%)
SITE 9 (FS)	2 (3.9%)
SITE 10 (EC)	5 (9.6%)
SITE 11 (LP)	3 (5.8%)

GP – Gauteng Province; WC – Western Cape Province; KZN – KwaZulu Natal Province; FS – Free State Province; EC – Eastern Cape Province; LP – Limpopo Province

6.3.2 Mean scores for IPC domains

Table 6.3 shows the mean scores for each of the seven domains. The highest mean score of 2.92 was obtained for care expertise, whereas effective group functioning obtained he lowest score of 2.55. None of the domains obtained a mean score of 4, which means that they collaborate fully as a CLP team.

Table 6. 3 Mean scores for IPC domains

Domain	Mean	Standard Deviations	Cronbach - α
Care expertise (8 items)	2.92	0.37	0.83
Shared power (4 items)	2.67	0.50	0.88
Collaborative leadership (10 items)	2.72	0.45	0.92
Optimizing professional role and scope (10 items)	2.70	0.46	0.93
Shared decision making (2 items)	2.69	0.50	0.73
Effective group function (9 items)	2.55	0.50	0.92
Competent communication (8 items)	2.63	0.44	0.87

6.3.3 Mean IPC domain scores by explanatory factor

Table 6.4 shows the summary of mean scores by explanatory factor. The mean IPC scores by domain did not differ by gender. None of the scores differed between males and females. The professional category called "doctor" scored higher overall. Compared to the therapists, the doctor category was more likely to obtain higher mean scores for shared power ($p< 0.01$), collaborative leadership ($p<0.04$), optimising professional role and scope ($p<0.03$), effective group function ($p<0.01$), competent communication ($p<0.04$) and overall ($p<0.02$). The mean scores on the domains of care expertise ($p<0.0005$) and shared power ($p<0.01$) differed across the CLP care centres.

Table 6. 4 Mean IPC domain scores by explanatory factor

Explanatory Levels		Care expertise	Shared Power	Collective leadership	Shared decision	Profession, role & scope	Group function	Communication	Overall
Gender	Male	2.95	2.76	2.83	2,72	2.76	2.65	2.71	2.77
	Female	2.89	2.58	2.61	2.67	2.64	2.46	2.57	2.63
Professional category	Doctor	2.98	2.83*	2.88*	2.73	2.83*	2.70*	2.76*	2.81*
	Therapists	2.85	2.48*	2.52*	2.64	2.56*	2.38*	2.5*	2.56*
CLP centres	Site 1 (GP)	2.90*	3*	2.6	2.63	2.33	2.28	2.38	2.59
	Site 2 (GP)	2.38*	2.95*	2.62	2.7	3	2.53	2.38	2.65
	Site 3 (GP)	3.04*	2.83*	2.7	2.83	2.73	2.85	3	2.86
	Site 4 (GP)	2.8*	1.8*	2.22	2.6	2.52	2.24	2.53	2.39
	Site 5 (GP)	3.03*	2.56*	2.71	2.5	2.63	2.63	2.66	2.67
	Site 6 (WC)	3.08*	2.75*	2.85	2.67	2.5	2.43	2.61	2.7
	Site 7 (WC)	2.85*	2.58*	2.71	2.75	2.73	2.44	2.52	2.66
	Site 8 (KZN)	3.25*	2.9*	3.05	2.9	3	3	3	3.06
	Site 9 (FS)	3.1*	2.85*	2.92	2.8	2.86	2.8	2.93	2.89
	Site 10 (EC)	2.25*	2.75*	2.5	2.5	2.8	2.33	2.25	2.48
	Site 11 (LP)	3.08*	2.58*	2.87	2.83	2.73	2.85	2.71	2.75

*Differences are significant (P<0.05)

6.4 Discussion

This was one of the first studies to use the RNAO's [142] IPC competency framework to analyse the collaboration among CLP team members at the 11 care centres in the South African public sector. The findings from our study showed that overall IPC was sub-optimal. We could not find similar studies that used the RNAO framework or any other IPC framework, in order to compare our study findings. Although the domains overlap, the mean scores for each of the seven domains were also sub-optimal and are discussed below.

Care expertise

The domain of care expertise, which measures the collaboration among HCP, patient and their families, obtained a mean score of 2.92. There were no differences by gender or health professional category. The differences in mean scores were influenced by CLP centre. Site 2 in Gauteng (GP) Province obtained the lowest mean score of 2.38, while site 8 in KwaZulu-Natal (KZN) obtained the highest mean score of 3.25.

Shared power

The domain of shared power, which measures the willingness to share power in decision making, authority and responsibility, obtained a mean score of 2.67. There were no gender differences found on the willingness to share power. The doctors were more likely to report shared power than other categories of health professionals. Other studies have also found that there is a professional hierarchy, with more power invested in physicians who dominate decision-making, and this could be a barrier to IPC [252-254]. Site 1 in GP obtained the highest mean score of 3 while Site 4 in GP obtained the lowest mean score of 1.8.

Collaborative leadership

In our study, Site 8 in KZN obtained the highest score of 3 for the domain of collaborative or shared leadership whilst Site 4 in GP had the lowest score of 2.2. The doctors were more likely to report collaborative leadership compared to the therapists. Another study has argued for collaborative leadership is necessary to ensure that the work atmosphere supports the health care team to enable quality care to patients [255].

Shared decision making

This domain measured the extent to which all team members, including patients, have the opportunity to contribute their knowledge and expertise, to optimise treatment goals. Joint decision making enables sharing of knowledge with the other team members and learning from each other for the benefit of patients [256]. Our study findings showed that Site 8 in KZN had the highest score of 2.9 while Site 5 in GP and Site 10 in Eastern Cape (EC) had lowest scores of 2.5. There were no statistically significant differences by gender, professional category and care centres. The mean scores imply that shared decision-making in CLP care requires improvement.

Optimising profession, role and scope

This domain evaluated the inter-disciplinary model of care that allows all team members to work to their full scope of practice, taking advantage of the synergies created by professionals working together. The highest scores of 3 were obtained by Site 2 in GP and Site 8 in KZN for this domain, while Site 1 in Gauteng had the lowest score of 2.3. The doctors were more likely to report optimising profession, role and care compared to the therapists. Other studies have found that this inter-disciplinary model of care involving physicians, pharmacists and nurses improved the management of hypertension in individuals with chronic diseases [257, 258].

Effective group functioning

This domain measures the extent to which the health-care system supports effective teamwork. Site 8 in KZN obtained the highest mean score of 3 while Site 4 in GP had a lowest score at 2.2. There were statistically significant differences by professional category, with the doctors more likely to report effective group function compared to the therapists.

Competent communication

This domain focuses on openness, honesty, respect for each other's opinions and effective communication skills. Communication is important to convey consistent messages that improves patient care. Site 3 in GP and Site 8 in KZN had the highest scores of 3 while the lowest score was obtained by Site 4 in GP at 2.2.The doctors were more likely to report higher scores on competent communication compared to the therapists.

A study in Nigeria found that insufficient number of health professionals and sociocultural issues hindered IPC among health care team members responsible for CLP [259]. The staff numbers found in our study are also small compared to staff numbers of CLP centres reported in other parts of Africa [104], Brazil [260], China [261], United Kingdom [13] and United States [157]. Hence, these small numbers influenced the scores.

Although the study was limited by the small sample size, we obtained the responses of all the health professionals involved in CLP care at each of the centres. Hence, we captured the universe of health professionals at each centre. The cross-sectional nature of the study means that we obtained the perspectives of health professionals at a point in time, using a scoring system. Further research is needed to determine the qualitative reasons for the differences in

centres scores. The potential social desirability bias was minimised by using a validated instrument, that was self-administered [145, 146].

There are several strengths of our study, which include the measurement of IPC using a validated instrument, obtaining baseline IPC data at all the CLP centres in the South African public sector, and initiating the discourse on IPC in the treatment and care of individuals with CLP.

Our study findings have implications for the treatment and care of individuals with CLP. The study revealed that the professional category, and to a lesser extent CLP centre, explained the differences in mean scores. In the majority of domains (shared power, collective leadership, optimizing profession role and scope, effective group function and effective communication), doctors obtained higher mean scores compared to therapists. Other studies have also found that doctors tend to dominate, both because of their training, professional status and socialisation [252, 262]. The complexity of the CLP condition and the long-term nature of treatment of individuals require a multidisciplinary team that practise IPC. The seven domains of the RNAO could be used to guide practical strategies to enhance IPC in the 11 CLP centres, starting with those that do not require additional resources. However, IPC requires institutional support and leadership, as well as mentoring and coaching to unlearn certain behaviours [79] [263]. Other strategies to enhance IPC include interdisciplinary round and clinical discussions, multidisciplinary meeting or video conferencing on patient management [256, 264].

6.5 Conclusions

IPC in CLP treatment and care has huge potential to enhance patient outcomes and quality of care. The study findings can be used as a foundation for improving communication and teamwork in CLP care, and promoting the coordination of complex care processes across the lifespan of the individuals with CLP.

6.6 Authors' contributions

PH conducted the study as part of PhD. LR is the primary supervisor of the PhD. JL is a biostatistician. PH and LR conceptualized and designed the PhD study. PH collected all the data, JL and PH conducted the statistics, with extensive input from LR. All authors analysed the data and contributed to the writing and editing of the manuscript, and read and approved the final version of the manuscript.

CHAPTER 7

Caregivers' Perceptions of Healthcare Provision and Support

7.1 Introduction

Congenital anomalies are a major public health concern worldwide, because of their contribution to infant and childhood mortality, chronic illness and disability [265]. Clefting of the lip with or without palate (CLP) is a major structural congenital defect that requires surgical intervention and has significant medical, social and psychological consequences for affected individuals and their families [265]. The worldwide prevalence is estimated at 1 in 700 live births, but this varies according to geographic location, socio-economic status and ethnic background [4, 266, 267]. In South Africa, CLP is amongst the five most common birth defects reported [92], with the prevalence ranging from 1.0 to 4.0 per 10 000 live births [92, 96, 193].

Research on CLP is extensive, and includes studies on prevalence, determinants and associated anomalies [92, 166, 193, 268-271], the diagnosis and medical management of CLP [128, 129, 198, 248, 272, 273]; feeding problems as well as speech, hearing, and dental complications [110, 195-197, 274]; long-term health outcomes [110, 195-197, 274, 275]; and the expressed communication needs and psychosocial issues faced by individuals with CLP and their parents [47, 125, 132, 217, 276-287].

Most of the research on experiences and perceptions of parents of children with CLP is in high-income countries (HICs) [125, 217, 248, 281, 282, 285]. In these countries, studies have found that parents experienced varying degrees of shock, anger, denial, distress and anxiety, and a sense of "loss of control" over the birth of a child with CLP [125, 217, 248, 281, 282,

285]. Mothers expressed feelings of guilt because they carried the infants to term [248]. In these studies, parents underscored the importance of appropriate and accurate information about CLP and health and social support with their children's condition at birth [217, 276, 278, 285, 287]. Some studies found that parents who had a delay in obtaining information from health professionals searched for this on the internet, which exacerbated their anxieties and distress [248].

In many low- and middle-income countries (LMICs), infectious diseases dominate the causes of infant and child mortality, and congenital anomalies account for a relatively small proportion of under-five mortality [179, 286]. Hence, there is a dearth of research on congenital anomalies in LMICs. Nonetheless, there is an emerging body of literature that describes parental emotions of shock, distress and anxiety [88, 278, 285, 288]. A study in Brazil found that frequent hospitalizations of children with CLP add to the parental feelings of stress, fear, anxiety and helplessness with the stress levels of parents higher prior to their children's corrective surgery [288]. Mothers of children with clefts expressed hurt after birth and attributed the cause of orofacial clefts to evil or ancestral spirits or the will of God. The parents also talked about the stigma experienced, as their children with oral clefts were often treated as outcasts in their communities [88, 283].

In a 2002 study from Pretoria, South Africa, in which the mothers' experiences of pre- and postnatal diagnosis of clefts were explored, it was discovered that the timing of diagnosis did not influence the need for emotional support, information, interaction with other parents of children with clefts, and a multidisciplinary team approach to care and treatment [285]. A 2014 study in a private hospital on parents' experiences of children with CLP found that parents were satisfied with treatment they received, but expressed dissatisfaction with

financial support for treatment, transport to the facility, and lack of parental education and information [278]. However, the experiences and views of parents in the public health sector might be different.

