CLEFT LIP AND PALATE CARE IN NIGERIA.

A thesis submitted to The University of Manchester for the degree of Masters of Philosophy in Orthodontics at the Faculty of Medical and Human Sciences

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ABBREVIATIONS

BCLP Bilateral cleft lip and palate

CDG Cleft Development Group

CFC Craniofacial condition

CL(A) Cleft lip and alveolus

CLAPA Cleft Lip and Palate Association

CL/P Cleft lip and / or palate

CPO Cleft palate only

CRANE Craniofacial Network

CSAG Clinical Standards Advisory Group

SLT Speech and language therapist

SN Specialist nurse

UCLP Unilateral complete cleft lip and palate

UICLP Unilateral incomplete cleft lip and palate

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ABSTRACT

BACKGROUND; The World Health Organisation has recommended the need to standardise cleft care globally. In Europe, the Eurocleft project was a concerted effort to improve on the standards of care for children with cleft lip and palate. Certain recommendations were made that were used to judge the standards of care offered, this eventually led to reorganization of services. Improving on standards of cleft care in Nigeria, would require a starting point, by determining what is currently being offered. Measurement of treatment outcome aims at reporting on the quality of care being offered. Up to date, there has been no Nigerian study that has reported on dentofacial outcomes in children affected with cleft lip and palate. This thesis, for the first time reports on dentofacial outcomes by assessing dental relationships in a selected Nigerian cleft population, it also reports broadly on cleft care services available in the country.

METHODS AND DESIGN: The design incorporated two methods of data collection; Semi-structured questionnaires administered to cleft care specialists to extract useful information about cleft care services in Nigeria.; Assessment of post-surgical study models of 18 of 5year old Nigerian children affected with UCLP using two commonly used indices (5 years old and the modified Huddart/Bondeham scales) to determine treatment outcome.

RESULTS: Treatment outcome reported in a selected Nigerian cleft population is good and compares favourably with treatment outcome in a European cleft population. The results are reliable with moderate to good inter-examiner and intra-examiner agreement.
The main form of treatment offered is surgery, which is sponsored mainly by one charity organisation. Shortcomings in services offered include lack of comprehensive care, such as speech therapy and Ear Nose and Throat services, generally cleft care services offered in the country fall below the WHO expected standards.

CONCLUSION: This is a single centre study that has determined treatment outcome of Nigerian children affected with CL/P. Findings are valid and reliable, though limited to a selected population.

RECCOMENDATIONS: Intercentre studies are recommended in future, this will allow for comparison as well as the use of a larger sample size.

DECLARATION

No portion of the work referred to in this thesis has been submitted in support of an application for another degree of this or any other university or other institute of learning.

DEDICATION

This thesis is dedicated to God the father, the Son and the Holy Spirit, from which I draw my inspiration to live, write and have my being.

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ABOUT THE AUTHOR OF THIS THESIS

I am a Dental Surgeon, Academic and Researcher with over 25year's experience. I have about 25 publications in peer-reviewed journals. My area of specialization is in Orthodontics with special interest in cleft lip and palate care.

My interest in cleft lip and palate care started about 10 years ago, when I was sponsored to train as a cleft specialist in a specialist hospital abroad. My experience in this hospital, motivated primarily by my interest led to my quest to contribute to the existing knowledge pertaining to the important field of CL/P, I decided to go in depth, by registering for a research degree (PhD Orthodontics) at the University of Manchester.

Over the last 3 years, I have been training as well as conducting research in the field of cleft at both the University of Manchester and Royal Manchester children Hospital. To date, I have been able to publish some of my findings in peer reviewed journals (see Appendix)

THESIS PRESENTATION

This thesis aimed at presenting broadly cleft care in Nigeria. It has for the first time reported treatment outcome in a Nigerian cleft population.

The thesis have been divided into 5 chapters It comprises of five main chapters, which have been set out as follows;

Chapter 1, is the introductory chapter and starts by describing cleft care in the developing world, it also discusses the condition cleft lip and /or palate (CL/P) and its consequences as well as treatment protocol. This chapter defines the hypothesis, aims and objectives of the study.

Chapter 2, presents the literature review with focus on the need for Evidenced Based Care in the field of CL/P.

Chapter 3, presents the methods and material used for the study.

Chapter 4, presents the results of the study.

Lastly chapter 5, presents the discussion of the study.

The references and appendices are attached .

CHAPTER 1: INTRODUCTION

1.0 Background

Every year, almost a quarter of a million babies are born with cleft lip and/or palate (CL/P) in the poorest part of the world (Mars et al., 2008). In Nigeria alone, there are likely to be between 55,000 and 200,000 children born with oro-facial clefts over the last 15 years (Oginni et al., 2014). The majority of affected children in developing countries receive limited treatment for this condition due to the scarcity of resources. One major identified problem with cleft care in developing countries is the associated stigma, which results in the reluctance of families of affected children to bring them for treatment (Oginni et al., 2014). This problem is further compounded by the unaffordability of care by affected families due to their low socioeconomic status and the fact that the Nigerian government does not offer free treatment to children affected with CL/P.

Comprehensive cleft care is expensive, and quality care requires enormous financial and human resources. Unfortunately, health care in Nigeria is not free, and is borne of an out-of-pocket basis; therefore, families seeking comprehensive cleft care in Nigeria must bear the cost of the treatment. Oro-facial conditions is also not considered a priority in a country, where Malaria and Ebola diseases are popular and has led to death of several thousand lives over years.

The rising prevalence of orofacial clefts in Nigeria over the last decade also calls for concern (Butali et al.,2014). Many of these children are from poor background and majority of their parents are unable to afford the cost of treatment .The Nigerian government on its own part has continually pledged to provide affordable health care to all its citizens by introducing health services schemes that would be affordable (www.ng.gov/2014). Such schemes aim at providing free health care for all children below the age of 5 years, it is hoped that it would be beneficial to Nigerian children born with oro-facial clefts.

Presently, however cleft care service provision in Nigeria is solely funded by one main charity organisation, the Smile Train (www.smiletrain.org). Questions may then arise as to why a country considered the giant of Africa with its enormous material resources would be unable to provide free or affordable health care to its citizens. Such questions remain unanswered, despite that many of the nation's citizens continue to live in abject poverty. However, on the international scene, Nigeria is portrayed as an oil-rich country with one of

the fastest growing economies in the world, having a whopping annual budget of 4.962 trillion Naira (approx. 983 billion dollars) (www.gov.ng/2013).

The unaffordability of health care by the citizens of Nigeria is further compounded by the health sector's chronic underfunding. For example, in 2014, healthcare was allocated just 5.6% of the budget (approximately N262.74 billion), which is below the anticipated 15% agreed previously by the African Union of Health Workers (www.gov.ng).

Presently, the Nigerian health care system precludes cleft care and services are provided mainly by foreign charity organisations. The contrast is seen in the United Kingdom, where cleft care services is sponsored by the government, with establishment of regional cleft care centres staffed with highly skilled specialists that are able to offer comprehensive care (CSAG, 1998).

The present government in Nigeria has promised to assist in financing the health care of its citizens. The first step towards achieving this pledge was the implementation of the National Health Insurance Scheme (NHIS) in 2005; a laudable project that appears to provide a solution to the issue of health care financing in Nigeria (www.nhis.org).

Non -intervention of the Nigerian government in cleft care services may actually be as a result of ignorance about the condition. Perhaps locating cleft care services within the Nigerian health care system may highlight grey areas and inform governmental agencies about the need for cleft care. This would be in agreement with the World Health Organisation report, which highlighted the need to reduce the burden of craniofacial diseases, need for evidence based care, improvement on the quality of cleft care, and improved access and availability of care (WHO, 2002).

There has been a gradual change in cleft care services in Nigeria over the last few years, this has been largely due to the intervention of cleft charity missions, who now offer free surgery to affected children in developing countries. Presently it is unclear, what impact such free surgery may have had on the quality of care received so far , thus creating a need to evaluate treatment outcomes in Nigerian children born with cleft of the lip and/or palate (CL/P).

Measurement of treatment outcome in children with CL/P, can give an accurate and reliable picture of cleft care services rendered, allow for criticism and create room for improvement. In general, treatment outcomes aim at assessing surgical care, speech, aesthetics and dental care received in children with CL/P.

Dento-facial outcomes in particular, aim at assessing post-surgical records of children affected with CL/P and has the advantage of evaluating quality of treatment offered, findings of such assessment thereafter, may lead to modification of surgical timing and technique (Shaw et al., 1992; Semb and Shaw, 2013). Reporting of treatment outcome may also inform health funding agencies by giving reliable evidence that may be helpful in taking health policies decisions (Bearn et al., 2001; Persson et al., 2013; Daskalogiannakis et al., 2010).

Presently in Nigeria, the main type of treatment offered to children with CL/P is surgical care, so it seems reasonable to assess post-surgical outcomes in affected children. The importance of outcome assessment can be seen as changes to cleft care services in the UK over the last two decades, where reports of poor treatment outcome led to reorganization of services into regional centres (Shaw et al., 1992; Sandy et al., 1998; Bearn et al., 2001).To date there has been no known published literature on dento-facial outcome for Nigerian children affected with CL/P.

To achieve the aims and objectives of the study, it was necessary to understand the subject matter, 'Cleft lip and palate'. The thesis continues by giving definition of CL/P, the sub-types of clefts and treatment protocol

1.1 DEFINITION OF CLEFT LIP AND PALATE (CL/P).

CL/P is one of the most common congenital conditions (Vanderas, 1987; Clinical Standards Advisory Group, 1998; Mosey and Little, 2002). In basic terms, cleft of the lip is the presence of one or two vertical fissure in the upper lip and alveolus. It may vary from a small defect to a complete cleft extending up to and through the floor of the nose. While cleft palate is the presence of one vertical fissure in the palate. CL/P is said to occur when the facial processes in an embryo do not completely fuse. The working definition for cleft of the lip and palate has been defined occurring as a cleft lip with or without cleft palate (CL/P) or a cleft palate without lip (CP) (ICBDMS, 2001).

The main cleft sub-types are isolated cleft lip (CL), cleft lip and alveolus (CLA), cleft palate only (CPO), unilateral incomplete cleft lip and palate (IUCLP), unilateral complete cleft lip and palate (UCLP), incomplete bilateral cleft lip and palate (IBCLP) and bilateral complete cleft lip and palate (BCLP).

Normal development of the oral cavity starts about the 4^{th} week of pregnancy, a lack of fusion of the primary palate in the 5th week of pregnancy manifests as cleft of the lip and

alveolus while lack of fusion of the secondary palate during the 8th week of pregnancy results in cleft of the palate (Sperber,2002).

1.2 CAUSES OF CL/P

The causes of CL/P are not well understood but there is a strong genetic link, complicated by the interaction of a range of environmental and lifestyle factors such as diet or smoking (Murray, 2002). The multifactorial concept of cause and effect of CL//P have been confirmed by observing the etiological heterogeneity in a series of cases of CL/P. Some are caused by mutant genes, others as a result of chromosomal abnormalities, specific environmental agents and a good number is caused by an interaction between genes and environmental factors (Mossey, 2009; Botto, 2002; Butali 2011). CL/P is said to be inherited, the inheritance trait has been evidenced by studies on siblings and mongoloid twins (Douglas, 1958;Asling et al., The main cleft sub-types are isolated cleft lip (CL), cleft lip and alveolus (CLA), cleft palate only (CPO), unilateral incomplete cleft lip and palate (IUCLP), unilateral complete cleft lip and palate (UCLP), incomplete bilateral cleft lip and palate (IBCLP) and bilateral complete cleft lip and palate (BCLP).

Association between environmental risk factors and CL/P such as maternal exposure to tobacco, alcohol, poor nutrition, infection, medicinal drugs and teratogens has been investigated, with maternal smoking and lack of folic acid being consistently linked with the risk of developing clefts (Mossey et al., 2007).

1.3 PREVALENCE OF CL/P

Cleft lip and palate occurs more frequently than isolated cleft lip and isolated palate (Calzolari et al., 2007, Tolarova and Cervanka, 1998). Generally CL/P affects males more than females (Niswander et al., 1972; Ritter et al., 2004). Gender distribution of cleft lip and palate reports that isolated cleft lip is more common in males while isolated Cleft palate is more common in females (Rittler et al., 2004).

The global prevalence of CL/P is 1:700 live births (WHO, 2002, 2005). Higher incidence has been reported among Asians with a prevalence as high as 2 per 1000(Gorlin et al.,2001).In the Chinese a prevalence of 1.12 per 1000 has been recorded (Cooper etal,.2000).These figures are slightly higher than a cleft prevalence of 1 per 1000 reported in White British(Department of Health, 1998; CRANE Project Team, 2009).Lower prevalence 0.3-0.9 per 1000 live births has been reported among the black populations (Butali 2009).Though it has been difficult ascertaining true prevalence in the black group due to bias in methodology

and reporting (Mossey et al., 2009). The prevalence of clefts in the mixed ethnic group is not known (Hernandez-Diaz 2000).

The World Health Organization in its series of reports on international collaborative research on craniofacial anomalies reported on the incidence and prevalence of clefts (WHO 2002, 2003). In general, both cleft lip (with or without cleft palate) and isolated cleft palate have an incidence of approximately 1 in 700 live births (WHO, 2002). It was also explained that although differences among countries do exist, birth prevalence of cleft lip (with or without cleft palate) is around 1:1000 births. Further, in two thirds of patients diagnosed with cleft, the left side is more frequently affected, and it is more prevalent in males than in females (WHO, 2002).

Higher incidence of CL/P has been reported among Asians while low incidence has been reported among African populations. In Europe, studies have revealed that 1.45-1.57 in every 1000 people are born with cleft lip and/or palate (Derijcke et al., 1996). In a study of all Danish children with clefts born between the years of 1976 and 1981 the incidence was 1.89 per 1000 live births (Jensen et al., 1988).

It has been difficult ascertaining the prevalence of CL/P in Nigeria, due to under reporting; previous studies have reported a prevalence of between 0, 2-0.3 per 1000 (Iregbulem et al., 1992).A very recent study have reported a prevalence of 0.5 per 1000 (Butali et al., 2014)

1.4 CONSEQUENCES OF CL/P

CL/P can impact on facial appearance and affect the health of children leading to adverse outcome on health, affecting feeding, speech, hearing, appearance, and dentition and quality of life (Rumsey and Harcourt, 2005). A higher morbidity and mortality rate has been reported in affected children than unaffected children (Mosey et al., 2009; Christensen et al., 2004). One study reported that 30-40% of children with CL/P were reported to have clinically significant behavioural and social difficulties such as shyness, reduced social competence, impulsive behaviour or learning disabilities/cognitive impairments (Endriga and Kapp-Simon, 1999; Hunt et al., 2005; Richman and Nopoulos, 2009). Complications associated with CL/P are facial disfigurement and temporary loss of function of affected parts of the oral cavity

1.5 COMPREHENSIVE CLEFT CARE.

The care of children with CL/P starts at birth and continues until adolescence. Treatment starts from childhood and continues until adulthood when facial growth is complete. The aim of treatment is complete rehabilitation so that these children can live a normal life. A multidisciplinary team approach to care has been recognized as the best option for treatment offered by a range of cleft specialists, following an established protocol (Marcusson et al., 2001; Hodgkinson et al., 2005; Clinical Standards Advisory Group, 1998).Caring for children affected with CL/P starts at birth and aims at holistic management and complete rehabilitation so that affected children can live a normal life. The general cleft protocol is described in Table 1 (Sommerlad 1994; Clinical Standards Advisory Group 1998; Hodgkinson et al 2005) and highlights the different stages of treatment as well as their timing.

Table 1.0: General protocol for CL/P

Age	Procedure	Specialist
Prenatal to 18 months	Emotional support; help with feeding / weaning.	Nursing
3 months	Primary surgery, Lip closure to improve function/alter appearance.	Surgeon
6 months	Palatal closure, Speech development, Placement of ventilation tubes to improve hearing.	Surgeon
9 months	Speech therapy, development and encouragement of speech, diagnosis of palatal dysfunction or incompetence.	Speech therapist
0-5 years	Audiology monitor hearing: support with either ear nose and throat surgery or hearing aids	Audiology
3-5 years	Surgical revision of lip and nose appearance to improve facial aesthetics, velopharyngeal surgery to improve speech.	Surgeon
From 7 years	Orthodontic use of appliances to correct teeth for treatment for good aesthetics and function. Done in permanent dentition.	Orthodontist
9-11 years	Secondary alveola grafting. Closure of or facial fistula.	Surgeon
17 to 20 years	Orthognatic surgery /re alignment of jaws to improve function and appearance.	Surgeon/Orthodontist
Across ages	Help for children to manage the demands of treatment, improve self-esteem and cope with teasing, transition to new schools, entering adulthood and the workplace.	Psychologists
Across ages	Assessment of family medical history to estimate Likelihood of CL/P recurrence in a future pregnancy.	Genetic counselling

1.5.1 Emotional support; help with feeding / weaning.

The potential emotional impact of the diagnosis and birth of a child with a long-term condition has been widely recognised, as has the need for adequate emotional support (Case 2000; Skirton 2006; Barr and McConkey 2007).

Research has revealed some of the emotional needs that parents may have upon hearing the news of their child's cleft. These include having the opportunity to talk, share their feelings and get emotional support from experienced professionals (Martin 1995; Strauss et al. 1995; Cleft Lip and Palate Association 1996; Byrnes et al. 2003; Johansson and Ringsberg 2004; Cleft Lip and Palate Association 2007). That is why early emotional support should be given to parents of children with CL/P.

Feeding has been recognised as a particular challenge in relation to the maintenance of a child's weight and growth as well as its potential emotional impact on parents (Endriga and Kapp-Simon 1999). Research has identified feeding as a main concern amongst parents of new born (Clifford and Crocker 1971; Brantley and Clifford 1979; Young et al. 2001; Farrimond and Morris 2004; Chuacharoen et al. 2009).

The ability to suck or feed efficiently may be compromised when an infant has a cleft (Reid et al. 2006; Reid et al. 2007).Feeding difficulties have been reported amongst parents of infants with clefts in one UK survey (n=100) (Oliver and Jones 1997).

1.5.2 Primary surgery, lip closure to improve function/alter appearance.

The importance of lip closure is its effect on the maxilla growth; lip repair creates tension within the muscle and introduces a medial pull on the anterior maxilla, thereby modifying growth of the maxilla. Seventeen different types of lip closure techniques were reported in one European survey (Shaw et al., 2001). The Millard rotational advancement technique (Millard, 1959) was recorded as most popular and used by 62.2% of European surgeons. The timing for surgery revealed that 46.6% of children with CL/P in Europe had completed lip repair at 3 months, while 91.7% of affected children had completed lip repair at 6 months. Some teams close the alveolus during lip surgery; others close the alveolus with the hard palate.

1.5.3 Palatal closure, speech development, placement of ventilation tubes to improve hearing loss.

In one European survey of 201 cleft teams, 21 techniques for palatal closure were reported (Shaw et al., 2000).About 49% of teams close the hard and soft palate in a single operation. It has been proposed that because palatal closure causes maxilla growth disturbances, that it should be delayed in affected children (Gilles and Fry ,1921; Schweckendick,1955Hotz and Gnoiski. 1986; Friede et al., 1986).However there is no evidence to support the fact that delayed palatal closure improved maxillary growth, but evidence has shown that delayed palatal closure causes speech problems (Robertson and Jolleys,1974;Witzel et al., 1984; Rohrich et al., 1996) .Presently, there is no agreement on how the right timing for closure of the hard palate or for how long palatal closure should be delayed to obtain to obtain the best maxillary growth without speech problems.

In one European survey of 201 cleft teams, 21 techniques for palatal closure were reported (Shaw et al., 2000).About 49% of teams close the hard and soft palate in a single operation. The most common method of palatal closure in Europe is the von Langebeck technique (von Langebeck, 1861), 28.7% of surgeons were reported to use this technique followed by the Wardill-Kilner pushback technique 19.7% (Shaw et al., 2000).

1.5.4 Audiology monitors hearing: support with either ear nose and throat surgery or hearing aids.

The main ENT problems in children with non-synrdromic clefts are otitis media with effusion (OME) also known as glue ear, non-suppurative otitis media (Dhilion, 1998; Albert etal., 1990; Handzic-cukeetal., 2001). OME occurs temporarily in children affected with non-syndromic C/LP and it is a treatable condition, and usually does not lead to hearing loss. Children with syndromic cleft present with other ENT problems such as atresia, meatal atresia and choanal atresia, hearing loss may occur in these children.

Studies have shown that children with non-syndromic cleft do not carry a higher risk of OME when compared to non-cleft children (Sheahan et al., 2003).In children with CL/P, OME is much less likely to resolve spontaneously when compared to children without CL/P , palatal repair does not necessarily lead to its resolution (Dhillion,1988). Some studies have shown that early palatal repair does improve hearing and speech (Sheahan et al., 2002, 2003, 2004). Early diagnosis of hearing loss and appropriate management lead to better outcome in

children with OME (Yoshigana-Itano et al., 1998). It is essential that newly diagnosed children with CL/P are screened for hearing loss and appropriate management commenced immediately. Early identification of hearing loss and treatable conditions such as otitis media with effusion (OME) is vital to optimising speech, language and cognitive development. Most developed countries, unlike developing countries run new born hearing screening programmes (NHSP) that avail children with CL/P the opportunity to be diagnosed early and receive prompt treatment. Early and routine insertion of ventilation tubes grommets to alleviate hearing problems have been advocated (Robison et al., 1999).

1.5.5 Speech therapy, development and encouragement of speech, diagnoses palatal dysfunction or competence.

Children with CL/P present with speech problems, formation of the sounds of speech may be influenced by deviations in oral structure and function (Harvold, 1970; Johnson and Sandy, 1999) and the risk of articulation problems is high in those with cleft lip and palate. Abnormalities of the hard palate, including an unrepaired cleft, a fistula after palate repair, maxillary collapse, or other abnormal configuration, affect tongue placement and posturing as well as airflow through the oral cavity thus altering phonation. Studies on 6 year old children with cleft lip and/or palate in Finland (Laitinen et al., 1998; 1999) suggest that as arch dimensions decrease and severity of the cleft type increases the risk of misarticulations.

The assessment of speech for children with cleft lip and/or palate has been the subject of considerable research effort in recent years (Sell et al., 1994; Harding et al., 1996; Wyatt et al., 1996) and has resulted in several objective scoring systems for cleft speech, including the Cleft Audit Protocol for Speech (CAPS) (Harding et al., 1996) and the Great Ormond Street Speech Assessment (GOSSPAS) (Sell et al., 1994, 1998).

1.5.6. Surgical revision of lip and nose appearance to improve facial aesthetics. velopharyngeal surgery to improve speech.

Correction of velopharyngeal incompetence (VPI) is the most important aspect of cleft care. Surgical intervention pharyngoplasty is indicated in affected children (Kawamoto, 1995; Pryor et al., 2006). Other revision surgery includes correction of fistulas, tongue flap closure, secondary rhinoplasty, alveola bone grafts, scar revision Abbe flap and orthognatic surgery.

The incidence of fistulas after palatal repair has been reported to be as high as 40% (Lehman 1992, 1995). Majority of fistulas occur in the hard palate and cause VPI, chronic rhinitis and malodour, therefore closure is important to improve on speech and hygiene.

More importantly, surgeons should take steps to prevent breakdown of fistulas, the following steps have been advocated;

i) Elevation of large palatal flap based on original incisions. ii) Excision of the scarred margins of the fistula so that no scar epithelium is left in the fistula site) Tension free closure of the nasal and oral mucosa) Use of anterior unscarred tissue to close anterior and large palatal defects') Bone grafting.

1.5.7 Orthodontic use of appliances to correct teeth for treatment for good aesthetics and function.

Children affected with CL/P require extensive and prolonged orthodontic treatment; this may be required in four different stages of the dentition and growth.

i) Pre surgical stage –infant orthopaedics using NAM appliances
ii) Late primary and mixed dentition stage- correction of anterior cross bites and occlusal discrepancies with removable appliances.
iii) Late mixed and early primary dentition stage-correction of occlusal anomalies and posterior cross bites using fixed appliances
iv) In the late adolescent stage after completion of facial growth-orthognatic surgery.

1.5.8 Psychological counselling for children with CL/P.

Psychological counselling aims at providing help for children to manage the demands of treatment, improve self-esteem and cope with teasing, transition to new schools, entering adulthood and the workplace. Studies largely in the USA has focused on the emotional difficulties parents caring for a child with a cleft may experience (Brantley and Clifford 1979; Palkes et al. 1986; Speltz et al. 1990; Krueckeberg and Kapp-Simon 1993; Speltz et al. 1993; Bradbury and Hewison 1994; Campis et al. 1995; Andrews-Casal et al. 1998; Slade et al. 1999; Pelchat et al. 1999b; Pope et al. 2005; Weigl et al. 2005). From the psychological perspectives, research aims to assess levels of stress amongst parents and investigate outcomes such as anxiety, depression and poor 'adjustment'.

1.5.9 Genetic Counseling

Genes that contribute to complex traits such as CL/P can be identified by using a combination of family collections, careful phenotyping, high-throughput genotyping, robust analytical strategies, and fine structure mapping and mutation characterisation.

Genetic counselling aims at assessment of family medical history to estimate likelihood of CL/P recurrence in a future pregnancy. The existing strategies for investigating gene involvement in CL/P are;

i) Animal models, particularly mouse and chick
ii) Studies of relevant populations through linkage disequilibrium
iii) Genetic linkage and association studies
iv) Chromosomal rearrangements
v) Studies of monozygotic twin discordance.

The thesis proposed the following hypothesis;

1.6 HYPOTHESIS

1. That the current status of cleft care in Nigeria may be below the World Health Organisation (WHO) recommended standards.
2. That the quality of treatment outcome in Nigerian children with CL/P may be dependent upon the surgical procedure and skill of the surgeon and rather than the type of service offered.