There remains a paucity of research on CLP in South Africa, and the perceptions and experiences of parents of children with CLP, especially studies that are representative of all provinces, of urban and rural settings, and of the public health sector. In light of insufficient scholarly attention on the psycho-social aspects of CLP as a congenital anomaly, this study focused on parental experiences of a child with CLP, and parents' perceptions of health care provision and support for their children in the public health sector.

7.2 Methods

7.2.1 Ethics considerations

The Human Research Ethics Committee (HREC) (Medical) of the University of the Witwatersrand in Johannesburg provided ethical approval to conduct this research (M1501536). Permission was also obtained from the relevant health care authorities. We adhered to standard ethical procedures, which included study information sheets, voluntary participation, informed consent, confidentiality, and anonymity in the management of data and reporting of study findings.

7.2.2 Study settings

We restricted the study to the public health sector in South Africa, as it provides health care services to the majority of the population in the country. The study setting consisted of all the 11 academic hospital centres that provide specialised care to individuals with CLP situated in six of South Africa's nine provinces. These centres were selected because they are referral

centres that provide specialised treatment to children born with CLP and multi-disciplinary teams of health professionals are available at these centres

7.2.3 Sampling

Although the number of participants in qualitative studies depends on the point at which data saturation is reached [289], we considered it important to obtain the views of parents at each of the specialised treatment centres. At each of the CLP care centres, we aimed to select a minimum of five and maximum of ten parents or caregivers of children with CLP, to take account of size and the catchment area of the specialised treatment centre. We selected caregivers purposively among those who attended the CLP clinic on the fieldwork day, and selected them as they arrived at each centre.

7.2.4 Data collection

On the fieldwork day, the principal investigator (PH) approached caregivers present at the relevant specialised treatment centre, explained the study verbally, handed an information sheet to each participant, and requested participation in the study. Those individuals who agreed to participate in the study were enrolled and in those instances where both parents were present at the clinic appointment, the mother was interviewed. Following informed, written consent, the PI used a semi-structured questionnaire (Appendix 7), designed in English, to conduct the interviews with the caregivers. We designed the questionnaire specifically for the study. The questionnaire contained 27 questions divided into three parts: socio-demographic information, the parental experiences of having a child with CLP, reactions from families, friends and the general public; perceptions of health care provision; and the availability of support services.

At each centre, PH conducted the interviews in a private room adjacent to the consulting room. The interviews were conducted in English, with clarification of terms into one of South Africa's local languages, where relevant, as PH is fluent in all local languages. Each interview lasted around 30 minutes. The responses of participants were written down verbatim, typed up and saved as individual Microsoft Word © documents soon after the interview.

7.2.5 Data analysis

The interviews were analysed using thematic content analysis [290]. The first step in the analysis was to look at participants' own words and phrases and without preconceived notions or classification. We then examined the language used by each participant in response to the following questions: What do the responses tell us about the experiences, feelings and perspectives of parents or caregivers? What is emerging about the experiences associated with having a child with CLP, health care provision or social support services?

To ensure trustworthiness, two additional researchers (an experienced qualitative researcher with health system experience and the primary supervisor) participated in the development of the codes by reading the parent responses independently from the PI in order to establish inter-coder agreement [290, 291]. Once the initial analysis was completed, the team liaised to discuss the codes generated independently, and to reach agreement on the codes and themes. Following inter-coder agreement and the themes, the PI analysed the interviews [290].

7.3 Results

7.3.1 Socio-demographic characteristics of participants

All parents or caregivers that were recruited at the specialised treatment centres, agreed to participate in the study; hence a 100% response rate was obtained. The study recruited 79 participants: 68 were biological parents, four were guardians, five were relatives, and there was one foster parent and one caregiver with a child from an orphanage. The mean age of the participants was 33.3 years (range 17-68 years). The majority of the participants were black African (72%), unemployed (67%), single (67%) and with only primary school education (58%). The majority of children were male, with a mean age of 3.8 (SD= ±4.3) years (Table 7.1).

Table 7. 1 Social and demographic characteristics of caregivers

Characteristics	n=79
Mean age in years (Standard Deviation)	33.3 (17.2)
Relationship to CLP Child	
Biological Parent	68 (86.1%)
Foster Parent	1 (1.3%)
Relative	5 (6.3%
Guardian	4 (5.1%)
Caregiver	1 (1.3%)
Race	
Black African	57 (72.2%)
Coloured	11 (13.9%)
Indian	2 (2.5%)
White	9 (11.4%)
Employment status	
Employed	22 (27.8%)
Unemployed	57 (72.2%)
Education status	
Primary school	46 (58.2%)
Secondary school	31 (39.2%)
Tertiary	2 (2.6%)
Marital status	
Married	26 (33%)
Single	53 (67%)
Children in each family including CLP Child	
Number of children – mean (Standard Deviation)	2.2 (±1.3)
Mean age in years (Standard Deviation)	3.8 (±4.3)

7.3.2 Caregivers' experiences and perceptions of health care provision and support services

Although overlapping, five broad themes emerged from the responses of study participants to the open-ended questions: emotional experiences following the birth of a child with cleft lip and palate; reactions from family, friends or the public; the burden of care experienced by caregivers; health system responsiveness and social support services (Table 7.2).

Table 7. 2 Caregivers' experiences and perceptions themes

Theme	Description
Emotional experiences of having a child with CLP	**Mother** • Self-blame • Shock • Feeling Scared • Crying • Acceptance of the child **Partner** • Acceptance of the child • Denial of the child • Supportive **Concerns about** • Speech • Schooling • Long term well-being
Reactions from family, friends and the public	**Family and friends** • supportive **Public** • Gossiping • Staring • Asking lots of Questions
Burden of care	• Feeding needs and challenges • Need for full-time and extra care • Many hospital visits - Transport costly - Frequent time off from work - Inability to work
Health system responsiveness	**Health provider attitudes and behaviour** • Pleasant attitude • Rude behaviour • Caring attitude **Treatment provision** • No payment for treatment • Satisfied with treatment • Lack of resources for mothers **Inadequate or lack of communication** • Little or no Information • Inadequate Counselling • Little or no explanation
Social support services	• Lack of public awareness • Nuances of counselling

7.3.3 Emotional experiences following the birth of a child with cleft lip and palate

A major theme that emerged from the caregivers' responses was the emotional experiences following the birth of a child with cleft lip and palate. The birth of a child with CLP evoked mixed feelings of shock, anxiety, distress, worry, sadness, and misery among parents because they did not know that their new-born baby would be born with a cleft lip and palate. Many mothers reported that they cried uncontrollably. At the same time, they also experienced happiness about their live born children, and some expressed a combination of feelings of shock and acceptance of the child "as a gift from God."

I was shocked because I have not seen a cleft before, but I accepted the baby as a gift from God (Parent 6; Site 8, KwaZulu Natal Province).

Another parent reported screaming and crying when she was shown her baby at birth:

I screamed and was shocked when they showed me the baby. I asked: "what happened to my baby?"... I cried. (Parent 10; Site10, Free State Province).

Most parents blamed themselves and expressed feelings of guilt because of the birth of a baby with a cleft lip and palate condition, especially those parents who reported smoking and alcohol during pregnancy.

I blame myself because I was smoking. I caused my baby to have the cleft (Parent 6, Site 6, Western Cape Province)

Some parents ascribed the CLP to delay in the first antenatal care visit to a health care facility or because they desired a baby with a different sex, than the baby who was born.

I am guilty because I went to hospital later and not early enough when I was pregnant (Parent 5; Site 5, Gauteng Province).

I think it is my fault because I wanted a girl so God did the opening in the mouth because he is a boy (Parent 1; Site 11, Limpopo Province).

The interviews revealed that there were differences in the reactions between mothers and fathers. In some instances, the fathers were reported to be supportive and loving.

The father loves her even though it was the first time he sees this since no one in our family has it (Parent 2; Site 6, Western Cape Province).

The father is supportive even though he does not live with us in the same house but he gives us money for milk, clothes and hospital visits (Parent 9; Site 4, Gauteng Province).

However, there were instances where mothers reported that the fathers rejected them and the babies. Some fathers denied the babies as theirs, either claiming that they do not "give birth to disabled children", or left and never returned.

The father denied the child saying he does not want an abnormal child (Parent 4; Site 8, KwaZulu Natal Province)

His father denied that it is his child immediately when he saw that he has a cleft condition (Parent 1; Site 9, Eastern Cape Province).

Many parents also expressed concerns about speech difficulties experienced by their children with CLP, which in turn impacted on future schooling and peer acceptance. They wished that their children should attend normal school and not a special school for children with disability. They also expressed concerns about the overall well-being of the child, hoping that the medical team would help their babies to become "normal":

We wish that the hole must be closed inside [his mouth] so that he is able to speak properly as he will be 4-years old next year and he must attend preschool (Parent 3; Site 11, Limpopo Province).

I wish that my child must be well; be able to go to school and be successful (Parent1; Site 9, Eastern Cape Province).

I wish that he must have normal schooling; today the medical world is much better equipped to handle this condition and I am confident that he will be helped (Parent 4; Site 4, Gauteng Province).

I do not know what to say to him when he is old, my son is scarred for life; I wish that he must be helped to be normal (Parent 4; Site 7, Western Cape Province).

7.3.4 Reactions from family, friends or the public

The nature and extent of support from family and friends was the second theme that emerged from the interviews. In some instances, caregivers expressed appreciation to their families and friends for their caring and understanding.

My family accepted my baby and they were supportive (Parent 5; Site 11, Limpopo Province).

There is no conflict in our family because of the child, the family has been working together to make sure that everything goes well with him (Parent 6; Site 8, KwaZulu Natal Province).

In other instances, mothers reported alienation from people they considered as their friends, who distanced themselves. This was interpreted by the mothers as their friends not wanting to hurt them [the parents] because they had not seen the cleft before.

In the case of children with isolated cleft palate, the parents indicated that they were not ashamed of their children in public because the hole was inside the mouth and could not be seen by other people. However, the majority of parents said that they were not comfortable or happy to show their children in public, because of stares, hurtful comments and/or gossiping. They expressed concerns about post-natal clinic visits indicating that mothers with "normal" children were always looking at their cleft children with curiosity.

I was very much ashamed especially in the clinic where other mothers were always looking at her; it was not nice (Parent 1; Site10, Free State Province)

Many parents reported that they only felt comfortable with the child in public after the lip surgery was done, commonly after three months of birth.

I was not comfortable with him in public before surgery because people look and ask lots of questions, but now [after surgery] it is okay to show him in public (Parent 1, Site 7, Western Cape Province).

I was not [comfortable], until the surgery was done; I keep her in the house as the whole village was gossiping (Parent 2; Site 8, KwaZulu Natal Province)

We did not show him in public until he was operated. We were scared of germs and also of people looking (Parent 5; Site 11, Limpopo Province)

I have accepted to show him in public places now that he is 8-years old but when he small it was very embarrassing because people were asking what is this (Parent 7; Site 4, Gauteng Province)

7.3.5 Burden of Care Provision

Another major theme that emerged from the interviews with parents was the burden of caring for a child with cleft lip and palate. This burden was expressed through the comments on the feeding needs and challenges of their children; the need for extra care or attention, and the necessity of "many" hospital visits to different health care providers.

Feeding difficulties were a constant thread in most of the interviews. The mothers reported that they were scared of the child choking during feeding because of the hole between the mouth and the nose. The comments below illustrate the caregivers' distress about feeding of their children:

I compared this baby to the older child who was normal- feeding is a problem as milk comes through the nose (Parent 2; Site 6, Western Cape Province).

Feeding is a problem because milk is coming out through the nose so we have to watch her all the time so that she is not choking (Parent 1, Site 10, Free State Province)

Caregivers reported that children with cleft lip and palate need greater attention because of feeding difficulties, the perceived vulnerability of these children relative to "normal" children, and frequent visits to health care facilities. Parents reported that they were not comfortable with leaving their children in the care of others. One said:

I have to be with my child all the time and I do not trust other people with her (Parent 6; Site10, Free State Province).

Others commented as follows:

This child needs more attention as he is a special child so I dedicate all my time for him to be well (Parent 4, Site 11, Limpopo Province)

Yes, my child needs more attention from me as my family were scared of her so I was the only one feeding her (Parent 2, Site 4, Gauteng Province)

Frequent hospital visits added to the burden of caring, because some parents in more rural areas had to travel long distances to specialised treatment facilities. Furthermore, the majority of parents used public transport which was costly especially to those who were unemployed. Others raised concerns that the frequent hospital visits and requesting time off from work affected their employment.