1.7 AIMS AND OBJECTIVES

The following are aims and objectives of the thesis

1. To report on the current status of cleft care in Nigerian using the WHO recommendations of standards for cleft care as a yardstick of measure.
2. To report on post-surgical treatment outcome in a selected Nigerian cleft population.
3. To compare treatment outcome in a Nigerian cleft population with that of a good practise archive population.

CHAPTER 2 : LITERATURE REVIEW

2.1 BACKGROUND

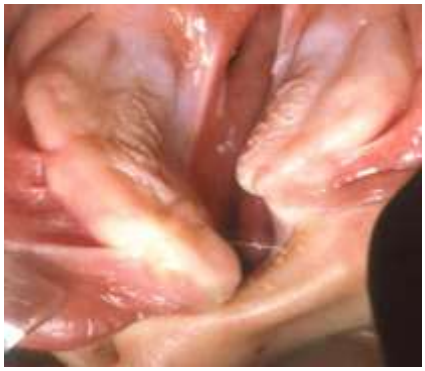
The purpose of this chapter is to review available literature on Evidence Based (EBD) cleft care. Several studies have been conducted in the field of CL/P, but few have actually provided evidence about cleft care. This literature review in itself is not exhaustive, but presents summaries of selected studies that meet its criteria.

2.2 METHODOLOGY

Traditional narrative method of searching relevant databases, including Scopus, Web of Science, PubMed and Google Scholar were using search terms such as : Unilateral cleft lip and Palate(UCLP), 'cleft care'/ and or Nigeria'; cleft care/and 'health care was done app. Inclusion and extrusion criteria were also applied; inclusion criteria were published studies on 'cleft lip and palate' 'cleft care' 'cleft care in Nigeria' to include web based articles and journals; exclusion criteria were unpublished articles on the subject matter. Only relevant literature on the subject matter were reviewed, The search yielded 46 papers on the subject topic, out of which 18 papers were relevant to the topic and was eventually summarised to give useful themes and salient points on key aspects of the literature review.

Of all the cleft subtypes, UCLP is said to be the most homogenous of the subtypes of clefts and occurs with sufficient frequency that reasonably sized samples can be gathered (Shaw et al., 1992; Bearn et al., 2001). It also represents a group of cleft disorders that require all the skills of a multidisciplinary team to achieve excellent outcome

Figure 2.1: Intra-oral view of UCLP



2.3 PREVALENCE OF UCLP

The World Health Organization has published a series of reports on international collaborative research on craniofacial anomalies (WHO 2002, 2003). In these reports, epidemiologic data on the incidence and prevalence of patients with cleft has been frequently presented.Consensus on the subject matter was agreed upon at two different meetings; Geneva (Switzerland) in 2000, and Utah (USA) 2001. In terms of epidemiology, it was reported that in general, both cleft lip (with or without cleft palate) and isolated cleft palate have an incidence of approximately 1 in 600 live births (WHO, 2002). Though there are differences in incidences among countries, the birth prevalence of cleft lip (with or without cleft palate) is around 1:1000 births and that in two thirds of patients diagnosed, the left side is more frequently affected. In parts of Europe, a study of all Danish children with clefts born between the years of 1976 and 1981 the incidence was 1.89 per 1000 live births (Jensen et al., 1988). Within this period of study, there was a distribution of 33.5% of cleft lip, 39.1% of UCLP and BCLP combined, and of 27.4% of isolated cleft palate. Of all the cleft subtypes, UCLP is said to be the most homogenous of the subtypes of clefts and occurs with sufficient frequency that reasonably sized samples can be gathered (Shaw et al., 1992; Bearn et al., 2001).Another study between 1912 and 1933 (Sanders, 1933) reported prevalence of cleft in 392 patients from Rotterdam that were operated on in two different hospitals. A prevalence of 20.6% was found for patients with BCLP and of 32.4% for those with UCLP. The remaining 47% had other combinations of clefts.

2.4 CHARACTERISTICS OF COMPLETE CLEFTS

Complete cleft of the lip and palate presents with retrusion of the maxilla and a reduction in size of the maxilla and mandible. Impairment of anterio-posterior growth of the maxilla is the most prominent feature of complete cleft (;Semb,1991;Capelozza et al., 1993).Increase in inter ocular width has also been reported (Dahl,1970;Aduss et al., 1971;Ishiguro et al.,1976; Ross andJohnston,1972;Semb,1991, Capelozza et al., 1993), however the facial width and basal maxillary width is said to be similar to that of a non -cleft population (Dahl,1970;Smahel and Brejcha,1983), Complete clefts are said to present with vertical changes with significant reduction of the posterior midfacial height and increased lower anterior facial height..

2.5 GROWTH PATTERN IN COMPLETE CLEFTS

There are differences in growth pattern of patients with complete clefts when compared to non-cleft patients, In a longitudinal study of 257 patients with UCLP and 90 patients with BCLP there was gradual reduction in maxillary prominence over the years when compared to the Non-cleft group (Semb,1991).

2.5.1 Factors influencing Facial growth disturbance in complete clefts.

In 1996, Semb and Shaw identified several factors that may disturb normal maxillofacial development, common among those identified are congenital dysmorphology, intrinsic variations and surgical iatrogenesis (Semb and Shaw, 1996).

2.5.1.1 Congenital dismorphology of the Midface.

UCLP typically presents with asymmetry of the anterior maxilla with an upward lifting of the pre-maxillary region, distortion of the Nasal septum with bulging and upward orientation of the anterior nasal spine. In one study (Kane et al., 2003) using Computed Tomography (CT) ,severe osseous dysmorphology in patients with UCLP were reported, further extreme asymmetry of the primary and secondary landmarks as well as displacement of the premaxilla towards the non-cleft side were reported. The findings of this study support the fact that UCLP affects the development of the entire face.

Quantitative morphometric analysis of children with UCLP can be done using matrix analysis of CT landmarks; in most cases anomalies were found to be evident from the initial early stage after failure of union of facial processes (Latham, 1973). These anomalies were suggested to be as a secondary response to extra capsular influences (Kimes et al.,1988),while it has been said to be possibly due to secondary distortion during the foetal period that may explain the variation in clinical variation after birth (Fergusson,1993). Variations in swallowing, tongue position bucco-pharyngeal neuro muscular activity have all been observed and may actually contribute to diversity of segmental relationship observed at birth.

2.5.1.2 Other intrinsic factors

Inherent differences in craniofacial morphology of patients with UCLP have been observe and reported (Mars and Hiuston1990; Capelozza etal., 1993). Consistent findings have shown that the size of the craniofacial dimensions including the maxilla is smaller, when compared to non-cleft patients.

2.5.1.3 Functional Adaptation

Children with UCLP have been observed to present with impairment of nasal airway, as a result of nasal capsule deficiency and hypertrophy leading to increased upper airway resistance (Drettner,1960;Aduss and Pruzansky,1967;Warren and Hairfiedl,1990).One utero study confirms the evidence of airway deficiency as a result of nasal capsule deficiency and hypertrophy (Siegel,1987) ,Nasal obstruction occurs in un operated cleft , this leads to functional and postural adaptation resulting in the characteristic mandibular form and position associated with clefts. Children with UCLP as a result of the maxillary narrowed arch, typically presents with lowered tongue position and mouth breathing.

2.5.1.4 Surgical Iatrogenesis

The impact of surgery on maxillary growth remains a central issue to be resolved (Shaw and Semb,1996). This is because surgically induced growth impairment have been reported several years ago (Harvold, 1954).There are controversies surrounding the impact of surgery on maxillary growth, as well as whether lip and palate surgery is more harmful in children with CL/P (Mars and Houston, 1990; Badrach, 1990; Nomando et al., 1992).Other aspects of surgery that may have an impact on the maxilla are the type of surgical technique, timing of the surgery and skill of the surgeon.

2.5.1.5 Operated versus Unoperated cleft

Studies have also shown that adults with un-operated complete clefts showed smaller and more protruded maxilla with increased anterior facial height (Capelozza et al., 1993; Mars and Houston, 1990). Studies in a Brazilian cleft population have also shown that Lip repair in isolated cleft of the alveolus and of the palate has no known deleterious effect on maxillofacial and maxilla mandibular growth (Nomando.1992). The converse is said to occur when surgical repair of the lip and palate is done in complete cleft of the lip and palate, its impact was shown to be more on the anterior-posterior dimension leading to downward growth rotation. Similar findings were also reported in other cleft population (Dahl, 1970; Bishara 1973). However surgical repair of the lip and palate does not seem to affect mandibular growth (Semb and Shaw, 2013).

2.5.1.6 Lip and Palatal Surgery in complete clefts.

Scar tissue formation after surgical repair especially in the maxillary, palatine and pterygoid sutures have been said to prevent forward and downward translation of the maxilla, thereby

impeding growth (Ross and Johnston,1972;Semb and Shaw, 1996).Animal studies has shown that it is the increase in pressure and restriction from the repaired lip on the maxilla that causes maxillary growth restriction (Bardach and Kelly,1988 ;Bardach etal.,1984).Similar studies in humans have also been conducted in cleft population to report the effect of lip and palate surgery on children with UCLP in some countries; Sri lanka (Mars and Houston,1990,Capelozza,1996), Danish (Dahl,1970) and Japanese population (Yoshida et al., 1992) substantial reduction in maxillary length was reported. They concluded that the impact of surgery is more on the maxillary base and dental arches resulting in reduced maxillary antero-posterior vertical dimension, rather than on the transverse dimensions (Dahl et al., 1981; Bergland and Sidhu.1974; Enemark et al., 1990). These studies also reported that surgical technique used in repair may influence degree of scar formation and determine the extent of malocclusion. Several authors have also drawn attention to the contraction of collagen fibres that arise in granulation tissue and may restrict growth of the maxilla (Graber, 1949, 1954; Kremenak et al., 1967; Ross and Johnston, 1972; Friede, 1977; Kremenak, 1984; Mars and Houston, 1990; Capelozza et al., 1996; Kuijpers-Jagtman and Long, 2000).

2.5.1.7 Surgical skill, Technique and Timing

Good surgical skill of the surgeon has been reported to reduce scar tissue formation in patients with UCLP with high volume operators likely to have better surgical skill than low volume operators (Shaw et al., 1992; Ross and Johnston,1972,Prahl Anderson and Ju,2006). Inter centre comparison studies of two centres reported better treatment outcome in one centre as a result of the high number of surgeries undertaken when compared to the other centre that had a lower number of surgeries undertaken (Shaw et al., 1992, Ross et al., 1990).
In summary, the impact of surgery in patients with UCLP; scar tissue formation and pressure on the maxilla from the lip after surgical repair are major factors that can restrict maxillary growth.

2.6 EVIDENCED BASED CLEFT CARE

Evidence based care allows the integration of the best research evidence with clinical expertise and patients values. The current scientific basis for the identification and dissemination for optimal clinical intervention and management of (CFAs) is weak and poses significant challenges to providers (Shaw et al., 2015). Currently, none of the element of treatment for cleft care has been subjected to the rigors of contemporary clinical trial design

and there is no firm evidence to determine new protocol of treatment for children with cleft lip and palate (Shaw et al., 2015).

2.6.1 Treatment Choices

Evidence provided in cleft studies for surgical care and other ancillary care such as pre-surgical orthopaedics, orthodontics, speech and secondary revision are based on disappointment from former practises rather than scientific evidence. For example, a survey of European cleft services revealed 194 different surgical protocol for Unilateral cleft lip and palate(UCLP) alone from 201 teams (Shaw et al., 2000), confirming the possible lack of consensus among surgeons as to the most effective surgical protocol for UCLP. Currently, there is weak evidence to confirm the best timing and most effective surgical protocol for children with UCLP (Shaw et al., 2015). The same can be said for most developing countries, Nigeria inclusive. This chapter continues with available evidence for treatment protocols in children with UCLP.

2.6.2 Pre-surgical Orthopedics (PSO)

Pre-surgical orthopaedics (PSO) has been a controversial issue because of its consistent lack of positive evidence in the literature (Kuijpers-Jagtman and Long, 2000; Shaw et al., 2015). The many advantages of PSO are said to be; improvement in feeding (Trankmann, 2000; Turner et al., 2001), maxillary growth redirection (Goldberg et al., 1988) and psychological effect (Jones et al., 1982). Advocates of pre-surgical orthopedics claims that it allows for alignment and correction of the nasal cartilage in children with CL/P, thereby minimizing the formation of scar tissue and producing better post-operative aesthetic results (Rosenstien et al., 2003;Grayson et al., 2008;Rubin et al., 2015; Mandwe et al., ,2015). The usual method is to use either a passive or active appliance to achieve treatment objective, which said to be the conversion of a wider cleft to a narrower cleft (Grayson et al., 2015; Aminpour and Tolliferson, 2008). Surgical advantage of PSO have been said to include less dissection and less surgical trauma (Mandwe et al., 2015). A previous study that determined the attitude of African cleft care providers towards PSO, reported a positive attitude towards its use (Adeyemi and Bankole, 2012).

However most of these studies are yet to be subject to rigorous clinical trial design that can provide sound scientific evidence of the advantage of PSO in children with UCLP. To authenticate such a claim, a randomised trial of 54 Dutch patients with UCLP, who had surgical repair using the same protocol in three different centres, revealed that PSO had no

effect on feeding and early growth of the babies (Prahl et al., 2005) .The patients were randomised into 2 groups, so that one group received PSO from two weeks after birth until operation of the posterior palate, the other group did not receive such type of intervention, the results showed that there was no difference between maxillary arch dimension and collapse between the two groups(Prahl et al., 2001; 2003;2005). Concerning dental arch assessment, also there was no known difference in occlusion between the two groups as well as facial aesthetics (Prahl et al., 2006).

In some respects, it is possible to make a stronger theoretical case can be made for PSO in patients with Bilateral cleft lip and palate (BCLP) especially because of the prominence of the pre-maxilla, though in one controlled retrospective comparison study, PSO was not found to be beneficial even in patients affected with BCLP (Ross and McNamara, 1994).

A systematic review of 53 aesthetic outcome measures in cleft patients after surgical repair, reported lack of consensus, validation and non-reproducibility of many of the aesthetic outcomes. This further confirms the lack of evidence for PSO in the treatment of children affected with UCLP.

Though very recently one study (Rubin et al., 2015) compared treatment outcome in patients who had PSO as part of their treatment protocol; specifically those who had Naso-alveola Molding (NAM) appliance were compared with patients who did not have the NAM appliance. The study reported that there was significant differences ($P<.0.001$) between patients who were NAM prepared and those who were not, it was concluded by cleft surgeons that children with CL/P who are NAM prepared are likely to have less severe clefts, and be among the best of surgical outcome cases and less likely to need revision surgery. However the likelihood of bias in this study is very high, this is because only photographs were rated by cleft surgeons, photographic rating of cleft treatment outcome have been reported to be a reliable method of assessing aesthetic outcome in children with CL/P (Asher Mc-Dade et al.,1991, Bearn et al., 2001). However sources of bias in cleft research has been identified (Shaw et al., 2015) and should be minimized by appropriate designs of comparisons. In summary, systematic review of randomized trials are general rule for making fair comparisons and providing evidence in research. Until such systematic reviews prove the validation and reliability of the effect of pre-surgical outcome on cleft outcomes, it may be difficult reaching a consensus and agreement as to what effect PSO has on treatment outcomes. Currently, there is weak evidence as to the long term effectiveness of PSO in children with UCLP (Shaw et al., 2015).

2.6.3 Surgery

Surgical repair of the lip and palate is indicated in children with CL/P, the impact of surgery on maxillary growth however remains a central issue to be resolved (Shaw and Semb, 1996). This is because surgically induced growth impairment have been reported several years ago (Harvold, 1954).There are controversies surrounding the impact of surgery on maxillary growth, as well as whether lip and palate surgery is more harmful in children with CL/P (Mars and Houston, 1990; Badrach, 1990; Nomando et al., 1992).There are also controversies concerning the most effective surgical technique, timing and skill. Intervention studies are advocated to resolve such controversies; empirical research in studies of health care intervention that do not use randomization is likely to give an overinflated view of effectiveness (Shaw et al., 2015).

2.6.3.1 Surgical choices in UCLP

The choice of surgical technique is a matter of operator preference, the usefulness of various techniques and their timing has been debated (Semb and Shaw,1998). For example, some operators prefer early bone grafting and to delay palatal closure, while others prefer late alveola bone grafting and early closure of the palate. A review of studies allows the variation in techniques and their advantages to be reported. For example, some studies show that early bone grafting and perioplasty in the neonatal period is detrimental to growth (Rehrmann et al., 1970; Robertson and Jolleys, 1968; 1974; Fride and Johson, 1974; Helliquist and Svardstrom, 1990). While other studies reported satisfactory growth after primary bone grafting (Nordin et al., 1983;Roseintein et al.,1982;1991), reduction of vertical facial growth was reported after neonatal bone grafting was reported in another study(Branstorm,1991).There are ongoing research to determine the most effective surgical technique and timing for children with UCLP.

2.6.3.2 Delayed versus Early palatal closure

The most controversial issue in surgical repair of UCLP is the timing of the hard palate (Shaw and Semb, 1998). Presently there is no consensus as to the influence of early or late palatal closure on maxillary growth. Several years ago, Gillies and Fry advocated delayed closure of the palate; the purpose was to avoid growth disturbance (Gillies and Fry, 1921).This was disputed by researchers (Witzel et al., 1984; bardach and Mooney, 1984) who advocated early palatal closure to prevent speech impairment. Some studies have advocated

delayed palatal closure (Friede et al., 2007; Lilja et al., 2006; Nollet et al., 2005; Mostled, 1999). In 2007, Friede reviewed published papers on delayed palatal closure in children with UCLP; the studies showed that two stage palatal closures in children with UCLP have excellent treatment outcome in affected children. Similar findings were reported in Goteborg, Sweden by Lilja and colleagues. One major study that showed the impact of delayed palatal closure on treatment outcome, was a meta-analysis of 1236 patients with UCLP (Nolliet et al., 2005), delayed palatal closure was associated with improved treatment outcome. Another study showed that treatment outcomes of centers in Nijmegen, Netherlands with the two stage palatal closure compared favorably with best centers in the Eurocleft studies (A, B&E).

One Scandinavian study (Farzanah et al., 2008) did a comparative analysis of 34 Adult patients with UCLP, who had surgical repair done at 8 months using the von-langenbeck technique and 27 Adult patients who had repair done at 18months by theWardhill technique. When compared with 151 controls, they discovered that early or late closure had no influence on facial growth. Researchers in other countries; Turkey (Savaci et al., 2005), Poland (Fudalej et al., 2007), Germany (Swennen et al., 2002) and Brazil (Silva Filho et al., 2001) have also conclude that early palatal closure have little influence on maxillofacial development. In a clinical trial conducted in Brazil, craniofacial morphology in children with complete UCLP treated with 2 different surgical protocols were evaluated; the Bauru protocol (lip closure mean age; 9 months and palatal closure mean age; 19months) was used to treat 53 affected children, and the Malek protocol (Lip and soft palate closure at 5.5 months, palatal closure at 20 months) was used to treat 22 children. The two stage palatal closure technique did not have much influence on growth.

In Germany, a multicenter study (Swenen et al., 2002) compared two different treatment protocol; Hanover technique with delayed closure of the palate and Malek technique with early closure of the palate. Thirty-six children with UCLP were treated with the Hanover technique, while 26 children with UCLP were treated with the Malek technique. The results showed that there was no difference in treatment outcome with either the early or late palatal closure. In a Turkish cleft population, Savaci and colleagues showed there was no difference in treatment outcome in children treated with 1 stage palatal repair before 10 months of age and those treated with a 2 stage palatal repair at 15 months. A similar study in a polish cleft population showed that there was good craniofacial symmetry and good maxillo-mandibular relationship with adequate overjet in children with UCLP after one stage palatal repair. In summary, palatal closure in children with UCLP can be done early before the age of 10

months or late after 12 months, advocates of early palatal closure claims that it improves speech, while advocates of late palatal closure claims it aids maxillary growth. However, most of the studies reviewed have shown that early or late palatal closure has no significant influence on maxillofacial growth. However due to the variability in craniofacial form and the difference in response to treatment, early or late palatal closure may be said to have some influence on maxillary growth in some populations (Mostled, 1999).

2.6.3.3 Primary bone grafting

Primary bone grafting is said to impair maxillary growth (Rehman, 1970; Robertson and Jolley, 1968; 1974; 1983; 1990). One study (Jolleys and Robertson.1972) reported the impact of primary bone graft on 14 pairs of patients, who were matched for type of cleft, amount of tissue deficiency and degree of cleft. One of the patients in each pair received autogeneous bone graft at the age of 15 months while the other did not and served as control. The patients were followed up at 5 and 11 years, there was maxillary growth impairment in those who received bone graft. Similar studies (Aduss and Prunzanky, 1967; Friede and Johanson.1974) have also reported the detrimental effect of primary bone grafting on maxillary growth and advocated the discontinuation of the procedure. More recent studies (Nordin et al., 1983; Roseintein, 1982; 1983) favor primary bone grafting, because they found no significant growth impairment in patients who had early bone graft.

2.6.3.4 Push back procedures

The impact of push-back procedure on maxillary growth when compared to palatoplasty without lengthening of the palate was also studied.Friede and colleagues found that palatal push back impaired palatal development in a Scandinavian cleft population (Friede et al., 1991). In a Japanese cleft population, the vomer flap was found to induce better maxillary growth when compared to the push-back procedure (Tanimo et al., 1997). Presently the cleft literature is yet to establish the most effective surgical procedure and best timing for surgical intervention in children with UCLP. It is hoped that future studies will establish these areas.

2.6.3.5 Secondary Procedures

Secondary surgical procedures such as pharyngeal flap closure, secondary alveola bone grafting and other secondary procedures such as lip and nose revision have been found to impair maxillary growth (Semb and Shaw, 1996)

2.6.3.6 Pharyngeal Flap Surgery

Direct restraint of maxillary growth was found after pharyngeal flap procedures (Subtelny and Pineda, 1978).Pharyngeal flap surgery has also been associated with mandibular adaptation as a result of changes in facial growth (Ren et al., 1983). However, in a study that compared 29 cases of UCLP with a control, there was no difference in growth pattern when compared with the control group (Semb and Shaw, 1990).

2.6.3.7 Secondary alveola bone grafting

The impact of secondary alveola bone graft on maxillary growth has been reported (Enemark et al., 1987; Semb,1988; Williams, 2003). Enemark and colleagues reported no growth impairment of the maxilla anteriorly and minor impairment of little significance in the vertically (Enemark,1987).Semb found out that secondary bone grafting had no influence on maxillary growth(Semb.1988).Williams and colleagues reported that increase in age and ethnicity are predictors of poor outcome of secondary borne graft in children with UCLP.

2.6.4 Multidisciplinary Treatment

The need for multidisciplinary approach has been stressed in the past, but there is lack of sufficient evidence based research to prove its effectiveness (Robin et al., 2006). Treatment of children with UCLP requires team work, with specialists such as surgeons, orthodontists, speech therapists, audiologists, psychologists and peadodontists. Long term results of treatment have been said to be unpredictable with successful cases of surgical repair ending up with severe growth retardation of the maxilla and an unacceptable appearance in adulthood (Semb, 1991).The consensus on fundamental elements of treatment of cleft lip and palate have been agreed previously to include multidisciplinary team work, centralization of care, high volume care,teamcontinuity,long-term planning from birth to adulthood, standardized protocols, documentation evaluation, follow-up studies, research, training and quality assurance (Shaw et al., 2001;Wellens and Vander poorten,2006). Recent advances in the impact of multidisplinary approach to treatment of children with cleft lip and palate reports an improvement in functional and aesthetic outcome (Tollerferson et al., 2008).

2.6.5 Treatment Outcome

The ultimate goal of cleft care is the restoration of the patient to a normal life as far as possible. However, ‘normalcy’ in children with cleft lip and palate represents the restoration

of different aspects of anatomic form and function in the parts affected by the cleft. Measurement of treatment outcome in essence aims at determining to what extent 'normalcy', has been restored as well as giving an indication of the deficits that persist despite treatment. Indices to measure facial appearance, speech, hearing, and dento-facial development have been developed for children with UCLP, with the aim of indicating treatment success or failure. The reproducibility and validity of such indices are important, ideally measured outcomes should be patient-centred, measuring those things that matter most to patients and their caregivers, rather than sophisticated measurements that may have little relevance to everyday life (Shaw et al., 2015). More recently, there have been suggestions to include qualitative measures within randomized trials and other comparison studies. These may reveal issues that are important to parents and patients that differ from those clinical outcomes selected for measurement in a trial .Therefore qualitative findings could inform the selection of a preferred treatment pathway, when significant difference are not be found with objective measures,.

Quality of life measures aim at identifying self-perceived variables that are of importance to the individual, in this case 'children with UCLP'. So that studies using qualitative approaches could facilitate the development of measurement tools that have been informed by the lay perspective. For example, it is known that measurements of aesthetic and functional outcomes in isolation are not good predictors of emotional (psychological) adjustment and well-being, and, consequently, there is a pressing need to identify the self-perceived variables that contribute to the quality of life of individuals with clefts.