I had to take two weeks leave for hospitalization when my child was operated. Taking time off from work for the hospital appointment affected my way of life, I don't get paid for being absent so many times from work (Parent 3; Site 5, Gauteng Province).

Some parents reported that they were unable to work or look for work, because they had to take care of their children.

7.3.6 Health system responsiveness

Some mothers reported that the treatment they received was good and that both nurses and the doctors provided information, counselled them, and reassured them, which made it easier to accept their babies with CLP.

I received good information from the nurses and doctor, they reassured me that my baby will be okay (Parent 10; Site 4, Gauteng Province).

The hospital doctors gave us the information about cleft, they told us that many other children are born like this and he [the baby] will be okay (Parent 1; Site 7, Western Cape Province).

The nurses and doctors were very nice to me and my child and they explained that my child is not the only one with this condition, so he will be helped (Parent 7; Site 8, KwaZulu Natal Province).

The comments about good quality of care tended to be at academic hospitals in large cities, because of the availability of a multi-disciplinary team of health professionals to support the parents.

However, the majority of parents reported several health system deficiencies that affected the care of their babies. These deficiencies included lack of information regarding the condition of their babies at birth, lack of explanation about CLP, and unhelpful and rude behaviour and attitudes of nurses at maternity clinics. These deficiencies contributed to their feelings of shock and distress.

I was never told until the next morning. I discovered it [cleft] myself when the baby was in the incubator. (Parent 7; Site 2, Gauteng Province).

[The cleft lip and palate] information was not enough, I used the internet and got more information about it [CLP] (Parent 2; Site 11, Limpopo Province).

I was not treated well. They [the nurses] were talking to each other and did not tell me anything about my baby, later they informed me that my child is not complete (Parent 8; Site1, Gauteng Province).

I was treated badly by the nurses, I was placed in a separate room and I was not told about my baby while the nursing staff was gossiping (Parent 4; Site 8, KwaZulu Natal Province).

Parents reported that they were happy with the free surgical treatment provided.

The doctors were helpful to both my girls, they [the girls] have been operated for free and they look pretty (Parent 4; Site 6, Western Cape Province)

The doctors are treating all these children very well; my child's lip was operated and I am very happy (Parent 6; Site 4, Gauteng Province)

I am very grateful to the doctors for fixing my son (Parent 6; Site 8, KwaZulu Natal Province)

I am happy with treatment even though it was delayed. He should have been operated when he was 6 months old, now he is 9 months. I was left behind by the transport to travel for treatment as it is not done in this hospital (Parent 1; Site 9, Eastern Cape Province)

7.3.7 Social support services

Social support service was the fifth theme that emerged from the parents, and includes the lack of public awareness and lack of counselling received by the mothers at the birth of their children with CLP. The majority of parents reported that they have never seen a cleft before and it was the first time in their family for a child to have this condition. Parents reported lack of counselling regarding this condition at the birth of their children.

I want the doctors and nurses to make public awareness especially in rural areas. I suffered for two months and my child could have died from the milk. The nurses in the clinics should know about cleft, and that it can be treated (Parent 2; Site 9, Eastern Cape Province).

I did not know at birth that my child has a cleft. I only saw the milk coming out the baby's nose and I was thinking that he has a small stomach and I am feeding him too much milk (Parent 3; Site 10, Free State Province).

I was not told what a cleft is and what causes it until today as I am speaking with you. You are the first person to explain cleft palate to me (Parent 1; Site 8, KwaZulu Natal Province).

Some parents reported that the hospital did not provide accommodation and lodging for them when their children were admitted for the surgical procedure. This was cited as a challenge because the parents' homes were far from the hospital and they had to sleep on the chairs for a number of days without food and facilities for their personal hygiene.

The nurses must have patience with us. When my child was admitted for the lip surgery, I slept on a chair for seven days waiting for my child to be discharged from hospital and did not have food. I wish that the hospital must give us [mothers] a place to sleep when we wait for our babies' surgery (Parent 5; Site 8, KwaZulu Natal Province).

7.4 Discussion

This is one of the first, comprehensive studies that explored the socio-demographic characteristics of the caregivers of children with CLP, the family impact of having a child with CLP, and their perceptions of health care provision and support services available at the

11 specialised academic centres in the South African public sector. The majority of study participants were single, unemployed, women who had completed primary schooling.

Although we did not ask about the nature and availability of family support, the findings suggest that the responsibilities for care provision of these children with CLP fall disproportionately on these women. Studies in the United States and the United Kingdom reported that the long term implications of parenting children with clefts include higher stress levels, anxiety and depression, a risk of maternal detachment, and lower cognitive functioning of the children [248, 292, 293]. Hence, the single mothers may require additional health and social service support. Furthermore, their socio-demographic characteristics need to be taken into account in health information, education and communication programmes in the South African public health sector. The specialist multi-disciplinary cleft team and the community healthcare services can play an important role in implementation of such programmes.

This PhD research provides unique parental perspectives of the effects on the family of children born with clefts. In our study, the participants expressed varying degrees of emotional reactions similar to studies done in other countries [104, 125, 217, 248, 281, 282, 285, 294]. Amidst these negative emotions, some parents expressed the view that the child is "a gift from God". This view may serve both as explanation of causation by a supernatural force, and as a coping strategy. A study from Nigeria also reported that Muslim parents who belonged to a specific ethnic group attributed CLP to the "will of God". The authors pointed out that the meaning of birth defects and the future faced by individuals with CLP are influenced by a complex set of social and cultural factors, that should inform the provision of health care services [295].

In our study, mothers' self-blame and feelings of guilt were both reinforced and exacerbated by the rejection of the babies' fathers and the courtesy stigma experienced from friends, the public, and uninformed nurses in health care facilities located outside the specialised academic centres. The notion of courtesy stigma, first described by Goffman [296], is the public disapproval evoked because of the association with a stigmatised individual or group, in this case a child with CLP. In this study, such courtesy stigma was expressed through staring, gossiping, and excessive questioning that made mothers feel uncomfortable and/or ashamed. A Ugandan study also found stigma narratives from mothers who reported that children with CLP were treated as "outcasts", while doctors reported that these children were not accepted by their communities [297]. Similarly, families may see a child with CLP as a "family shame", a feeling that may influence the social position of such families. A ten-year review done by the Smile Train of their work in 33 African countries found that stigma influenced case reporting, family relations, utilisation of health care services, and ability to conduct research [298]. Our findings point to the need for public awareness campaigns on CLP, as part of the South African Department of Health's overall strategic plan on non-communicable diseases. Nurses, who work in health facilities outside academic facilities, and in rural areas where there are no specialised treatment centres, require continuing professional education on CLP.

The mothers gave moving accounts of the burden of caring for a child with cleft lip and palate, especially feeding difficulties before the surgery was done and the necessity of numerous hospital visits to different health care providers. Feeding problems of babies with CLP at birth are universal, and detailed guidelines exist on feeding approaches, advice and information for mothers, and the management of complications. These guidelines could be adapted both for parents as well as for health professionals outside specialised health facilities.

The mothers' narratives on the burden of care should be viewed within the context of the emotional impact of children with CLP, and the experiences of courtesy stigma. A study in Brazil found that frequent hospital visits added to family stress and anxiety [288]. In this study, the mothers' narratives of the health system deficiencies were more acute in rural and non-specialised health care facilities, and this may worsen their stress. The inequities and complexities of care provision in the South African public health sector have been well described, and are illustrated through the comments on the "free and good" treatment they received, as well as the information, counselling and reassurance from doctors and nurses in the specialised academic centres. This means that there is a solid foundation to build on and opportunities to expand good practice models on information, counselling and social support to rural and non-specialised health care facilities. This is important because evidence suggests that lack of parental social support is one of the factors that influence the nature and quality of the attachment children form with their main caregivers [47, 248].

In light of the 2015 Sustainable Development Goals that emphasise universal health coverage and inclusive societies, this study provides important information on the issues that need to be taken into account in the provision of health services that are responsive to the needs of caregivers and children born with CLP. The study did not explore the use of traditional healers, or existing family support mechanisms. These are study limitations. Nonetheless, it is one of the first studies that explored the experiences of families on their child with CLP, and caregiver perspectives on health care and social service provision in the public health sector. The study participants were selected purposively, and represent all the specialised academic centres in South Africa's public health sector, rather than the views of parents at one centre. The study has generated new knowledge on health system deficiencies for

individuals with orofacial clefts in South Africa, which can be used to shape future treatment efforts, both by multi-disciplinary teams at specialised centres and community-based services.

The experience of well-established CLP treatment facilities in HICs provide a useful framework for care provision, that is applicable to resource-constrained settings [248]. These included: customised information and communication programmes that enhance the understanding of CLP among parents; genetic counselling as part of the package of care; teaching parents the skills to handle and feed the baby to enhance their confidence and to reduce anxiety; forums or opportunities for parents to express their feelings and come to terms with taking care of a baby with CLP; informing parents about treatment, care and rehabilitation of their children, and the members of the health care team; social support, which could include the child support grant, access to other parents, and parent support groups [248].

7.5 Conclusions

This study has highlighted the perceptions and the experiences of caregivers regarding health care service provision and support for their children with CLP in the South African public health sector. The study underscores the importance of sufficient information regarding cleft lip and palate condition, supportive families and friends, and optimal functioning of the health care system.

The study findings have implications for the integrated management of individuals with cleft lip and/or palate. Such management should include: addressing the information needs of parents and caregivers; the education and training of health care providers outside specialised centres; raising public awareness of birth defects, including CLP and social support programmes.

7.6 Acknowledgements

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7.8 Authors' contributions

PH is a PhD student. LR is the main supervisor of the PhD. PH and LR conceptualised and designed the PhD study. PH collected all the data, PH and LR coded the interviews with assistance of another researcher. All authors contributed to the writing and editing of the manuscript, and read and approved the final version of the manuscript.

CHAPTER 8

Discussion and Recommendations

8.1 Introduction

This mixed methods PhD study has generated new knowledge on cleft lip and palate (CLP) prevalence, treatment and care of the condition in the public health sector, inter-professional collaboration among health care teams, and the psychosocial issues faced by affected families. However, there is a policy vacuum on the overall management of congenital anomalies in general, and CLP in particular, evidenced by an outdated policy of 2005 [58]. Since the PhD research was conducted, a study of under-five mortality showed that the proportion of deaths due to non-natural causes, congenital disorders and non-communicable diseases has increased [67]. This suggests the need for health policy makers to develop pro-active strategies that prioritise prevention of these set of conditions, while at the same time ensuring cost-effective, high quality care for those individuals with CLP. The findings of this PhD study can contribute to the development of these strategies.

In this concluding chapter, I have integrated and discussed the key findings of this thesis (Chapters 4 to 7) and proposed recommendations for the comprehensive and integrated management of CLP in South Africa. In Section 8.2, I have summarised and discussed the key findings and recommendations of the PhD study. In Sections 8.3 and 8.4, I have highlighted the scholarly contribution and policy impact of the PhD respectively. Section 8.5 contains recommendations for further research.

8.2 Key Findings and Recommendations

8.2.1 Summary of key findings

The key findings of my PhD are summarised in Table 8.1 and discussed below.

Table 8. 1 Summary of the key findings of the PhD

Key PhD findings
• The estimated prevalence rate of CLP in South Africa's public sector was 0.3 per 1000 live births. • The clinical profile showed that in females, cleft palate (CP) was more common, whereas cleft lip and palate (CLP) was more common in males.
• Surgical repair of the lip and palate (10/11 centres) and speech therapy (7/11 centres) dominated the type of CLP treatment provided in the public health sector. • Although all 11 centres reported a multidisciplinary team approach for CLP care provision, there were gaps in the health professional categories, which in turn influenced the types of treatment provided. • Only six centres provided written, standard treatment protocols for CLP.
• Inter-professional collaboration was sub-optimal for all the seven domains of: care expertise; shared power; collaborative leadership; shared decision making; optimising professional role and scope; effective group function; and competent communication. • The highest mean score of 2.92 was obtained for care expertise, whereas effective group functioning obtained the lowest score of 2.55. • Professional group, and to a lesser extent CLP centre, explained the differences in mean scores.
• The majority of caregivers of children with CLP were single, unemployed, women who had only completed primary schooling. • Caregivers narrated their mixed feelings of shock, anxiety, and sadness at the birth of their children with CLP, and highlighted the burden of care provision, amidst their experiences of health system deficiencies and insufficient social support services.