2..6.5.1 Measuring Treatment Burden

Measurement of treatment burden has received little attention in cleft studies; yet cleft care can be burdensome, bearing the combined total number of operations and other treatment episodes, along with appointments for the first 20 years of life of these children (Semb, 2011). The desire of patients and parents is to reach a point where the stigma associated with having a cleft is completely eradicated, and as such parties involved are likely to accept proposals and comply with protocols of care that are recommended by specialists. Also because the consequences of cleft lip or palate may be apparent through every phase of childhood and adolescence, the disciplines involved in care are in a position to recommend some form of intervention. There is also the pain and suffering and the disruption to family life, employment, and school attendance, may have an adverse effect on the patient's sense of self-determination or

locus of control. Supplementary orthodontic interventions such as NAM, pre-surgical orthopedics, primary dentition orthodontics, and maxillary protraction have been said to impose extra burden on patients (Severens et al. 1998; Kujipers-Jagtman et al., 2000). To measure treatment burden, clinical trials will need to accurately record the total number of ancillary interventions and clinical visits in addition to surgical episodes. This recording was done for speech therapy visits in the Scandcleft project, and very large variations among patients were revealed (Nollet et al., 2006). There was positive correlation between speech problems up to age 5 years and attendance, creating an extra burden on the patient and family. However, studies on 'treatment burden' are few with small sample size of patients, hence subject to a number of biases and to large variations in outcome measures. There is a great need to conduct well-designed, authoritative evaluations of the effectiveness of speech and language therapy.

2.6.5.2 Measuring Cost Benefit

Global economic recession with shrinking budgets make it mandatory for clinicians to measure cost benefits of treatment offered. Surgical operations for children with CL/P are expensive treatment episodes, so that successful initial operations that can minimize the need for multiple secondary revisions are highly desirable. The application of health economics techniques to the field of cleft care is yet to be reported (Shaw et al., 2015). Economic prioritization models tend to use decision analysis and simulation to assess the resource costs and patient benefits of current treatment patterns and the "cost effectiveness gap" or potential gain from alternative surgical procedures for cleft care.

The literature review summarised findings on treatment outcome as well as measures used to determine treatment outcome in children with CL/P. The following treatment outcome measures have been reported previously for children affected with CL/P; assessment of craniofacial form and soft tissue profile using cephalometic x-rays; assessment of facial and naso-labial appearance using photographs; Dental arch relationships using study casts, speech assessments using clinical video and audio assessments (Shaw et al., 1992; Mostled et al, 1992; Asher-Mcdade et al., 1992 ;Mars et al.,1992; Bearn et al., 2001).

2.6.5.3 Measuring Dental Arch Relationships

Dental arch relationships in children with UCLP can be assessed using either cephalometric x-rays, photographs or study models. Assessment of study casts are however one of the most reliable method of assessing surgical outcome in children with UCLP (Attack et al., 1997; Mars et al., 1987), In general, study casts of patients have been used to determine several

dental and occlusal variables such as tooth size and arch width. Early studies used measuring callipers or gauges to record measurements on study casts (Hunter and Priest 1960; Andrews,1972; White,1974),the use of measuring callipers still remains one of the most common traditional method of measuring study casts (Redahan and Lagerstrom,2003).

2.7 EVIDENCE BASED CARE FOR CHILDREN WITH CL/P IN NIGERIA

Few studies (Oginni et al., 2014; Olasoji et al., 2011; Adeyemo et al., 2009) have reported on cleft care in Nigeria. The majority (69.2%) of hospitals in Nigeria began cleft care delivery between 2006 and 2010, with 73.3% having designated cleft clinic locations and 66.7% offering interdisciplinary care, services offered include cheiloplasty and palatoplasty. Other aspects of cleft care are provided sparingly in most centers due to paucity of manpower. Challenges with hospital administration, securing bed and theater spaces, drug availability, and performing laboratory investigations were the common limitations reported (Oginni et al., 2014). Majority of the hospitals are government owned hospitals and 80% began cleft care between 2006-2010. Nigerian cleft care specialists are mainly surgeons/anesthetist. Multidisciplinary approach is rarely practiced; surgical repair of the cleft is the main form of treatment offered to children with UCLP (Olasoji et al., 2011). Audit of cases seen at the hospitals are not done.

Three main approaches have been adopted to alleviate the burden of care in developing countries, they are; establishment of high-volume indigenous centres; financial support for indigenous cleft centres to sponsor free surgery for children with CL/P and surgical missions (WHO 2002, Peterson et al., 2005). In Nigeria, the main approach to cleft care is through financial support for indigenous cleft centres by one main charity organisation (Smile Train) who partners with indigenous hospitals to provide free surgery. Such intervention has been reported to lead to an increase turn out of affected children (Onah et al., 2008; Onah and Ezinwa, 2012). However there are many affected children, who are unable to access and receive care (Oginni et al., 2014). This is because majority of the partner hospitals are located in major cities and towns thus making access to care, difficult for affected families who incidentally hail from rural towns and villages (Oginni et al., 2014).

2.8 THE NIGERIAN HEALTH CARE STRUCTURE

The Nigerian health system is somewhat complex including both private and public health providers. Private Health Providers include Non-governmental Organisations (NGOS),

Community based Organisations, Religious organisations and Traditional care providers and Private health providers. NGOs and community based organisations are non- profit organisations that aim to finance health care by offering free treatment, private providers aim at providing health service for the purpose of profit making.

2.8.1 Public Health Care Sector

The Public health care sector is funded mainly by the government and operates a three-tier structure: tertiary, secondary and primary levels of health care, with responsibilities at Federal, State and Local government levels. All three levels of health care are involved in major health function, stewardship, financing and service provision.

The Tertiary level of care is coordinated by the affairs of the Federal Ministry of Health (FMH) and is involved with health policy, budgeting, health management information systems and international relations on health matters. In general, the FMH coordinates health services at the tertiary level of care. The Secondary level of care is coordinated by the State Ministry of Health (SMH) and is responsible for secondary care hospitals, and the regulation of and technical support for primary care services.The Local government coordinates the affairs of Primary Health Care (PHC) and is responsible for primary health care at community level. The community plays a major role in health care delivery and serves as the support structure for the implementation of primary health care services.

The Federal Ministry of Health (FMH) is at the helm of affairs of the Nigerian health system, its mission is to develop, implement and plan programmes that allow quality, efficient and affordable services to be delivered in Nigeria. It comprises of several departments to include; Family Health Department (FHD) - Concerned with creating awareness of reproductive, maternal, neonatal and child health. Its role is to ensure balanced nutrition for infants and young children, as well as for their mothers; Department of Public Health (DPH) which undertakes health promotion, surveillance, prevention and control of diseases. It also coordinates the formulation, implementation and evaluation of public health policies and guidelines; Department of Planning Research and Statistics (DPRS) that undertakes research in collaboration with other health departments, agencies and institutions. It also develops health policies and budgets, in addition to monitoring their implementation; Department of Hospital Services (DHS), oversees the country's federal tertiary hospitals, teaching hospitals and eye centres and develops oral health and nursing policies, and is involved in the coordination of Nursing and Midwifery programmes.; Department of Food and Drug (DFD)

Services, this department formulates guidelines and policies on food and drugs, ensuring ethical delivery of pharmaceutical services nationwide. It also coordinates the affairs of the Institute of Pharmaceutical Research and Development, as well as the National Association of Food and Drug Administration and Control (NAFDAC).

2.8.2 Integrating Cleft Care Services within the Federal Ministry of Health (FMH).

Cleft care services can be suitably placed in the following departments; Family Health Department (FHD); this department may be involved in the care of the health of the family of children with oro-facial clefts; including support and counselling and inform families about the risk of having a child with CL/P.; Department of Planning, Research and Statistics ; this department may be involved in cleft research and auditing, so that problems associated with cleft care can be highlighted and reported with possible solutions proffered ;Department of Public Health; this department may create awareness of oro-facial cleft and help dismiss the associated stigma as well as promote cleft care; Department of Food and Drug (DFD) Services - Children with oro facial cleft may be undernourished as a result of feeding difficulty associated with unrepaired cleft. The responsibility of this department would be to ensure that affected children are adequately nourished;Department of Hospital Services ;This department may ensure provision of comprehensive cleft care for children affected with oro facial cleft, through adequate funding and staffing of the hospital with specialists.

2.8.3 The State Ministry of Health (SMH) and Local Government.

The State government provides secondary health care and replicates all the function of the federal ministry at the state level. The Local Government Authority (LGA) provides primary health care at the grass root, majority of children with CL/P are reported to be from rural areas. The LGA may integrate cleft care by organising outreaches, campaigns and health education for children.

2.9 CURRENT PROVISION OF CLEFT CARE SERVICES IN NIGERIA.

Presently, cleft care is offered at two tiers of health care in Nigeria, namely Tertiary and Secondary health care levels, it is not offered at the primary health care level. At the tertiary level of care, cleft services are provided by specialists working in teaching and other federal hospitals, including military and naval hospitals. Provision of care is sponsored mainly by non-governmental organisations (NGOs), who are in partnership with these hospitals. Treatment offered at this level is mainly surgical care and rarely comprehensive, secondary

care such as orthodontic and speech therapy is not offered. Health promotion and oro facial cleft outreach programs are conducted in the rural areas by specialists working in tertiary hospitals. Recruitment of children born with CL/P for surgical repair is done during such outreach programs. At the secondary level, cleft care is offered mainly in private hospitals by cleft specialists; the majority of whom work in state hospitals but also do private practise. The services offered are largely surgical repair done in private hospitals on a fee for service basis. Health promotion and awareness of oro-facial conditions is rarely offered at this level.

Table2.1 Showing the three tiers of health care in Nigeria and cleft care services offered

Level of Care	Provider	Cleft care services offered
Tertiary care	Federal Government of Nigeria	Primary surgical repair Health education/support Campaigns/Outreaches to recruit patients
Secondary care	State government	Primary surgical repair Health education/support
Primary care	Local Government	No services available

2.9.1 Financing cleft care in Nigeria

In general terms, health care financing system involves the means by which funds are generated, allocated and utilised for health care. It involves three basic steps; collecting revenue; pooling of resources and purchasing services (Carrin et al.,2007;Gottret etal.,2006).The most commonly used mechanisms in implementing health care financing are ;tax based financing; out of pocket payments; donor funding and health insurance (Gottret etal.,2006).The success of the different mechanism can be measured by the overall efficiency of equity of access; health outcomes; revenue generation and efficiency, user and provider behaviour (Palmer et al., 2004).

Health care in Nigeria is financed by a combination of tax revenue, out-of-pocket payments, donor funding, and health insurance (WHO, 2009). Tax-based systems are health financing systems where government revenues are the main source of health care expenditure (WHO, 2004). Funds are usually generated through taxation or other government revenues. Revenues

are raised at the federal, state, or local government levels. However, the federally generated revenue which is shared according to a formula forms the majority of the funds for the other tiers of government.

The total government health expenditure as a proportion of THE was estimated as 18.69% in 2003(Soyinbo et al., 2002; 2003) and has increased to 27.6% in 2013 (WHO, 2010-2014). The budgetary allocation for health is still below the 15% signed by the Nigerian government in the Abuja declaration (WHO, 2009), given this level of government spending, it will be very difficult to provide essential health care services, and health care in Nigeria will always be at the peril of underfunding by the government. For example, in 2014, healthcare was allocated just 5.6% of the budget (approximately N262.74 billion), which is below the anticipated 15% agreed previously by the African Union of Health Workers (wwwgov.ng).

2.9.2 Out of pocket payment

Out of pocket is payment for services at point of use. The charges levied for health care services are referred to as user fees to include any combination of drug costs, medical material costs, entrance fees, and consultation fees (Largarde and Palmer,2006).Out-of-pockets account for the highest proportion of health expenditure THE and has varied over the years between 64.59% to 74% (Soyibo, 1999; 2002; Soyibo et al., 2003;2005). The implication of this is that households bear the highest burden of health expenditure in Nigeria.

2.9.3 Donor Funding

Donor funding, refers to financial assistance given to developing countries to support socio economic and health development. Financial assistance to Nigeria has not been tremendous,it has witnessed a declining trend since the return of the democratic governance in 1999 (World Bank, 2010). The major challenges in Nigeria with donor funding are effective coordination of the funds and tracking donor resource flow (WHO, 2009). Cleft care in Nigeria thrives solely on donor funding, the Smile Train Organisation is a cleft charity organisation that sponsors free surgical repair for affected children (www.smiletrain.org). The main aim of Smile Train is to finance and support care in children affected with CL/P from poor families by funding surgical care. While its mission is to provide a sustainable approach to a single solvable problem: CL/P. The organisation adopts an approach to support indigenous cleft care units by establishing partnerships with hospitals. The organisation thrives on the

principle of operating of one of the most cost-efficient cleft care systems. Partner hospitals are given grant to cover cost of surgical repair alone, which is estimated at a cost of $250 per child, grants are then given based on the number of patients seen at each hospital.

2.9.4 Health Insurance Scheme

One system of health insurance is the Social Health Insurance (SHI) is of financing health care through contributions to an insurance fund that operates within a tight framework of government regulations (Kutzin, 1998). It provides a pool of funds to cover the cost of health care; every citizen is required to make contributions. Governments may contribute on behalf of the poorest and the unemployed; employers also usually contribute on behalf of their employees (Largarde and Palmer, 2006).Another system is the National Health Insurance Scheme (NHIS) which was established by the Nigerian government under Act 35 of 1999 with the aim of improving access to health care and reducing the financial burden of out-of-pocket payment for health care services (http//nhis.gov.ng). The present government in Nigeria has promised to assist in financing the health care of its citizens. The first step towards achieving this pledge was the implementation of the National Health Insurance Scheme (NHIS) in 2005; a laudable project that appears to provide a solution to the issue of health care financing in Nigeria.