8.2.1 Epidemiology of CLP

My PhD study was one of the first studies in democratic South Africa to estimate the prevalence of CLP in the public health sector (Chapter 4). Although the estimated prevalence

of 0.3 per 1000 live births is similar to the prevalence estimated more than three decades ago in the cities of Cape Town, Johannesburg and Pretoria [93-98], updated information is needed for health care planning and resource allocation. The clinical profile found of CP in girls and CLP in boys, was similar to existing evidence on CLP [87, 299, 300].

In my Ekhaya Lethu conceptual framework (Chapter 3), I proposed that the policy on congenital anomalies and population based surveillance system to be the foundation of the CLP comprehensive care. However, our study found that this foundation is weak since the surveillance system does not exist and the genetic policy has not been updated for more than 10 years. Therefore, our study findings should inform the development and implementation of a population-based surveillance system on congenital anomalies that was envisaged in the 2005 national policy [58]. WHO has noted that a population based surveillance system that is able to capture congenital anomalies accurately will enable the country to obtain accurate information on the burden of, and risks for, congenital anomalies, and refer affected infants to specialised centres for treatment and care [301]. The surveillance system will also inform prevention programmes and assist with health policy development [301].

8.2.2 CLP care

The estimated prevalence of 0.3 per 100 live births means that about 300 children are born with CLP each year in the South African public health sector. The condition of CLP results in a huge treatment and care burden, both on affected families (Chapter 7) and the health care system. This PhD was one of the first studies to analyse CLP treatment and care at all the 11 specialised academic centres. Surgical repair of the lip and palate (10/11 centres) and speech therapy (7/11 centres) dominated the types of CLP treatment provided in the public health

sector, with gaps in other types of treatment provided, There is a dearth of research on CLP treatment and care, except a 1953 study conducted at a Johannesburg private clinic [95].

The *Ekhaya Lethu* conceptual framework has proposed two essential pillars of care, namely system responsiveness and care environment. These pillars are weak, and the national policy should be updated. One of the issues to consider is the creation of a limited number of centres of excellence in cleft care. This will reduce the 11 centres through regionalisation, where for example the five centres in Gauteng Province could be reduced to two, one in Tshwane and the other in Johannesburg. These centres of excellence for cleft care would have the following benefits: increase in the critical mass of HCPs; enhance CLP expertise; development of standard treatment protocols; cost-effective CLP treatment; improved quality of care; create greater benefits for IPC; allow parent education and involvement in care; and facilitate research or evaluation of long-term treatment outcomes audits.

The roof of the Ekhaya Lethu is proposed to be protective and consists of a multidisciplinary team of specialised HCPs that work together in a coordinated manner. These HCPs can be organized through reviving of the South African Cleft Lip and Palate Association (SACLPA), which has ceased to function in the last ten years. The association will strengthen the discipline; provide the forum for continuing professional development, policy and health care advocacy. It has been shown where these associations are active in America [114], United Kingdom [302] and India [303], that CLP care is coordinated, and there is greater collaboration with one another to ensure the provision of the necessary treatment for a child with CLP as well as support and counselling to the parents.

8.2.3 IPC

IPC is fairly new in South Africa and there has been growing calls for inter-professional education to enhance IPC [147]. Our study was the first studies to use the RNAO's [142] IPC competency framework to analyse the collaboration among CLP team members. The findings from our study showed that overall IPC was sub-optimal. The comprehensive management of the CLP condition and the long-term nature of treatment of individuals require a multidisciplinary team that practise IPC. Our findings have implications for the improvement of IPC, which would be enhanced by centre and treatment reforms proposed above. These reforms will be possible through the regional approach of cleft care in South Africa. It has been shown that IPC requires institutional support, leadership, and that mentoring and coaching are enablers for IPC [79, 263]. Therefore, the revitalisation of SACLPA will encourage dialogue in IPC, enhance the discourse on the concept and improve awareness of IPC among members of the health care team.

8.2.4 Caregivers' perception

Our study findings showed that the responsibilities to care for children with CLP fell mostly on caregivers who were single women, unemployed with only primary education schooling. My *Ekhaya Lethu* House of Care conceptual framework (Chapter 3) proposed that the parents or caregivers should form one of the pillars of Ekhaya Lethu and they must be well- informed, motivated, empowered and prepared to participate in the treatment of their children. The communities in which the caregivers reside are also important part of Ekhaya Lethu for the advocacy and mobilisation of resources as well as participation in parents' support groups.

This study findings highlighted several health system deficiencies that were experienced by caregivers and these exacerbated the emotional trauma experienced by these parents. There is

a great need for public awareness campaigns on CLP, as part of the updated national policy on congenital anomalies. The study underscores the importance of HCPs' training on current approaches to CLP care, which would be enhanced through regionalisation of CLP services.

8.3 Summary and recommendations

In this section, I have drawn on the study's conceptual framework presented in Chapter 3 and the key findings of the study to propose key recommendations (Table 8.2)

Table 8. 2: PhD study recommendations

Recommendations	Individual HCP	Authorities (National, Provincial)	CLP care centres	Caregivers of children with CLP
1. Proper record keeping and register of craniofacial anomalies through active population based surveillance system.	√	√	√	
2. Updating the national policy on congenital anomalies	√	√	√	
3. Improve access to CLP care for all individuals in South Africa through regionalisation of centres.	√	√	√	√
4. Raising public awareness on CLP	√	√	√	√
5. Development of national standard protocol for CLP treatment and management	√	√	√	√
6. Addressing the information needs of parents and caregivers	√	√	√	√
7. Enhance the education and training of health care providers on congenital anomalies	√	√	√	
8. Audit of CLP treatment outcomes	√	√	√	
9. Enhance IPC among CLP team members	√	√	√	√
10. Incorporation of IPE in training of HCPs as a strategy to improve IPC	√	√	√	
11. Development and support of parents support groups	√	√	√	√
12. Revitalisation of the SACLPA to develop standards of care, share continuous professional education, conduct and share research and monitor all HCPs involved in management of individuals with CLP	√	√	√	√

8.3 Scholarly contribution of the PhD study

8.3.1 Generating new knowledge

The PhD study has generated new knowledge on the epidemiology and clinical profile of individuals with CLP in South Africa.

Study 2 was the first comparative study that analysed the treatment and care of individuals with CLP in the South African public sector, and it generated new empirical knowledge on describing the members of the CLP team in South Africa, describing the types of treatment rendered and analysing the treatment protocols.

Study 3 measured IPC among health professionals who are part of the health care teams involved in the treatment of CLP, and generated new knowledge on IPC in this context. It was one of the first studies to be done in the country on IPC in South Africa, using a validated instrument.

Study 4 explored caregivers' perceptions on having a child with CLP, the psychosocial issues faced by parents and families, experiences of healthcare provision and social support for children born with cleft lip and palate. The study has generated new knowledge and has highlighted the importance of caregivers' involvement in the ongoing management of CLP.

8.3.2 Methodological innovation

This PhD study setting included all 11 specialised academic centres in the South Africa, therefore it is representative of CLP care in the public sector.

Studies 1 was the first study to include all individuals with CLP in public sector to estimate the prevalence of CLP, therefore it is representative of the data in the public sector.

Study 2 is the first to analyse CLP care in the South African public sector, and the findings can be used to improve management and care.

Study 3 on CLP team IPC would enhance the discourse on IPC, a subject that has not received much attention in South Africa. IPC will be enhanced by the inclusion of interprofessional education in the training of all members of multidisciplinary team [147].

Study 4 was also the first for South Africa, and can be used for similar studies in other congenital anomalies in South Africa, and in other LMICs.

8.4 The Policy impact of the PhD study

I am a Specialist Orthodontist in the Department of Orthodontics, Wits School of Oral Health, and am involved in the treatment of individuals with CLP. The publication of Study 4 and the findings of Study 1 and Study 2 facilitated the provision of the recommendations of the research to decision makers in the country at an early stage of the PhD study. I was invited by the NDoH, Directorate of Genetic Disease on a stakeholders' meeting to discuss strategies on cleft care and involvement of NGOs in cleft management, including national data on CLP (part of Study 1 and 2).

Following the NDoH meeting of CLP stakeholders, I was invited to be part of discussion with the Smile Foundation, an NGO operating in South Africa, to collaborate on the development of CLP surveillance in South Africa (part of Study 1).

In 2017, I was part of the panel on CLEFT17 International Congress on Clefts in India, where we were grouped to present under the session "*Challenges for developing cleft care in Africa."* This session had representative from six African countries including Uganda, Ghana, Nigeria, Zimbabwe, Nigeria and South Africa. I was the representative for South Africa, and I presented my pilot study results (Part of Study 4).

My PhD has also triggered interest in the topic of orofacial clefts at the Wits School of Oral Health Sciences. This was illustrated by the hosting of a *Symposium on Cleft Deformities* in August 2018. I was liaising with organising the committee and I presented a paper entitled "*Psychosocial aspect of cleft lip and palate treatment,*" as part of my PhD Study 4. This symposium attracted multidisciplinary presenters including maxillofacial surgeons, orthodontists, plastic surgeons, prosthodontists, speech therapists, and geneticists. The NDoH was also part of the symposium, and officials have expressed an interest in continuing discussions around policy development.

8.5 Recommendations for further research

This PhD study was a descriptive study on orofacial clefts as an important first step. Numerous possible areas for future research are listed below:

- Predictors of trends and variations in epidemiology and clinical profile of CLP in South African provinces.
- A study on genotypic-phenotypic correlations of orofacial clefting in South Africa.
- An evaluation of clinical outcomes, and the quality of care comparisons across the 11 academic centres.
- Comparison of treatment and care of individuals with CLP between the public and private health sectors, including the extent to which CLP treatment is performed in the private sector.
- Evaluation of the human resource challenges to service provision for children with CLP.
- The economic and financial costs of the management of individuals with CLP in the public health sector, to assist with resource allocation decisions.

- The differences in IPC scores among specialised centres and professional groups, and the reasons for these differences.
- Caregivers' experiences of utilising alternative source or model of care.
- Quality of life for individuals with CLP.

8.6 Conclusions

A population-based congenital anomalies surveillance system is essential to ensure the collection of accurate information that will assist with health policy development, prevention strategies, planning of service delivery, and long-term treatment outcomes. This PhD generated new knowledge on the epidemiology and care for individuals with cleft lip and palate in South Africa. The *Ekhaya Lethu conceptual model* is proposed for the comprehensive management and care of individuals with CLP. The comprehensive management of CLP is likely to guide the development of a national clinical protocol, promote inter-professional collaboration, enhance public awareness campaigns on CLP, promote continuing professional education on CLP, and promote equal access to care for all children born with CLP in South Africa. This would be in line with the provisions of the South African constitution that aim to 'put children first."

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APPENDICES

Appendix 1 Ethics Clearance Certificate

R14/49 Prof. Phumzile Hlongwa

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

CLEARANCE CERTIFICATE NO. M150536

NAME: (Principal Investigator)	Prof. Phumzile Hlongwa
DEPARTMENT:	Oral Health Sciences 4 Gauteng Province Hospitals, 3 Western Cape Hospitals, King Edward VIII Hospital, Eastern Cape : Nelson Mandela Academic Hospital, Free State : Universitas Hospital Polokwane-Mankweng Hospital Complex
PROJECT TITLE:	Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa
DATE CONSIDERED:	29/05/2015
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Prof Laetitia Rispel and Prof T Dandajena
APPROVED BY:	Professor P Cleaton-Jones, Chairperson, HREC (Medical)
DATE OF APPROVAL:	18/11/2015

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 Secretary in Room 10004, 10th floor, Senate House, University.

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**.

PHlongwa — Principal Investigator Signature

18/11/2015 — Date

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

Appendix 2 CLP Team Participants Information Sheet

Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa

Introduction

Good day. My name is Phumzile Hlongwa. I am conducting a study on cleft lip and palate (CLP) in order to determine the epidemiology and care of individuals with CLP in SA. The study is being done for a postgraduate degree.

Why are we doing the study?

The purpose of the study is to obtain information from the professional team treating individuals with CLP on who are the members of the team and how well they work together. We hope that the information obtained from the study will be used to assist us to improve the provision of CLP treatment services in the country.

How we will do the study?

The study will obtain information of all individuals with CLP treated at the 11 academic centres in South Africa from 1 January 2012 to 31 December 2013. The researcher would like to ask the professional team questions regarding their service for CLP. It should take around 30 minutes to answer these questions.

The team members will be requested to participate in a survey by answering a questionnaire about their personal experiences regarding CLP care. Our role is to listen and to understand what you are saying. There is no right or wrong answer. How you answer the questions will not affect your work in the hospitals and we will not tell the hospital management what *you* say to us. We will put your answers into an overall report and nobody will know who said these things. We cannot help you with any complaints that you have with the hospital and we cannot tell the hospital management if you are happy with the services.

How do I know that the information I give will be kept confidential?

The information that you provide to me will be kept confidential and will be used for the purpose of this study only. Everything that you say will be treated as private and confidential. I will complete the questionnaire with the answers that you give, but I will not tell people your name or any other information that could be used to identify you. This means that no other person apart

from my supervisors and me will know that you took part in the study, or know how you answered the questions. The answers given by you and other participants will be looked at and the combined information will be written-up in a form of a report. Your name will not be used in any report.

Did you get permission to carry out the study?

Permission to carry out this project was obtained from the University of the Witwatersrand Research Ethics Committee. The health department has also approved the study.

Will there be any benefits from participating?

There will be no direct benefits to anyone who participates in the interviews, as participation is voluntary. This means you have the right to refuse to answer the questionnaire or if you do agree to answer the questionnaire you can leave out the answers you are uncomfortable with and you can stop the interview at any time.

Will there be any harm from participating?

We don't foresee any risks to you taking part in the research apart from taking up your time. If you feel upset during the interview, we can stop the interview at any time. It is not a problem if you don't want to be interviewed. However, we would really appreciate it if you do share your thoughts and feelings about the questions we will be asking you. We hope that the information we will get from you will be used to help us to improve treatment services for individuals with CLP.

Who do I contact if I want to ask more questions?

We will be happy to answer any questions you have about this study. The University of the Witwatersrand Research Ethics Committee has approved this research. If you have any questions about your rights as a study participant, or questions or concerns about any aspect of the study, you may contact the Chairperson of the Ethics Committee, Professor Peter Cleaton-Jones, on (011) 717 1234. If you have questions about the research, you may also contact the principal investigator: Phumzile Hlongwa on 072 664 4664.

Thank you for your assistance with this study.

Appendix 3 Parent Participants Information Sheet

Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa

Introduction

Good day. My name is Phumzile Hlongwa. I am conducting a study on cleft lip and palate (CLP) in order to determine the epidemiology and care of individuals with CLP in SA. The study is being done for a postgraduate degree.

Why are we doing the study?

The purpose of the study is to obtain information from the parents and /or care givers of children with CLP regarding the support services they receive for their children. We hope that the information obtained from the study will be used to assist us to improve the provision of CLP treatment services in the country.

How we will do the study?

The study will obtain information of all individuals with CLP treated at the 11 academic centres in South Africa between 1 January 2012 to 31 December 2013. The researcher would like to ask parents and/or care givers of children with CLP few questions regarding the support services and their perception of CLP services. It should take around 30 minutes to answer these questions.

Mothers of children with CLP are going to be asked question about their personal circumstances regarding their child with CLP. Our role is to listen and to understand what you are saying. There is no right or wrong answer. How you answer the questions will not affect any future care that your child receives at any of the hospitals and we will not tell any staff what *you* say to us. We will put your answers into an overall report and nobody will know who said these things. We cannot help you with any complaints that you have with the hospital and we cannot tell the staff if you are happy with their services.

How do I know that the information I give will be kept confidential?

The information that you provide to me will be kept confidential and will be used for the purpose of this study only. Everything that you say will be treated as private and confidential. I will complete the questionnaire with the answers that you give, but I will not tell people your name or any other information that could be used to identify you. This means that no other person apart

from my supervisors and me will know that you took part in the study, or know how you answered the questions. The answers given by you and other participants will be looked at and the combined information will be written-up in a form of a report. Your name will not be used in any report.

Did you get permission to carry out the study?

Permission to carry out this project was obtained from the University of the Witwatersrand Research Ethics Committee. The health department has also approved the study.

Will there be any benefits from participating?

There will be no direct benefits to anyone who participates in the interviews, as participation is voluntary. This means you have the right to refuse to answer the questionnaire or if you do agree to answer the questionnaire you can leave out the answers you are uncomfortable with and you can stop the interview at any time.

Will there be any harm from participating?

We don't foresee any risks to you taking part in the research apart from taking up your time. If you feel upset during the interview, we can stop the interview at any time. It is not a problem if you don't want to be interviewed. However, we would really appreciate it if you do share your thoughts and feelings about the questions we will be asking you. We hope that the information we will get from you will be used to help us to improve treatment services for individuals with CLP.

Who do I contact if I want to ask more questions?

We will be happy to answer any questions you have about this study. The University of the Witwatersrand Research Ethics Committee has approved this research. If you have any questions about your rights as a study participant, or questions or concerns about any aspect of the study, you may contact the chair of the ethics committee, Professor Peter Cleaton-Jones on (011) 717 1234. If you have questions about the research, you may also contact the principal investigator: Phumzile Hlongwa on 072 664 4664.

Thank you for your assistance with this study.

Appendix 4 Record Review Form

Epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa

For official use only

1. Participant number		☐☐☐☐
2. Study site	☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5 ☐ 6 ☐ 7 ☐ 8 ☐ 9 ☐ 10 ☐ 11	☐☐☐☐☐☐
3. Date of review:	DD/MM/YY	☐☐☐☐☐☐
4. Date of birth	DD/MM/YY	☐☐☐☐☐☐
5. Record complete		☐Yes…1 ☐ No…0 ☐ Other …9

SECTION 1: BACKGROUND INFORMATION

For official use only			
☐	101.	Gender	☐ Male…0 ☐ Female…1
☐	102.	Age at time of first consultation (indicate 0 if at birth)	_____
☐	103.	Population group	☐ Black…1 ☐ Coloured…2 ☐ Indian…3 ☐ White…4 ☐ Other…9 (specify)……
☐	104.	Foreign National	☐ No…0 ☐ Yes…1
☐	105.	Province of residence at birth	☐ EC…1 ☐ Free State…2 ☐ Gauteng…3 ☐ KZN…4 ☐ Limpopo…5 ☐ Mpumalanga…..6 ☐ North West…..7 ☐ Northern Cape …..8 ☐ Western Cape…..9 ☐ Other ……..10

SECTION 2: CLINICAL INFORMATION

For official use only			
☐	201.	Type of defect	☐ Cleft lip…1 ☐ Cleft palate…2 ☐ Cleft lip & palate…3 ☐ Other…9, please specify……..
☐	202.	Description of cleft	☐ Unilateral…1 ☐ Bilateral…2 ☐ Palate…3 ☐ Midline…4 ☐ Other…9, please specify ……..
☐	203.	Cleft laterality	☐ Left…1 ☐ Right…2 ☐ Left & Right…3 ☐ Palate…4 ☐ Midline…5 ☐ Other…9, please specify ……..
☐	204.	Position of the cleft	☐ Lip…1 ☐ Alveolar…2 ☐ Palate…3 ☐ CLEFT LIP AND PALATE …4 ☐ Lip & alveolar…5 ☐ Other…9, (please specify)…….

SECTION 3: TREATMENT/ CARE

For official use only			
☐	301.	Information on treatment on record	☐ No…0 ☐ Yes…1 ☐ Incomplete…2
☐	302.	Number of consultations in previous 12 months	_____
☐	303.	Internal referrals described	☐ No…0 ☐ Yes…1 ☐ Incomplete…2
	304.	Stage of treatment	☐ Plastic surgery…1 ☐ Maxillo-facial surgery…2 ☐ Orthodontics…3 ☐ Speech therapy…4 ☐ Psychology…5 ☐ Genetics…6 ☐ General Dentistry…7 ☐ ENT…8 ☐ Other…9

END OF FORM

Appendix 5 Current Approaches to Care Questionnaire

Comparative Analysis of Healthcare Provision to Individuals with Cleft Lip and Palate at the Specialized Academic Centres in South Africa

For official use only

1. Participant number ☐☐☐☐

2. Study site
   - ☐ 1
   - ☐ 2
   - ☐ 3
   - ☐ 4
   - ☐ 5
   - ☐ 6
   - ☐ 7
   - ☐ 8
   - ☐ 9
   - ☐ 10
   - ☐ 11

   ☐☐☐☐☐☐

3. Date of interview DD/MM/YY ☐☐☐☐☐☐

STATEMENT OF CONSENT

I have been given an information sheet and I understand the objectives of the study.

I further understand that my

responses will be kept confidential and that it is up me whether or not to complete this questionnaire.

It has been explained

to me that even if I choose not to complete this questionnaire, I should still return the questionnaire to the researchers and

indicate **No** in the space below.

My refusal to participate will in no way prejudice me**.**

I agree voluntarily to complete the questionnaire **(please tick).** ☐ **Yes**

☐ **No**

IF YOU AGREE TO PARTICIPATE, PLEASE ANSWER ALL OF THE FOLLOWING QUESTIONS. TICK/ MARK OR CIRCLE THE BOX NEXT TO THE APPROPRIATE ANSWER.

SECTION 1: CARE AND MANAGEMENT

For official use only

	101.	Which point of care do you provide for individuals with CLP	☐ Plastic surgery…1 ☐ Paediatric surgery…2 ☐ Dentistry…3 ☐ Maxillo-facial & oral surgery…4 ☐ Orthodontics…5 ☐ Speech therapy…6 ☐ Other…9, please specify ……
	102.	Which of the following services do you provide at the centre (Tick all that apply)?	
		a) Feeding plate	☐ No...0 ☐ Yes...1
		b) Speech Therapy	☐ No...0 ☐ Yes...1
		c) Genetic counselling	☐ No...0 ☐ Yes...1
		d) Lip and palate repair	☐ No...0 ☐ Yes...1
		e) General dental treatment	☐ No...0 ☐ Yes...1

☐		**f)** Orthodontic treatment	☐ No...0 ☐ Yes...1
☐		g) Orthognathic treatment	☐ No...0 ☐ Yes...1
		h) Other (please specify):	
For official use only			
☐	103.	Is there a specific treatment protocol for CLP?	☐ No…0 ☐ Yes…1
☐	104.	If yes, please could we have a copy of the protocol?	☐ No…0 ☐ Yes…1
☐	105.	Is there a protocol for internal referrals?	☐ No…0 ☐ Yes…1
☐	106.	If yes, please could we have a copy of the protocol?	☐ No…0 ☐ Yes…1
☐	107.	How would you describe care provision at the Centre?	☐ individual specialist…1 ☐ Team approach…2 ☐ Hybrid approach…3 ☐ Other…9 (please specify)…… …… …… ……

SECTION 2: TEAM MEMBERS

For official use only			
☐	201.	Which of the following groups are members of the team at the Centre? (Tick all that apply)?	
☐		a) Geneticist/Genetic Counsellor	☐ No...0 ☐ Yes...1
☐		b) Plastic Surgeon	☐ No...0 ☐ Yes...1
☐		c) Orthodontist	☐ No...0 ☐ Yes...1
☐		d) ENT Surgeon	☐ No...0 ☐ Yes...1
☐		e) Maxillo-Facial Surgeon	☐ No...0 ☐ Yes...1
☐		**f)** Paediatric Dentist	☐ No...0 ☐ Yes...1
☐		g) Paediatric surgeon	☐ No...0 ☐ Yes...1
☐		h) Psychologist	☐ No...0 ☐ Yes...1
☐		i) Professional nurse	☐ No...0 ☐ Yes...1

For official use only			
☐		j) Other, please specify …… …… …… ……..	☐ No...0 ☐ Yes...1
☐	202.	Do the members of the team meet to discuss CLP care?	☐ No...0 ☐ Yes...1
☐	203.	How often do you meet?	☐ Weekly…1 ☐ Monthly…2 ☐ quarterly…3 ☐ Other…9 (please specify)…….
☐	204.	Is there a team-leader?	☐ No...0 ☐ Yes...1
☐	205.	When was the team established?	☐ less than one year ago…1 ☐ 1-2 years ago…2 ☐ 3-5 ears ago …3 ☐ more than 5 years ago…4 ☐ Other ….9 (specify)……..
☐	206.	Do the team members participate in continuous professional development courses?	☐ No...0 ☐ Yes...1

Thank you for participating

Appendix 6 CLP Team IPC Questionnaire

Inter-professional Collaboration among Cleft Lip and Palate Healthcare Team Members in South Africa

FOR OFFICIAL USE ONLY

1. Participant number ☐☐☐☐

2. Study site
   - ☐ 1
   - ☐ 2
   - ☐ 3
   - ☐ 4
   - ☐ 5
   - ☐ 6
   - ☐ 7
   - ☐ 8
   - ☐ 9
   - ☐ 10
   - ☐ 11

   ☐☐☐☐☐☐

3. Date of interview DD/MM/YY ☐☐☐☐☐☐

STATEMENT OF CONSENT

I have been given an information sheet and I understand the objectives of the study.