The NHIS is organized into the following social health insurance programs :Formal Sector; Urban Self-employed; Rural Community; Children Under-Five; Permanently Disabled Persons; Prison Inmates; Tertiary Institutions; School students; Voluntary Participants; Armed Forces, Police and other Uniformed Services (http//nhis.gov.ng) . It is only the formal sector SHIP that is currently operational in Nigeria (NHIS program on line).

The aim of the NHIS is to provide equitable access to health care for its citizens and at the same time providing financial protection. The intention is to remove users' fees, thereby removing the financial burden of 'out of pocket payment 'on families. Unemployed and poor people can benefit from the NHIS on special schemes; the government is now focusing on making the scheme mandatory for every Nigerian and aims to get every citizen enlisted by December 2015 (Agba et al., 2010). Funding of the NHIS comes from a combination of resources; employer and employees. Employers contribute 10% of the equivalent of the employee's annual salary while the employees contribute 5% of their annual salary to make a total of 15 % of the annual salary equivalent per employee. The Nigerian government has a

large workforce, unconfirmed sources estimates that about 70% of its citizens are government workers.

The Government pays its own contribution for its entire employer; in addition it has committed huge financial resources to ensure its success of the program. The benefit package is for employees and their spouse along with 4 biological children. Treatment offered under the NHIS is comprehensive and includes both out-patient and in- patient services to include hospital admission, maternity, eye, dental and pharmaceutical service and surgery.

The under 5 year old scheme was introduced by the government under the NHIS, its aim is to offer free treatment to all Nigerian children under the age of 5 years (www.nhis/org).

Cleft care funding may be considered under the 5 year old scheme; firstly because CL/P is a congenital condition affecting children in this category. Secondly most affected families are from poor background thus fitting in with one of the aims of the scheme and lastly because surgical services to include dental health care and education are available on this scheme.

Though other factors such as poor medical facilities, shortage of medical personal, lack of awareness, and poor funding have been identified as challenges that affect the efficacy of NHIS in Nigeria, nevertheless it is obvious the scheme has come to stay (Ibiwoye,2008; Sanusi,2009).

Table 2.2: Showing available funding for general health and cleft care in Nigeria

Types of funding	General health care	Cleft care
Health Insurance Scheme	Available	Not-available
Donor Funding	Available	Available
Out of pocket payment	Available	Available

2.10 CHAPTER SUMMARY

In conclusion, a narrative literature review on cleft lip and palate care has been presented in this chapter, the following points summarises the review.

- UCLP is the most prevalent of all types of clefts.
- Multidisciplinary treatment is the best approach to cleft care.

- There is a need for evidence based cleft care.
- The main form of cleft care in Nigeria is surgery
- Funding from cleft care in Nigeria, comes mainly from charity organisations,

CHAPTER 3 MATERIALS AND STUDY METHODOLOGY

3.1 INTRODUCTION

This chapter describes the materials and research methods used for the study. Two outcomes; 'standards of cleft care in Nigeria ' and 'treatment outcome in a selected Nigerian cleft population' were determined .

The chapter continues by discussing the study design, sample size, selection and data collection process.

3.2 STUDY DESIGN

The study design was retrospective and cross-sectional, two sets of data were collected; questionnaire response form cleft care coordinators and Dental models from 5 year old Nigeria children affected with CL/P. The questionnaires collected useful information about the current status of cleft care in Nigeria, Dental arch relationships were measured on Dental models to determine treatment outcome.

3.2.1 Questionnaire design

The aim of the questionnaire was to determine the standards of cleft care in Nigeria. The World Health Organisation established standards of cleft care previously (WHO,2002), hence the questionnaire was designed using the WHO standards of cleft care as a yardstick of measure. Three main domains in cleft care were covered; Health care needs; Service organisation and Practise guidelines. The questionnaire consisted of questions that covered these three domain of standards of cleft care as recommended by the World Health Organisation (WHO,2002) .

The informants were cleft care coordinators working in Nigeria hospital. Before administering the questionnaires, it was needful to identify hospitals offering cleft care services in Nigeria.

3.2.2 Recruitment

There are 53 identified cleft care centres in Nigeria with cleft care coordinators; only 40 out of the 53 cleft care coordinators participated in the questionnaire study. Data for treatment outcome was collected from only one of such centres coordinated by a surgeon, who allowed his patients to participate in the study. Firstly it was necessary to identify hospitals that offer cleft care in Nigeria, this was done using the internet google search tool with relevant search words. Presently all hospitals that offer cleft care in Nigeria are now in partnership

with the Smile Train organisation, so it was easier identifying the hospitals through the Smile Train website (www.smiletrain.org). Names of 53 hospitals that offer cleft care were eventually retrieved from their website, contact numbers and e mail addresses of coordinators were also retrieved. The geographical location of identified hospitals is shown in Table 3.1

Table 3.1: Geographical Location of Identified Cleft care centres in Nigeria.

Geopolitical zone	Number of cleft care centres that participated	Percentage
South West	12	30%
South South	2	5%
South East	9	22.5%
North West	2	5%
North East	6	20%
North Central	9	22.5%
Total	40	100%

3.2.3 Procedure to administer the questionnaire

The cleft team coordinators were then contacted by telephone or by e mail, and their willingness to participate in the study was sought. For those willing to participate, questionnaires were administered over the telephone and lasted for about 30 minutes. The questionnaire covered questions on the following aspects of cleft care; pre and post natal diagnosis, counselling; pre-surgical care; primary surgery; alveolar bone grafting; orthodontic/dental care; secondary surgery; speech therapy; otology; clinical genetics; psychological intervention and support groups (Appendix A). Responses were recorded and analysed. The reliability and validity of the questionnaire was tested by conducting an initial pilot survey with six cleft team coordinators; this allowed questions that were confusing or ambiguous to be reworded for better and clearer meaning. Eventually a questionnaire design that had been tested with clear meaning and wording was administered to collect useful data which was then analysed.

3.2.4 Data collection from 5 year old children

When conducting studies, the researcher must have forethought as to where and how the sample will be selected, this is to enable the findings of such research to be applicable to the intended population (Pelham and Balton,2006).The study population allowed a representative sample for the study to be selected.

3.2.5 Study population

The study population was the Federal teaching hospital, Gombe, Nigeria and was identified during the process of administering questionnaires to cleft care givers.The hospital is a specialist teaching hospital located in the North East Region of Nigeria and offers care to children affected with cleft lip and palate. The cleft care co-ordinator indicated his willingness to help with recruitment of patients as participants for this study.

Cleft care services in this centre began in 2000; very few children were seen initially in this hospital until the smile train came on board in 2007. However between the years 2000 to 2007, a total of 400 children affected with oro-facial cleft conditions have been seen at this hospital, this allowed for the selection of the sample size.

3.2.6 Sample size

Generally speaking research in the field of cleft has been plagued with the problem of sample size, it is important to note that in any given population, children with cleft lip and palate are relatively small in number. A practical way to calculate sample size is to base it on the prevalence of cleft lip and palate in the population. This was however not practicable because of the limitation of selection of sample to just one hospital. The question as to the ideal age at which to measure treatment outcome also arises, usually the impact of surgery on the soft and hard tissue in children with cleft lip and palate should be obvious after some months, though final treatment outcome can be measured after the age of 18 years when growth is complete. Measuring treatment outcome is however possible at any age, if the objectives and aims for measurement are stated clearly. Since previous studies have successfully reported outcomes in 5 year old children and have produced a reliable and valid yardstick, it seems reasonable to use such a yardstick for this study.

The study design allowed restrospective recall of patients that had previously undergone surgery at the hospital. Though the hospital had seen over 400 patients over a period of 10

years, the study was only interested in 5 year old affected children. Hence the sample size was purposive, all willing children and their parents were included in the study. Only 25 parents willingly gave consent for their children to participate in the study and were included in the study. Their impressions were taken and dental models fabricated afterwards. Overall 18 out of 25 dental models fabricated were then rated, this is because 7 models were of poor quality and did not show the required details.

3.2.7 Sampling technique

A cluster sampling of all children with cleft lip and palate that have received care at the designated hospital over a 5 year period (January 2008 to January 2013) was done to identify children that fell into the age selection criteria for the study. Their Case notes were retrieved, and relevant information to include patient's bio data, contact address and telephone numbers, type of cleft, side of cleft, type of operation were all recorded on a data extraction form (Appendix B). There were inclusion and exclusion criteria for the sample selcetion.

3.2.7.1 Inclusion criteria

The following inclusion criteria was considered for selection of the study sample.

- Only children born with complete unilateral cleft lip and palate (UCLP) born between January 2008 and December 2012.
- Children with a soft tissue band up to 5mm in width were included.
- Only children whom their parents gave voluntary consent were selected.

3.2.7.2 Exclusion criteria

- Children who present with other types of cleft than complete Unilateral Cleft lip and Palate (UCLP).
- Children presenting with a soft tissue band that is greater than 5mm.
- Children who present with a cleft as well as a syndrome.
- Children who present with cleft as well as learning difficulties and severe disability
- Children who did not have primary surgery performed in Nigeria.

3.2.7.3 Sample characteristics

Dental models of 18 (4 Females and 14 Males) Nigerian affected children out of the 25 originally collected were assessed to determine treatment outcome. These were mixed with a consecutive series of 37 European children (28 Males and 9 Females) retrieved from the Eurocran Good Practise Archive. A total of 55 Dental models were eventually rated. The characteristics of the Nigerian participants were retrieved from the patients case note and included Age, Gender and Side of cleft. Surgical repair of the lip was done at a mean age of 3.3 months using the Millard technique. Palatal repair was done at a mean age of 18.6 months using the von Langebeck technique. All surgeries were performed by only one surgeon. The Characteristics of the Nigerian and European samples are shown in Tables 3.2 & 3.3.

Table 3.2 Characteristics of the Nigerian sample

Model number	Side of Cleft	Gender	Age (Years and Months)
001	RC	Male	5.6
002	LC	Male	5. 4
003	RC	Male	5. 3
004	RC	Male	5 .2
005	LC	Female	5 .11
006	RS	Male	5.7
007	LS	Male	5 .7
008	RC	Male	5 .9
009	LC	Male	5 .9
010	RC	Male	5 .8
011	LS	Male	5. 11
012	RC	Male	5 .10
013	LC	Female	5 .7
014	RC	Female	5 .5
015	LC	Male	5 .3
016	LS	Male	5 .2
017	LC	Male	5 .6
018	RS	Female	5 .4
019	RS	Female	5.5
020	LS	Female	5.6
021	RC	Male	5.3
022	RC	Male	5.1
023	RS	Male	5.2
024	LS	Female	5.3
025	RC	Male	5.4

Table 3.3: Characteristics of the European sample

Model number	Side of Cleft	Gender	Age
1C	LS	Female	4.82
2C	RC	Female	6.01
3C	LC	Female	5.01
6C	RS	Male	4.96
7C	LS	Male	6.02
8C	RC	Female	6.02
10C	LS	Male	7.44
12C	LS	Male	5.10
14C	LC	Male	5.03
16C	RC	Male	5.70
19C	RC	Female	6.01
20C	LS	Male	6.03
21C	LC	Male	6.01
23C	LS	Female	6.23
24C	LS	Male	5.17
27C	LS	Male	5.06
28C	RS	Female	5.05
29C	LS	Male	6.01
33C	RS	Male	6.11
34C	LS	Male	4.92
35C	LC	Male	6.03
37C	LC	Female	6.03
43C	LC	Male	6.00
44C	RS	Male	6.06
45C	LC	Female	6.10
46C	LC	Male	6.01
48C	LC	Male	6.01
50C	LC	Male	5.93
52C	RS	Male	5.87
53C	LC	Male	6.01
61C	RC	Male	6.05
62C	RC	Male	5.99
63C	RC	Male	6.01
64C	RS	Female	6.01
65C	RC	Female	6.01
66C	LC	Male	6.02
67C	LC	Female	6.02

Table 3.4 Sample Characteristics (Gender &Age)

Nigerian Sample				European Sample			
Gender	**Number/Percentage**	**Mean Age**	**Range**	**Number**	**Percentage**	**Mean Age**	**Range**
Boys	**14(77%)**	**5.48**	**5.2-5.11**	**25**	**67.5%**	**5.81**	**4.92-7.44**
Girls	**4(23%)**	**5.42**	**5.4-5.11**	**12**	**32.5%**	**5.77**	**4.82-6.10**

Table 3.5 Sample Charactristics (Gender and side of cleft)

Nigerian Sample				European Sample			
Side of Cleft	**Gender**	**Number**	**Percentage**	**Side of Cleft**	**Gender**	**Number**	**Percentage**
Right	**Male**	**7**	**38.8%**	**Right**	**Male**	**8**	**21.6%**
	Female	**2**	**11.2%**		**Female**	**6**	**16.2%**
Left	**Male**	**7**	**38.8%**	**Left**	**Male**	**17**	**46%**
	Female	**2**	**11.2%**		**Female**	**6**	**16.2%**

Table 3.6 Treatment Protocol for the Nigerian and European affected Children

Treatment	Nigeria Sample	European Sample
PreSurgical Orthopaedics(PSO)	**No**	**No**
Lip repair	**Yes (Mean Age 3.1months)**	**Yes (Mean Age 3.3months)**
Palatal repair	**Yes (Mean Age 18.1 months)**	**Yes (Mean Age 17.2 months)**
Surgical technique Lip repair.	**Millard**	**Millard**
Surgical technique Palate repair	**von Langebeck**	**von Langebeck**

3.2.8 Procedure

Parents of children selected were contacted by telephone and informed about the study and their consent was sought for participation in the study. Those who consented were given a date to attend the clinic with their children. On the day of appointment, they were given a participant information sheet (PIS) that had detailed information about the study, in addition verbal explanation was also done. Parents signed the consent form on behalf of their children, since they were minor.