I further understand that my responses will be kept confidential and that it is up me whether or not to complete this questionnaire.

It has been explained to me that even if I choose not to complete this questionnaire, I should still return the questionnaire to the researchers and indicate **No** in the space below.

My refusal to participate will in no way prejudice me**.**

I agree voluntarily to complete the questionnaire **(please tick).** ☐ **Yes**

☐ **No**

SECTION 1 – BACKGROUND INFORMATION

OFFICE USE ONLY

□ 101. What is your gender?
□Male (0)
□Female (1)

□ 102. What is your age in years?…….

□ 103. Which of the following groups is your profession?
□Geneticist/Genetic Counsellor(1)
□Plastic Surgeon(2)
□Orthodontist(3)
□ENT Surgeon(4)
□Maxillo-Facial Surgeon(5)
□Paediatric Dentist(6)
□Paediatric surgeon(7)
□Speech therapist(8)
□Other, please specify(9)……

□ 104. Do you hold any formal qualification on CLP care?
□ No… 0
□ Yes… 1

□ 105. Do you participate on continuous professional development courses?
□ No… 0
□ Yes… 1

SECTION 2 – CARE EXPERTISE

	Inter-disciplinary care requires collaboration between health –care professionals and patients and their families and circle of care in order to identify and take advantage of each person's care expertise. To support inter-disciplinary practice, practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does'nt apply (0)
201.	Support the participation of patients/clients, their families, and / or community representatives as integral partners alongside health-care personnel					
202.	Share information with patients/clients (or family and the community) in a respectful manner and in such a way that it is understandable, encourages discussion and enhances participation in decision making					
203.	Ensure that appropriate education and support is provided to patients/clients, family members and others involved with care or service					
204.	Listen respectively to the expressed needs of all parties in shaping and delivering care or services					
205.	Conduct a collaborative inter-professional assessment to identify what expertise is required and then individualize for each patient/client					
206.	Coordinated effort to find the best expert for the patient/client					
207.	Patients/clients are full participants in their own care					
208.	Include specific contributions and collective knowledge as dictated by the complexity of the patients/clients needs					

SECTION 3 – SHARED POWER						
	Willingness to share power is a commitment to create balanced relationship through democratic practices of leadership, decision making, authority and responsibility. To support inter-personal practice, practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
301.	Leverage opportunities for all team members to contribute					
302.	Create balanced power relationships					
303.	Establish a safe environment to express diverse opinions					
304.	Consider points of view of all care providers					
SECTION 4 – COLLABORATIVE LEADERSHIP						
	Collaborative leadership (also called reciprocal or shared leadership) is a people – and relationship – focused approach based on the premise that answers should be found in the collective (the team). To support inter-disciplinary practice, practitioners collaboratively determine who will provide group leadership in any given situation by supporting:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
401.	Work with others to enable effective patients/client outcomes					
402.	Advance interdependent working relationships among all participants					
403.	Facilitation of effective team processes					
404.	Establish a climate for collaborative practice among all participants					
405.	Co-create a climate for shared leadership and collaborative practice					
406.	Apply collaborative decision making principles					
407.	Integrate the principles of continuous quality improvement to work processes and outcomes					
408.	Share accountability that addresses power and hierarchy					
409.	Utilize structures and processes to advance exemplary care					
410.	Work with others to enable effective patients/client outcomes					
SECTION 5 – SHARED DECISION MAKING						
	Shared decision-making gives all team members, including patients, the opportunity to contribute their knowledge and expertise, to arrive collaboratively at an optimal goal. To support inter-disciplinary practice, /practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
501.	Recognize and respect each other's knowledge and expertise, regardless of occupation and formal position					
502.	Willing to accept responsibility for decisions					

SECTION 6 – OPTIMIZING PROFESSION, ROLE AND SCOPE						
	Exemplary inter-disciplinary care let all team members work to their full scope of practice and takes advantage of the synergies professionals working together can create. To support inter-disciplinary practice, practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
601.	Describe their role and others'					
602.	Recognize and respect the diversity of other health and social care roles, responsibilities, and competencies					
603.	Perform their own roles in a culturally respectful way					
604.	Communicate roles, knowledge, skills, and attitudes using appropriate language					
605.	Consider the roles of others in determining own professional roles					
606.	Access others' skills and knowledge appropriately through consultation					
607.	Consider the roles of other in determining own professional and inter-professional roles					
608.	Integrate competencies/roles seamlessly into models of service delivery					
609.	Demonstrate knowledge application of own profession/roles/scope					
610.	Explore and integrate roles of others					

SECTION 7 – EFFECTIVE GROUP FUNCTION						
	A health-care system that supports effective teamwork can improve the quality of patient care, enhance patient safety, and reduce workload issues that cause burnout among professionals. To support inter-disciplinary practice, practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
701.	Understand the process of team development					
702.	Develop a set of principles for working together that respects the ethical values of members					
703.	Effectively facilitate discussions and interactions among team members					
704.	Participate, and be respectful of all members' participating , in collaborative decision making					
705.	Regularly reflect on their functioning with team learners/practitioners and patients/clients families					
706.	Establish and maintains effective and healthy working relationships with learners/practitioners, patients/clients, and families, whether or not a formalized team exists					
707.	Respect team ethics, including confidentiality, resource and allocation, and professionalism					
708.	Collaborate and engage together to formulate implement and evaluate care					
709.	Assess, Practise and reflect upon effective group processes					

SECTION 8 - COMPETENT COMMUNICATION						
	Competent communication – openness, honesty, respect for each other's opinions and effective communication skills – is a part of all domains of inter-disciplinary practice. To support inter-disciplinary, practitioners are able to:	Never (1)	Rarely (2)	Sometimes(3)	Always(4)	Does not apply (0)
801.	Establish team work communication principles					
802.	Actively listen to other team members including patients/ clients/families					
803.	Communicate to ensure common understanding of care decisions					
804.	Develop trusting relationships with patients/clients/families and team members					
805.	Effectively use information and communication technology to improve inter-professional patient/client/community-centered care					
806.	Is clear, focused, transparent and respectful					
807.	Constructively manages conflict					
808.	Maintains and enhances the relationship					

Thank you for participating

Appendix 7 Caregivers' Questionnaire

Caregivers' Perceptions of Healthcare Provision and Support for Children Born with Cleft Lip and Palate

FOR OFFICIAL USE ONLY

1. Participant number ☐☐☐☐

2. Study site
   - ☐ 1
   - ☐ 2
   - ☐ 3
   - ☐ 4
   - ☐ 5
   - ☐ 6
   - ☐ 7
   - ☐ 8
   - ☐ 9
   - ☐ 10
   - ☐ 11

   ☐☐☐☐☐☐

3. Date of interview DD/MM/YY ☐☐☐☐☐☐

STATEMENT OF CONSENT

I have been given an information sheet and I understand the objectives of the study.

I further understand that my responses will be kept confidential and that it is up me whether or not to participate in an interview.

It has been explained to me that even if I choose not to participate, I should indicate **No** in the space below.

My refusal to participate will in no way prejudice me**.**

I agree voluntarily to participate in the interview **(please tick).** ☐ **Yes**

☐ **No**

IF YOU AGREE TO PARTICIPATE, PLEASE ANSWER ALL OF THE FOLLOWING QUESTIONS. TICK/ MARK OR CIRCLE THE BOX NEXT TO THE APPROPRIATE ANSWER.

SECTION 1 – BACKGROUND INFORMATION

OFFICE USE ONLY

□ 101. What is your relationship with the CLP child?
□Biological parent (1)
□Foster parent (2)
□Relative (3)
□Guardian (4)
□Care-giver (5)
□Other (9)specify……

□ 102. What is your age in years? …….

□ 103. Which of the parent are you?
□ Mother(1)
□ Father (2)
□ Other (9) specify

□ 104. Race
Black……..(1)
Coloured…(2)
Indian……(3)
White…..(4)
Other …..(9)

□ 105. What is your employment status?
□ Employed.(1)
□ Unemployed (2)
□ Other (9) specify……

□ 106. What sex is your CLP child?
□Male (0)
□Female (1)

□ 107. How old is your CLP child?.....Years.....Months

□ 108. What type of cleft does your child have?……

□ 109. How many children do you have including this one?.....

□ 110. What is your marital status?
□Single (1)
□Married (2)
□Other (9)

□ 111. What is your highest education status?
□None (0)
□Matric (1)
□Tertiary education
□Other (9) specify……..

SECTION 2 – FAMILY IMPACT

OFFICE USE ONLY

□ 201. How has your work or the other parent been affected by your CLP child?

□ 202. Has your child required more attention from you or the other parent?

□ 203. How have the other family members responded on your CLP child?

□ 204. Have you or the other parent been upset because of the CLP child?

□ 205. Do you or the other parent have any feelings of guilt with the birth of this child?

□ 206. What are your most concerned issues on the child's future?

□ 207. Are you comfortable with your child in public places?

□ 208. How does your partner or other children coping with this child?

□ 209. Has your child's condition caused disagreement or conflict in the family?

□ 210. Do you blame yourself or the other parent for your child condition?

□ 211. Has your child's condition caused financial difficulties for your family?

SECTION 3 – SUPPORT SERVICES

OFFICE USE ONLY

□ 301. Were you informed before birth about your CLP child condition?

□ 302. Did you receive counseling regarding your child CLP condition and how was it done?

□ 303. Were the causes of CL P condition explained to you and did you understand?

□ 304. Was termination of pregnancy an option for you due to the condition of your child? or Would you have considered to terminate pregnancy should you have known during pregnancy?

□ 305. How was your CLP child treated at birth?

□ 306. How can you describe the information you received at the birth of your CLP child?

□ 307. What information did you receive regarding your CLP child treatment?

□ 308. Do you feel that having a CLP child affected your way of life?

□ 309. How has your CLP child affected your relationship with your friends?

□ 310. How has your CLP child affected your family relationship?

□ 311. How were your affected by CLP child's condition at birth?

□ 312. Were you satisfied with the information given at the birth of your child?

□ 313. Do you think your child has received necessary treatment from birth till now?

□ 314. Were you satisfied with the support given by the medical team?

□ 315. Was the treatment for your CLP child provided at an easily accessible hospital?

□ 316. Which of the following medical team members played a role in your child's treatment?

k)	Geneticist/Genetic Counsellor	☐ No...0 ☐ Yes...1
l)	Plastic Surgeon	☐ No...0 ☐ Yes...1
m)	Orthodontist	☐ No...0 ☐ Yes...1
n)	ENT Surgeon	☐ No...0 ☐ Yes...1
o)	Maxillo-Facial Surgeon	☐ No...0 ☐ Yes...1
p)	Paediatric Dentist	☐ No...0 ☐ Yes...1
q)	Paediatric surgeon	☐ No...0 ☐ Yes...1
r)	Speech Therapist	☐ No...0 ☐ Yes...1
s)	Professional nurse	☐ No...0 ☐ Yes...1

	t) Other, please specify…… …… …… ……..	☐ No...0 ☐ Yes...1

□ 317. Do you have any recommendations on the care of CLP children?

_____
_____
_____
_____
_____
_____
_

Thank you for participating

Appendix 8 Provincial Health Departments Approvals

Eastern Cape Department of Health

Enquiries:	Madoda Xokwe	Tel No:	040 608 0830
Date:	08 April 2016	Fax No:	043 642 1409
e-mail address:	zonwabele.merile@echealth.gov.za		

Prof P Hlongwa

RE: Epidemiology and care of individuals with cleft lip and palate in South Africa. (EC_2016RP15_718)

The Department of Health would like to inform you that your application for conducting a research on the abovementioned topic has been approved based on the following conditions:

1. During your study, you will follow the submitted protocol with ethical approval and can only deviate from it after having a written approval from the Department of Health in writing.
2. You are advised to ensure, observe and respect the rights and culture of your research participants and maintain confidentiality of their identities and shall remove or not collect any information which can be used to link the participants.
3. The Department of Health expects you to provide a progress on your study every 3 months (from date you received this letter) in writing.
4. At the end of your study, you will be expected to send a full written report with your findings and implementable recommendations to the Epidemiological Research & Surveillance Management. You may be invited to the department to come and present your research findings with your implementable recommendations.
5. Your results on the Eastern Cape will not be presented anywhere unless you have shared them with the Department of Health as indicated above.

Your compliance in this regard will be highly appreciated.

SECRETARIAT: EASTERN CAPE HEALTH RESEARCH COMMITTEE

Ilanga elilgqambileyo!

health
Department:
Health
PROVINCE OF KWAZULU-NATAL

DIRECTORATE:
Health Research & Knowledge Management (HKRM)

330 Langalibalele street,
Private Bag X9051 PMB, 3200
Tel: 033 395 2805/3189/3123 Fax: 033 394 3782
Email: hrkm@kznhealth.gov.za
www.kznhealth.gov.za

Reference: **HRKM80/16**
KZ_2016RP4_852

15 April 2016

Dear Prof P Hlongwa
(Wits University)

Subject: Approval of a Research Proposal

1. The research proposal titled **'Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa'** was reviewed by the KwaZulu-Natal Department of Health (KZN-DoH).

   The proposal is hereby **approved** for research to be undertaken at King Edward VIII Hospital.

2. You are requested to take note of the following:
   a. Obtain support letters from the relevant district managers and make the necessary arrangement with the identified facility before commencing with your research project.
   b. Provide an interim progress report and final report (electronic and hard copies) when your research is complete.

3. Your final report must be posted to **HEALTH RESEARCH AND KNOWLEDGE MANAGEMENT, 10-102, PRIVATE BAG X9051, PIETERMARITZBURG, 3200** and e-mail an electronic copy to hrkm@kznhealth.gov.za

For any additional information please contact Ms G Khumalo on 033-395 3189.

Yours Sincerely

Dr E Lutge
Chairperson, Health Research Committee
Date: 19/04/15

Fighting Disease, Fighting Poverty, Giving Hope

LIMPOPO
PROVINCIAL GOVERNMENT
REPUBLIC OF SOUTH AFRICA

DEPARTMENT OF HEALTH

Enquiries: Latif Shamila

Ref:4/2/2

Hlongwa P
WITS
HUMAN RESEARCH ETHICS COMMITTEE

Greetings,

RE: Epidemiology and Care of individuals with Cleft Lip and Palate in South Africa

The above matter refers.

1. Permission to conduct the above mentioned study is hereby granted.
2. Kindly be informed that:-
    - Research must be loaded on the NHRD site (http://nhrd.hst.org.za) by the researcher.
    - Further arrangement should be made with the targeted institutions, after consultation with the District Executive Manager.
    - In the course of your study there should be no action that disrupts the services.
    - After completion of the study, it is mandatory that the findings should be submitted to the Department to serve as a resource.
    - The researcher should be prepared to assist in the interpretation and implementation of the study recommendation where possible.
    - The above approval is valid for a 3 year period.
    - If the proposal has been amended, a new approval should be sought from the Department of Health.
    - Kindly note, that the Department can withdraw the approval at any time.

Your cooperation will be highly appreciated.

Head of Department

30/05/2016
Date

18 College Street, Polokwane, 0700, Private Bag x9302, POLOLKWANE, 0700
Tel: (015) 293 6000, Fax: (015) 293 6211/20 Website: http/www.limpopo.gov.za

Appendix 9 Academic Centres Approvals

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

CHARLOTTE MAXEKE JOHANNESBURG ACADEMIC HOSPITAL

Enquiries:
Mr. J. Maepa
Office of the Clinical Director
Tell: (011): 488-3365
Email:Johannes.maepa@gauteng.gov.za
30 March 2016

Dear Prof Phumzile Hlongwa

STUDY TITLE: Epidemiology and Care of Individuals with Cleft Lip and Plates in South Africa.

Permission is granted for you to conduct the above recruitment activities as described in your request provided:

1. Charlotte Maxeke Johannesburg Academic Hospital will not anyway incur or inherit costs as result of the said study.
2. Your study shall not disrupt services at the study sites.
3. Strict confidentiality shall be observed at all times.
4. Informed consent shall be solicited from patients participating in your study.

Please liaise with the HOD and Unit Manager or sister in charge to agree on the dates and time that would suit all parties.

Kindly forward this office with the results of your study on completion of the research.

Supported/~~not supported~~

Dr. M.I. Mofokeng
Clinical Director
DATE: 14/4/2016

Approved/~~not approved~~

Ms. G. Bogoshi
Chief Executive Officer
DATE: 14/4/2016

health
Department:
Health
PROVINCE OF KWAZULU-NATAL

DIRECTORATE:

Physical Address: 800 Bellair Road, Mayville, 4058
Postal Address: Private Bag X08, Mayville, 4058
Tel: 0312401059 Fax: 0312401050 Email: ursulanun@ ialch.co.za
www.kznhealth.gov.za

Office of The Medical Manager
IALCH

12 September 2016

Prof P Hlongwa
Wits University

Dear Prof Hlongwa

Re: Approved Research: Ref No: HRKM80/16: Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa.

As per the policy of the Provincial Health Research Committee (PHRC), you are hereby granted permission to conduct the above mentioned research once all relevant documentation has been submitted to PHRC inclusive of Full Ethical Approval.

Kindly note the following.

1. The research should adhere to all policies, procedures, protocols and guidelines of the KwaZulu-Natal Department of Health.
2. Research will only commence once the PHRC has granted approval to the researcher.
3. The researcher must ensure that the Medical Manager is informed before the commencement of the research by means of the approval letter by the chairperson of the PHRC.
4. The Medical Manager expects to be provided feedback on the findings of the research.
5. Kindly submit your research to:

   The Secretariat
   Health Research & Knowledge Management
   330 Langaliballe Street, Pietermaritzburg, 3200
   Private Bag X9501, Pietermaritzburg, 3201
   Tel: 033395-3123, Fax 033394-3782
   Email: hrkm@kznhealth.gov.za

Yours faithfully

.....

Dr L P Mtshali
Medical Manager

health
Department:
Health
PROVINCE OF KWAZULU-NATAL

DIRECTORATE:
Office of The Medical Manager
IALCH

Physical Address: 800 Bellair Road, Mayville, 4058
Postal Address: Private Bag X08, Mayville, 4058
Tel: 0312401059 Fax: 0312401050 Email: ursulanun@ ialch.co.za
www.kznhealth.gov.za

Reference: HRKM80/16
Enquiries: Medical Management

12 September 2016

Prof P Hlongwa
Wits University

Dear Prof Hlongwa

RE: PERMISSION TO CONDUCT RESEARCH AT IALCH

I have pleasure in informing you that permission has been granted to you by the Medical Manager to conduct research on: **Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa.**

Kindly take note of the following information before you continue:

1. Please ensure that you adhere to all the policies, procedures, protocols and guidelines of the Department of Health with regards to this research.
2. This research will only commence once this office has received confirmation from the Provincial Health Research Committee in the KZN Department of Health.
3. Kindly ensure that this office is informed before you commence your research.
4. The hospital will not provide any resources for this research.
5. You will be expected to provide feedback once your research is complete to the Medical Manager.

Yours faithfully

.....

Dr L P Mtshali
Medical Manager

Province of the
EASTERN CAPE
HEALTH

Research Ethics Committee • Level 3 • Nelson Mandela Academic Hospital • Sissons Street -Fortgale •
Private Bag X 5152 • MTHATHA • Eastern Cape • 5100 • REPUBLIC OF SOUTH AFRICA •
Tel.: +27 (0)47 502 4546 • Fax: +27 (0)47 502 4968

26 September 2016

Prof Phumzile Hlongwa
School of Public Health
University of the Witwatersrand
Johannesburg

Research Ethics Approval

Project Title:

Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa.

The above-mentioned study has been approved by the Research Ethics Committee.

Prof N.T Tonjeni
Chair: Research Ethics Committee
Nelson Mandela Central Hospital
Mthatha

Western Cape Government
Health

Dr AS Booysen
Manager: Medical Services
Email: Tony.Booysen@Westerncape.gov.za
Tel: +27 21 658 5788 fax: +27 21 658 5166

Prof P Hlongwa
Red Cross War Memorial Children's Hospital

Dear Prof P Hlongwa

APPROVAL OF RESEARCH

PROJECT TITLE: EPIDEMIOLOGY AND CARE OF INDIVIDUALS WITH CLEFT LIP AND PALATE IN SOUTH AFRICA

It is a pleasure to inform you that approval is hereby granted to conduct the above-mentioned study at Red Cross War Memorial Children's Hospital.

Yours sincerely,

Dr AS Booysen
Manager: Medical Services
Date: 17.05.16

Permission to access Records / Files / Data base at Steve Biko Academic Hospital

TO: Dr Ernest Kenoshi [Name]
Chief Executive Officer/Information Officer

Steve Biko Academic Hospital

FROM : Prof Phumzile Hlongwa [Name]
Investigator

Wits Oral Health Centre Hospital

Re: Permission to do research at Steve Biko Academic **Hospital**

TITLE OF STUDY: Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa

This request is lodged with you in terms of the requirements of the Promotion of Access to Information Act. No. 2 of 2000.

I am a researcher / PhD student at the School of Public Health at the University of the Witwatersrand /

Wits Oral Health Centre Hospital. I am working with Prof Rispel and Prof Dandajena as my supervisors.
I herewith request permission on behalf of all of us to conduct a study on the above topic on the hospital / clinic grounds. This study involves access to patient records. This study involves clinical research.

The researchers request access to the following information: clinical files, record books and data bases.

We intend to publish the findings of the study in a professional journal and/ or to present them at professional meetings like symposia, congresses, or other meetings of such a nature.

We intend to protect the personal identity of the patients by assigning each individual a random code number.

We undertake not to proceed with the study until we have received approval from the Faculty of Health Sciences Research Ethics Committee, University of Pretoria.

Yours sincerely

P Hlongwa

Signature of the Principal Investigator

Permission to do the research study at this hospital / clinic and to access the information as requested, is hereby approved.

Title and name of Chief Executive Officer: M E KENOSHI

Name of hospital / clinic: STEVE BIKO ACADEMIC HOSPITAL

Signature: _____ Date: 7/7/2016

DR ME KENOSHI
CHIEF EXECUTIVE OFFICER
STEVE BIKO
ACADEMIC HOSPITAL

Official Stamp

The Research Ethics Committee, Faculty Health Sciences, University of Pretoria complies with ICH-GCP guidelines and has US Federal wide Assurance.
- FWA 00002567, Approved dd 22 May 2002 and Expires 28 August 2018.
- IRB 0000 2235 IORG0001762 Approved dd 22/04/2014 and Expires 22/04/2017.

UNIVERSITEIT VAN PRETORIA
UNIVERSITY OF PRETORIA
YUNIBESITHI YA PRETORIA

Faculty of Health Sciences Research Ethics Committee

21/09/2016

Approval Certificate
New Application

Ethics Reference No.: 278/2016

Title: Epidemiology and Care of Individuals with Cleft Lip and Palate in South Africa

Dear Prof Phumzile P Hlongwa

The **New Application** as supported by documents specified in your cover letter dated 23/06/2016 for your research received on the 23/06/2016, was approved by the Faculty of Health Sciences Research Ethics Committee on its quorate meeting of 21/09/2016.

Please note the following about your ethics approval:
- Ethics Approval is valid for 2 years
- Please remember to use your protocol number (**278/2016**) on any documents or correspondence with the Research Ethics Committee regarding your research.
- Please note that the Research Ethics Committee may ask further questions, seek additional information, require further modification, or monitor the conduct of your research.

Ethics approval is subject to the following:
- The ethics approval is conditional on the receipt of **6 monthly written Progress Reports**, and
- The ethics approval is conditional on the research being conducted as stipulated by the details of all documents submitted to the Committee. In the event that a further need arises to change who the investigators are, the methods or any other aspect, such changes must be submitted as an Amendment for approval by the Committee.