Every child who participated in the study was given an Identity number (ID) for the purpose of data collection. The dental models of the children were labelled with the identity numbers afterwards.

Data collection involved impression taking of the upper and lower jaws of each of the child to reproduce study models. All other relevant data, to include patient's details were recorded on a data extraction form. Impressions were taken with the aid of rubber based impression material loaded inside a paediatric impression tray. Prior to this, the child had been sited upright in a dental chair and was asked to open the mouth to allow for the impressions to be taken, thereafter, a squash bite of the child's occlusion was taken with a wax wafer, this was kept alongside the impressions taken for the purpose of correct articulation of the study casts . Poorly taken impressions were discarded; only impressions that showed good reproducibility of the dental arch details were retained. The impressions were then sent to a dental laboratory

in the United Kingdom, where an experienced dental technician fabricated study cast models. The study casts were then trimmed according to specifications, and were labelled in pairs (upper and lower jaw) with the child's ID number. Initially Dental models of 25 (5 year old) Nigerian children were reproduced of which 18 were eventually rated.

3.3 DATA ANALYSIS

Two sets of data were collected for this study; Questionnaires and Dental models.

3.3.1 Analysis of Questionnaires

The questionnaire were analysed as follow;

3.3.1.1 Data validation of questionnaires

Data validation ensures questionnaires are completed and data is consistent, it is best to avoid unclear questions that would not be answered by most respondents this prevents bias in analysis. The mode for data collection for this study was over the telephone, so the PI had to read out the questions over the telephone, responses recorded was in multiple choice format which made it easy for analysis. There were very few unanswered questions. Responses were then partitioned into themes surrounding cleft care.

3.3.1.2 Response partitioning

Homogenous partitioning of responses allowed analysis to be easier and faster, for example demographic data were sub grouped into male and female to allow for better comparison.

3.3.1.3 Data coding

Data coding allows responses to be coded, simply put, it allows conversion of nominal to ordinal scale data so that it can be analysed statistically. Grouping together of similar responses are then analysed. Responses from participants were then coded into several subgroups of cleft care, such as Diagnosis, Record, Comprehensive cleft care; Treatment protocol this allowed findings on current status of cleft care in Nigeria to be reported.

3.4 ANALYSIS OF DENTAL CASTS

The Dental models fabricated were trimmed according to specifications, and labelled in pairs (upper and lower jaw) with the child's ID number. Prior to this, the study casts were

inspected carefully by the Principal Investigator (PI) and some were discarded prior to rating due to loss of history details of the patients and poor impression taking technique so that some details were poorly represented. Eventually only 18 out of the 25 study casts were of good quality, only these were assessed. The remaining 7 dental models were discarded.

The dental models form the Nigerian sample was mixed with a consecutive series of 37 models from the Eurocran Good Practise Archive which were matched for age and gender .This allowed for comparison between the Nigerian and European samples. A total of 55 models were eventually assessed by two examiners (BS) and (AA).Dental arch relationships was measured using two indices; 5 year old index and the modified Huddart/Bodenham, this was used to predict postsurgical treatment outcome. Tne use of the two indices are further described;

3.4.1 The 5 year old index

The 5 year old index was developed by Attack and colleagues with the aim of assessing dental arch relationship in children born with UCLP. It is a standardized, valid and reliable method that can be used to report surgical outcomes in 5 year old children (Attack et al., 1997).The yardstick is graded on a five point scale 1-5, (from excellent to very poor) and considers the anterio-posterior, transverse and vertical aspects of occlusion. To score appropriately, the yardstick uses a set of reference models and scores are awarded by comparing the study models under investigation with a reference set of models consisting of each of the five grades.

Study casts for this study was scored by two orthodontists, one of them (BS) an expert in scoring with several years of experience in cleft care, and the other was the principal investigator (AA) a researcher, who had no experience in scoring. The models scored had been previously labeled with the children's ID number; they were then randomized and re-labeled with new numbers to allow for anonymity and to avoid bias during scoring. Fifty five models (18(Nigerian) and 37(European) models of 5 year old children with UCLP were scored by the specialists on two separate occasions and at an interval of one week apart this was to ensure inter and intra rater reliability and minimize the influence of memory loss on results.

Table 3.7: The 5 year old index (Attack et al., 1997)

Group	General studies	Outcome
1	Positive overjet with average inclined or retroclined incisor. No crossbites/openbites; Good maxillary shape and palatal vault anatomy	Excellent
2	Positive overjet with average inclined or proclincd incisors Unilateral crosshite or crossbite tendency +/- Open bite tendency around cleft site	Good
3	Edge-to-edge bite with average incline or proclincd incisors: or reverse overjet with retroclined incisors Unilateral crossbite,+/- Open bite tendency at cleft site.	Fair
4	Reverse overjet with average inclined or proclined incisors Unilateral crossbite +/- bilateral crossbite.	poor
5	Bilateral crossbite, Poor maxillary arch form and palatal vault.	Very poor

3.4.2 Modified Huddart Bondeham MH/B scoring system

The Huddart and Bondeham was originally designed as a scale to evaluate arch form, and intended for use in the primary dentition (Huddart and Bodeham, 1972). The scale was later modified for use in the mixed dentition; the Modified Huddart Bondeham MH/B scoring system (Mosey and Gray, 2005). The MH/B system requires that the relationship of all upper to the lower teeth from the first permanent molar forward be given a score to reflect maxillary arch constriction.

Rules were also drawn up for some circumstances; for example when one central incisor was missing the other central incisor was scored. When a canine was unerupted, its score was determined by the mid-point of the maxillary alveolar ridge. Where a premolar was absent due either to non-eruption or hypodontia, a score was allocated equivalent to the adjacent

premolar, if erupted. If no premolars were erupted, the same rule as for the canine was applied, i.e. the score was determined by the mid-point of the maxillary alveolar ridge.

The laterals are not scored, as well as the permanent molar before the age of 6 years (maximum range scores;-18 to+2). After 6 years of age, first permanent molars are scored; (Maximum range scores -22 to +2).The same specialists (BS) and (AA) scored the models using the MH/B system, to ensure inter and intra examiner ratter reliability scoring was done twice at an interval of one week apart.

Scores determined from both the 5 year old and the MH/B scoring system were analysed, inter and intra examiner ratings were evaluated using the weighted kappa (Fleiss,1981). Kappa values were rated as follows; values<0.20 (poor); 0.21-0.40 (fair); 0.41-0.60 (good); 0.81-1.00 (very good) and 1.00 (perfect agreement). For the 5year old index mean scores were used to assess the quality of treatment. The results are presented in chapter 4.

CHAPTER 4 RESULTS

This chapter presents the results of the thesis; two outcomes were determined; treatment outcome in a selected 5 year old Nigerian cleft population and current status of Cleft Care in Nigeria.

4.1 TREATMENT OUTCOME

Treatment outcome was determined by measuring post-surgical dental arch relationships in 5 year old Nigerian children affected with CL/P. Two indices were used ; the 5 year old index and the modified Huddart/Bodenham scale. The results are presented in Tables 4.1- 4.9 . The overall mean scores for the Nigerian sample was 2.3 and that of the European sample was 2.9 (Table 4.1). The percentage distribution of scores is shown in Table 4.3-4.7, the Nigerian sample showed that 50% of cases were in groups 1 and 2 (good quality), and 50% of cases were in group3 (fair quality). The statistical analysis of data is shown in Tables 4.8-4.11.

Table4.1 : Mean scores of dental models assessed

Scores	**Overall Mean Scores (5 year old index)**
Nigerian cleft population	**2.3**
European cleft population	**2.9**

Table 4.2 : Percentage distribution of scores (5 year old index)

Centre A (Nigerian sample)

Grading	**No of Models**	**Percentage**
Score 1-2	**9**	**50**
Score 3	**9**	**50**
Score 4- 5	**0**	**0**

Centre B (European sample)

Grading	**No of Models**	**Percentage**
Score 1-2	**10**	**27%**
Score 3	**22**	**59%**
Score 4- 5	**5**	**14%**

Table 4.3 : Over **all distribution of scores**

Centres A & B	
Score 1-2	19
Score 3	31
Score 4&5	5

Table 4.4 Overall percentage of Poor Scores

Centres	(Scores 4 &5)	
Centre A	0%	
Centre B	9%	

Table4.5: Showing comparison of Single Centre poor scores with the mean 5 year old index score in Centres A&B

Centres	**Percentage of poor scores**	**Mean scores**
Centre A	**0%**	**2.3**
Centre B	**14%**	**2.9**

Table 4.6: Showing two sample t test with equal variances

Group interval	Obs	Mean	Std. Err	Std.Ved	(95% Conf.
1	18(Nigerian)	2.333333	2061156	8744746	1.898467
2.768199		2.925676	1710748	1.040608	2.57872
2	37(European)				
3.272632					
(Combined) - 3.007686	55(Combined)	2.731818	1375982	1.020456	2.45595
Diff 0215037					

Table 4.7: Showing two sample Wilcoxon rank-sum (Mann-Whitney) test

Source	Obs	Rank sum	Expected
1	18(Nigerian)	392	504
2	37(European)	1148	1036
Combined	55(Combined)	1540	1540

P=0.0 Significant differences exists between Nigerian and European sample.

Table 4.8 : Showing Mean Scores- H/B System.

Tooth	Cleft Side	Non Cleft Side	P value
Central Incisors	-0.80	-0.51	.000(Significant)
Canines	-1.07	-0.53	..309
1st molar	-.84	-.44	.208
2nd Molar	-1.09	-0.93	.000(Significant)

Table 4.9: Showing frequency of scores -H/B System

Tooth	Cleft Side	Percentage %	Non Cleft Side	Percentage %
Central Incisors -3	13	23.6	3	5.5
-2	5	9.1	6	10.4
-1	13	23.6	9	16.4
0	22	40.0	36	65.5
2	2	3.6	1	1.8
Canines -2	37	67.3	10	18.2
-1	6	10.9	9	16.4
0	12	21.8	36	65.5
1st molar -2	24	43.6	2	3.8
-1	10	18.2	20	36.4
0	21	38.2	33	60.0
2nd Molar -2	14	25.5	3	5.5
-1	11	20.0	13	23.6
0	30	54.5	38	69.1

4.10 **Study Model Ratings : Raw Scores (Huddart Bodenham)- Rater 1**

CLEFT SIDE					NON-CLEFT SIDE			
Model	2nd Molar	1st Molar	Canine	Central Incisors	Central Incisors	Canine	1st Molar	2nd Molar
1	-1	-1	-2	0	0	0	0	0
2	-1	-1	-2	0	0	0	0	0
3	-1	0	0	0	0	0	0	0
4	-1	-1	-2	0	0	0	0	0
5	-1	-1	-2	0	0	0	0	0
6	0	0	0	0	0	0	0	0
7	-1	-2	-2	-2	0	0	0	0
8	-2	-2	-2	-2	-2	0	0	0
9	-1	-1	-1	0	-2	0	0	0
10	0	0	0	0	0	0	0	0
11	0	0	-2	0	0	0	0	0
12	0	0	0	-1	0	0	0	-1
13	-1	0	0	0	-1	0	0	-1
14	-2	-2	-2	0	0	0	0	0
15	-2	-1	-2	0	0	0	0	0
16	-2	-2	-2	0	0	0	0	0
17	-2	-2	-2	-3	-3	-2	-1	-1
18	-1	-1	-1	0	0	0	0	0
19	0	0	-2	0	0	0	0	0
20	-1	-1	-2	0	0	0	0	0
21	0	-1	-1	-1	-1	0	0	0
22	-2	-2	-2	0	0	0	0	0
23	-2	-2	-2	0	0	-1	-1	-1
24	-1	-2	-2	-3	-3	-1	-1	-1
25	-1	-1	-1	0	0	-1	-1	-1
26	0	-2	-2	0	0	0	0	0
27	-1	-2	-1	-2	-2	0	0	0
28	-2	-1	-2	-1	-1	0	0	0
29	-1	-1	-2	-2	-2	-1	-1	0
30	-1	0	-2	0	0	0	0	0
31	0	-2	0	0	0	0	0	0
32	-1	-1	-2	0	0	0	0	0
33	-1	0	-1	-2	-2	-2	-2	-2
34	0	-2	-1	0	0	0	0	0
35	-2	-1	-2	-3	-3	-1	-1	-1
36	-1	0	-2	0	0	0	0	0
37	-1	0	-2	-2	-2	-2	-2	-2
38	-2	-1	0	0	0	0	0	0
39	-2	-1	0	0	0	0	-2	-2
40	-1	0	-1	0	0	-1	0	0
41	0	0	-1	0	0	0	0	0
42	0	-1	-1	0	0	0	0	0
43	-1	-1	-1	0	0	0	0	0
44	-1	-1	-2	0	0-	0	0	0
45	-1	-1	-2	0	0	0	0	0
46	0	0	0	0	0	-1	-1	-1
47	-1	-2	0	0	0	-1	-1	-1
48	-1	-2	-2	-1	-1	0	0	0
49	-2	-2	-2	-1	0	0	0	0
50	0	0	0	0	-3	-2	-2	-2
51	0	-1	-2	-3	-3	-3	-2	-2
52	-1	-1	0	0	0	0	0	0
53	-1	-1	0	0	0	-1	-2	-1
54	0	0	-1	-1	-1	0	0	0
55	0	0	0	0	0	0	0	0