We wish you the best with your research.

Yours sincerely

Dr R Sommers; MBChB; MMed (Int); MPharMed,PhD
Deputy Chairperson of the Faculty of Health Sciences Research Ethics Committee, University of Pretoria

The Faculty of Health Sciences Research Ethics Committee complies with the SA National Act 61 of 2003 as it pertains to health research and the United States Code of Federal Regulations Title 45 and 46. This committee abides by the ethical norms and principles for research, established by the Declaration of Helsinki, the South African Medical Research Council Guidelines as well as the Guidelines for Ethical Research: Principles Structures and Processes, Second Edition 2015 (Department of Health).

☎ 012 356 3084 deepeka.behari@up.ac.za http://www.up.ac.za/healthethics
✉ Private Bag X323, Arcadia, 0007 - Tswelopele Building, Level 4, Room 60, Gezina, Pretoria

Western Cape Government
Health

TYGERBERG HOSPITAL
REFERENCE: **Research Projects**
ENQUIRIES: **Dr GG Marinus**
TELEPHONE:**021 938 5752**

Ethics Reference: M150536

TITLE: **Epidemiology and care of individuals with cleft lip and palate in South Africa.**

Dear Prof Phumzile Hlongwa

PERMISION TO CONDUCT YOUR RESEARCH AT TYGERBERG HOSPITAL.

1. In accordance with the Provincial Research Policy and Tygerberg Hospital Notice No 40/2009, permission is hereby granted for you to conduct the above-mentioned research here at Tygerberg Hospital.

2. Researchers, in accessing Provincial health facilities, are expressing consent to provide the Department with an electronic copy of the final feedback within six months of completion of research. This can be submitted to the Provincial Research Co-Ordinator (Health.Research@westerncape.gov.za).

DR GG MARINUS
MANAGER: MEDICAL SERVICES [RESEARCH CO-ORDINATOR]

DR D ERASMUS
CHIEF EXECUTIVE OFFICE
Date: 15 June 2016

Administration Building, Francie van Zijl Avenue, Parow, 7500 Private Bag X3, Tygerberg, 7505
tel: +27 21 938-6267 fax: +27 21 938-4890 www.capegateway.go.v.za

health
Department of Health
FREE STATE PROVINCE

8 July 2016

Prof P Hlongwa
Department of Orthodontics & Paediatric Dentistry
School of Oral Health Sciences
5 York Road
Parktown
JOHANNESBURG

Dear Prof Hlongwa

EPIDEMIOLOGY AND CARE OF INDIVIDUALS WITH CLEFT LIP AND PALATE IN SOUTH AFRICA

Universitas Academic Hospital will only grant permission when the following criteria have been met:

√ that you obtain Ethical Clearance from the Human Research Ethics Committee of the relevant University;

√ that the Hospital incurs no cost in the course of your research;

√ that access to the staff and patients at Universitas Academic Hospital will not interrupt the daily provision of services; and

√ that prior to conducting the research you will liaise with the supervisors of the relevant sections and introduce yourself with permission letter and to make arrangements with them in a manner that is convenient to the sections.

Dr. B.A. Benganga
11 JUL 2016
Acting Head of Clinical Services
Universitas Academic Hospital

Yours sincerely

DR B A BENGANGA
ACTING HEAD: CLINICAL SERVICES

UNIVERSITAS ACADEMIC HOSPITAL
Private Bag X20660, Bloemfontein, 9300
Clinical Services, First Floor, Logeman Street, Universitas, Bloemfontein, 9301
Tel: (051) 405 3557 Fax: (051) 405 3634
E-mail address: bengangaba@universitas.fs.gov.za

Sefako Makgatho Health Sciences University
Research & Postgraduate Studies Directorate
Sefako Makgatho University Research Ethics Committee
(SMUREC)

Molotlegi Street, Ga-Rankuwa 0208
Tel: (012) 521 5617/3698 | fax: (012) 521 3749
Email: lorato.phiri@smu.ac.za
P.O. Box 163 Medunsa 0204

Prof P Hlongwa
University of Wits
Department of Oral Health Sciences

Dear Prof Hlongwa

RE: PROF P HLONGWA – REQUEST FOR PERMISSION TO CONDUCT A STUDY AT THE MEDUNSA ORAL HEALTH CENTRE (CLEFT CLINIC)

The committee **NOTED** an e-mail dated 29 April 2016, responding to SMUREC recommendations.

Study Title:	Epidemiology and care of individuals with Cleft Lip and Palate in South Africa
Principal Investigator:	Prof P Hongwa
Institution:	University of Wits, Department of Oral Health Sciences
Clearance Certificate No.:	M150536
Approval date:	18 November 2015
Type of Research:	PhD in the School of Public Health

SMUREC **APPROVED** and **GRANTED** the researcher permission to conduct the research study at the Medunsa Oral Health Centre (CLEFT Clinic)

Yours Sincerely,

DR C BAKER
DEPUTY CHAIRPERSON SMUREC

04 May 2016

SEFAKO MAKGATHO
HEALTH SCIENCES UNIVERSITY
SMU Research Ethics Committee
Chairperson
Date: 04/05/2016

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

MEDICAL ADVISORY COMMITTEE
CHRIS HANI BARAGWANATH ACADEMIC HOSPITAL

PERMISSION TO CONDUCT RESEARCH

Date: 8 April 201(

TITLE OF PROJECT: Epidemiology and care of individuals with cleft lip and palate in South Africa

UNIVERSITY: Witwatersrand

Principal Investigator: P Hlongwa

Department: Public Health

Supervisor (If relevant): L Rispel

Permission Head Department (where research conducted): not yet

Date of start of proposed study: April 2016
Date of completion of data collection: Dec 2020

The Medical Advisory Committee recommends that the said research be conducted at Chris Hani Baragwanath Hospital. The CEO /management of Chris Hani Baragwanath Hospital is accordingly informed and the study is subject to:-

- Permission having been granted by the Human Research Ethics Committee of the University of the Witwatersrand.
- the Hospital will not incur extra costs as a result of the research being conducted on its patients within the hospital
- the MAC will be informed of any serious adverse events as soon as they occur
- permission is granted for the duration of the Ethics Committee approval.

.....
Recommended
(On behalf of the MAC)
Date: 08 April 2016

.....
Approved/Not Approved
Hospital Management
Date: 12/04/16

Appendix 10 John Wiley & Sons' Permission

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Jul 29, 2018

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Requestor type	University/Academic
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Number of figures/tables	1
Original Wiley figure/table number(s)	Figure 1
Will you be translating?	No
Title of your thesis / dissertation	Epidemiology and care for individuals with cleft lip and palate in South Africa
Expected completion date	Aug 2018
Expected size (number of pages)	200
Requestor Location	Prof. Phumzile Hlongwa P.O. Box 244 Blue Valley Centurion, 0096 South Africa Attn: Prof. Phumzile Hlongwa
Publisher Tax ID	EU826007151
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Appendix 11 Declaration: Student's contribution to article

I, **Phumzile Hlongwa,** student number **1021703,** declare that this Thesis is my own work and that I contributed adequately towards research findings published in the article(s) stated below which are included in my Thesis.

Signature of Student ……

Date…….

Name of Primary Supervisor: Professor Laetitia C Rispel

Signature of Primary Supervisor

……**Date**……..

Agreement by co-authors: By signing this declaration, the co-authors listed below agree to the use of the article(s) by the student as part of his/her Thesis/Dissertation/Research Report. In cases where the student is not the 1st author of a published article, the primary supervisor must explain (under comments) why the student is entitled to use the paper for his/her degree purposes.

Article 1: Title: "People look and ask lots of questions": caregivers' perceptions of healthcare provision and support for children born with cleft lip and palate.

Journal name, year, volume and page numbers: *BMC Public Health*. 2018; **18**(1):506. DOI. 10.1186/s12889-018-5421

Authors	Name	Signature	Date
1st author	Phumzile Hlongwa		
2nd author	Laetitia C Rispel		
3rd author			
4th author			
5th author			
6th author			

Comments by primary supervisor:

……

……

…… ……..

Appendix 12 BMC Public Health Permission

From: "Romel Jake Cruz" <romeljake.cruz@springernature.com>
To: Phumzile Hlongwa
Subject: 00908481 RE: Permission to reproduce published journal article: "People look and ask...
Date: 31 July 2018 01:14:03 PM

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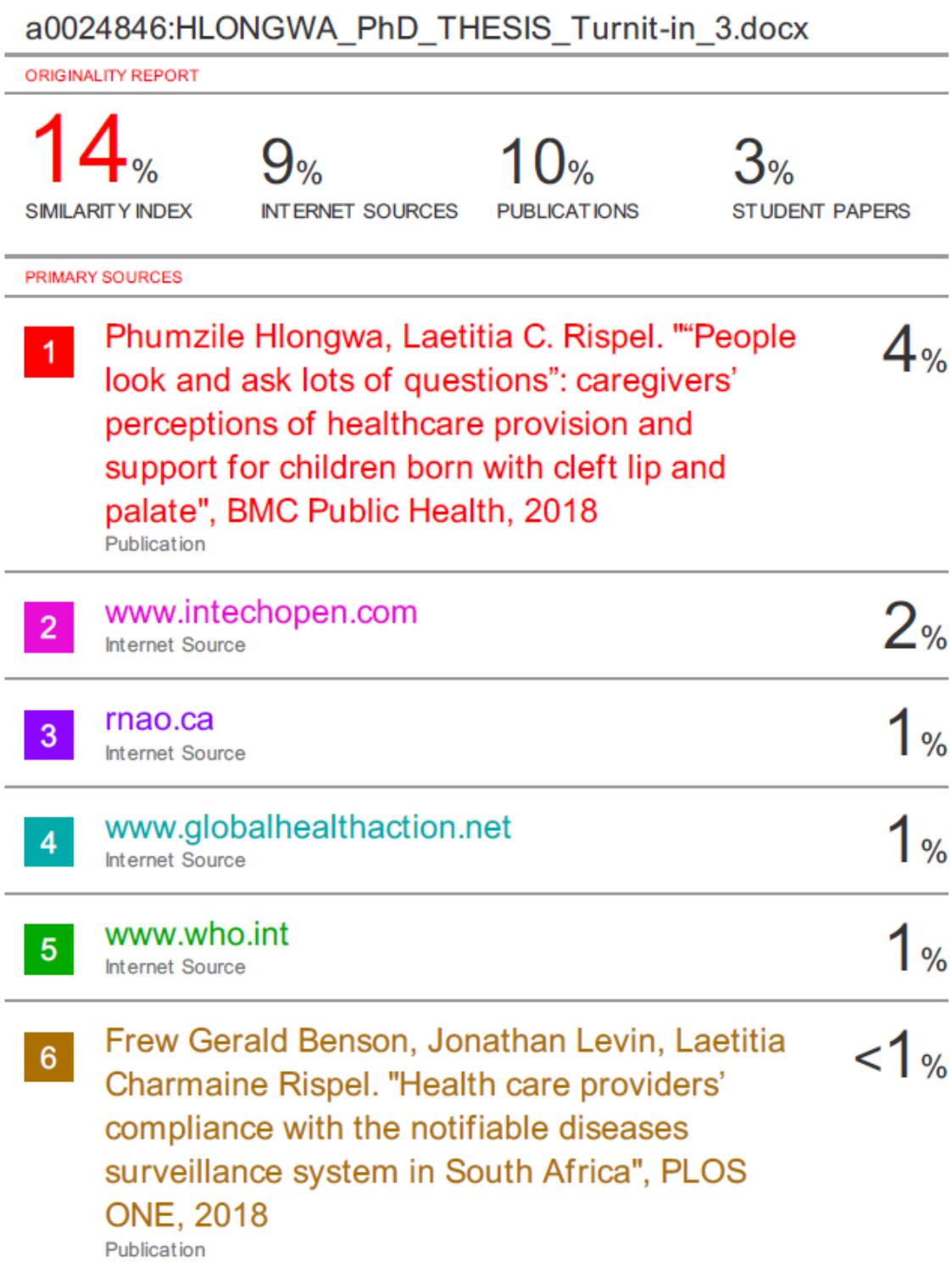
With kind regards,

Romel Jake Cruz

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Appendix 13 PhD Turnit-in Report



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ORIGINALITY REPORT

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SIMILARITY INDEX	INTERNET SOURCES	PUBLICATIONS	STUDENT PAPERS

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