Table 4.11 ; Showing Study Model Ratings : Raw Scores (Huddart Bodenham)-Rater 2

CLEFT SIDE					NON-CLEFT SIDE			
Model	2nd Molar	1st Molar	Canine	Central Incisors	Central Incisors	Canine	1st Molar	2nd Molar
1	-1	-1	-1	0	0	0	0	0
2	0	-1	-1	0	0	0	0	0
3	0	0	0	0	0	0	0	0
4	0	0	-2	0	0	0	0	0
5	-0	0	-2	0	0	0	0	0
6	0	0	0	0	0	0	0	0
7	0	0	-1	-2	-2	0	0	-1
8	-1	-1	-2	-2	-2	-1	-1	-1
9	-1	-1	-2	0	-2	0	0	0
10	0	0	0	0	0	0	0	0
11	0	0	-2	0	0	0	0	0
12	-1	-1	-1	-1	0	0	0	-1
13	0	0	0	0	-1	0	0	-1
14	-2	-2	-2	0	0	0	0	0
15	-1	-1	-2	0	0	0	0	0
16	-2	-2	-1	0	0	0	0	0
17	-1	-2	-2	-3	-3	-2	-1	-1
18	0	0	0	0	0	0	0	0
19	0	0	-2	0	0	0	0	0
20	0	-1	-2	0	0	0	0	0
21	0	-1	-1	-1	-1	0	0	0
22	-1	-2	0	0	0	0	0	0
23	-2	-2	-2	-1	0	0	0	0
24	0	-2	-2	-3	-3	-1	-1	-1
25	-1	-1	-1	0	0	-1	-1	-1
26	0	-1	-1	0	0	0	0	0
27	-1	-2	-1	-2	-2	0	0	0
28	0	-1	-2	-1	-1	0	0	0
29	-1	-1	-2	-2	-2	-1	-1	-1
30	0	0	0	0	0	-2	-1	-1
31	0	-2	0	0	0	0	0	0
32	-1	-1	-2	0	0	0	0	0
33	0	0	-1	-2	-2	-2	-2	-2
34	0	-2	-1	0	0	0	0	0
35	-2	-1	-2	-3	-3	-1	-1	-1
36	-2	0	-2	0	0	0	0	0
37	-2	0	-2	-2	-2	-2	-2	-2
38	-1	-1	0	0	0	0	0	0
39	-2	-1	0	0	0	0	-2	-2
40	0	0	0	0	0	-1	0	0
41	0	0	-1	0	0	0	0	0
42	0	-1	-1	0	0	0	0	0
43	-2	-1	-2	0	0	0	0	0
44	-1	-2	-2	0	0-	0	0	0
45	-1	-2	-2	0	0	0	0	0
46	0	0	0	0	-1	-1	0	0
47	-2	-1	0	0	0	0	0	0
48	-1	-1	-1	-2	-2	0	0	0
49	0	0	0	0	0	-2	0	0
50	0	0	0	0	-2	-2	-1	-1
51	-2	-2	-2	-3	-3	-2	-1	-0
52	-2	0	0	0	0	0	-1	-1
53	-1	-1	0	0	0	0	-1	-1
54	-1	-1	0	0	0	-1	-1	0
55	0	0	-0	0	0	0	0	0

Table 4.12: Showing the level of agreement between raters (Kappa statistics):1st session.

Examiner	Indices	Weighted Kappa	Agreement
BS vs AA	5 years old index	0.53	Moderate
BS vs AA	MH/B index	0.85	Very good

Table 4.13: Showing the level of agreement between raters (Kappa statistics):2nd session.

Examiner	Weighted Kappa	Agreement	
BS vs AA	5 year old index	0.85	Very good
BS vs AA	MH/B index	0.60	Moderate

Figure 4.1:Showing the study models of 5 year old children wirh UCLP that were rated

Figure 4.2: Modelrating session: showing the two raters (AA) and (BS) .

Figure 4.3: Chart showing the mean scores of the Nigerian and European sample

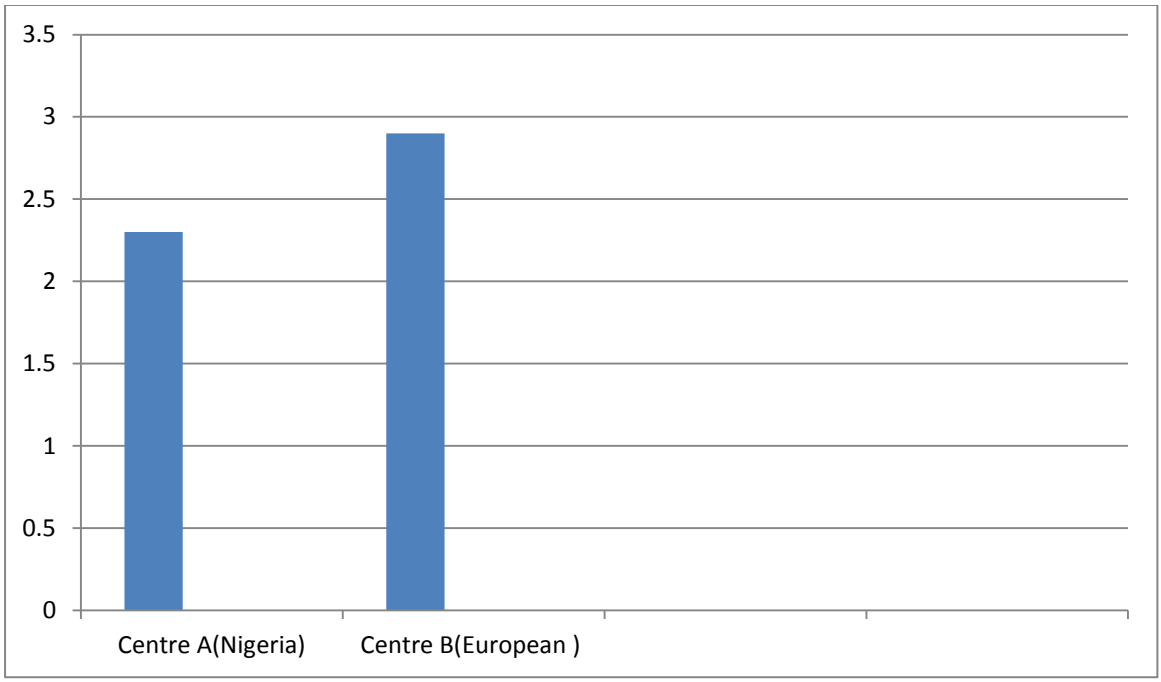
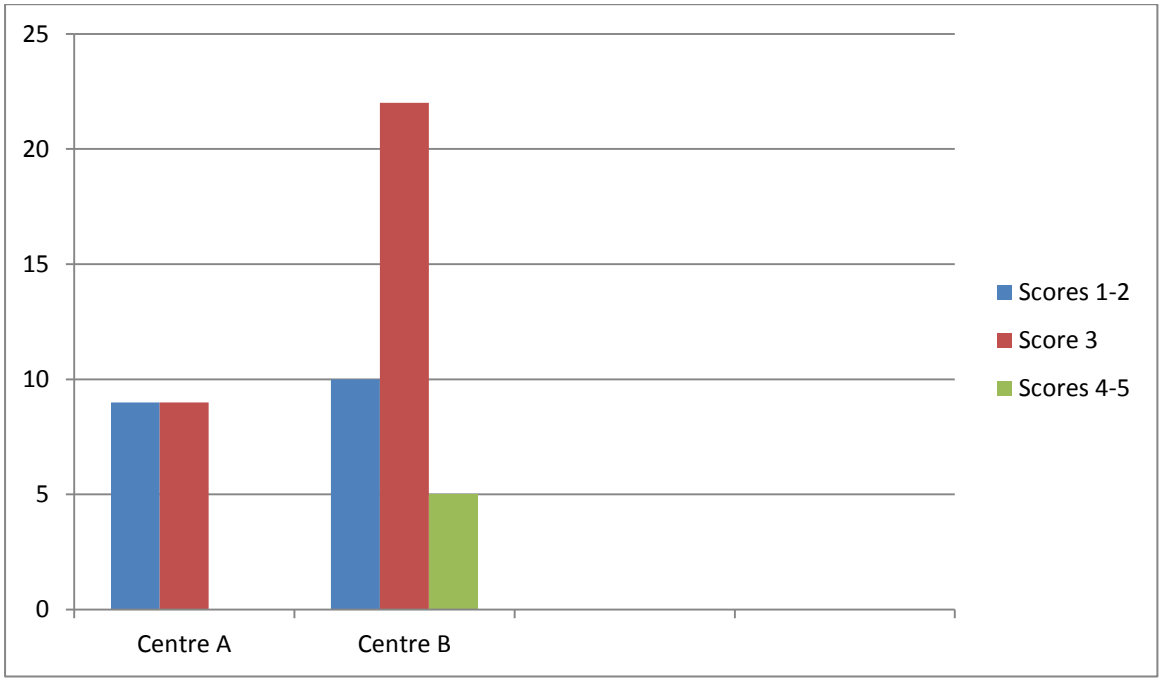


Figure 4.4: Chart showing the percentage distribution of scores between Centre A &B



4.2 STANDARDS OF CLEFT CARE

Out of the 53 hospitals identified, only 40 (75%) cleft care coordinators participated in the survey. The questionnaires examined three domain pertaining to cleft care; Organisation of services; Practise guidelines and Health care needs. The results are presented;

4.2.1 ORGANISATION OF SERVICES.

4.2.1.1 Cleft care services available and offered in Nigeria

Fifty three (53) hospitals offer cleft care services in Nigeria, all are located in either major cities or towns. Majority (n=46; 86.7%) of the hospitals are government owned while very few (n=7; 13.3%) are privately owned (Table 4.1). Forty six (n=46; 86.7%) of these hospitals began cleft care between 2006-2010. Cleft care services in all hospitals (n=40; 100%) that participated in the study are provided independently by their hospitals, there are no regional cleft care centres. Majority (92.5%) of the Cleft care specialists are surgeons either (Oral and maxillo facial or plastic surgeons) and act as co-ordinators of services offered.

The main form of treatment available for children with CL/P in Nigeria is primary surgical repair of the lip and/or palate (n=40; 100%)..Multidisciplinary care in the form of surgery, orthodontics and otology care are offered in some Nigerian hospitals (n=6:15%), speech therapy is rarely done.

Table 4.14 Showing the main providers of cleft care services in Nigeria

Type of provider	Number of hospitals	Cleft specialists
Government	**46(87.7%)**	**Surgeons: OMS and plastic (37:92.5%)** **Orthodontists 2(10%)** **ENT surgeons 1(2.5%)**
Private	**7(13.3%)**	**Surgeons mainly (100%)**

Table 4.15 Showing the WHO recommendation for Health Care Needs for affected children affected and the Current Status of Cleft care in Nigeria.

Health care Needs	WHO Standards	Current status in Nigeria
Pre natal diagnosis/ Neonatal Emotional support and Professional Advice	Prenatal diagnosis recommended and emotional support and professional advice to be given by a cleft specialists.	Currently not offered, professional advice available only after birth.
Neonatal Nursing	Specialist's advice on feeding should be given by nurses.	Specialist's advice on feeding given by doctors and in few hospitals

4.2.2 – PRACTISE GUIDELINES- The WHO recommendations and the current status in Nigeria is shown in Table 4.12

Table 4.12: Showing the WHO recommendations of practise guidelines for cleft care and the current status in Nigeria.

Type of services	**Eurocleft reccomendations**	**Current status of cleft care in Nigeria**
Multidisciplinary care	Multidisciplinary care should be practised and is recommended as the best form of care.	Multidisciplinary care is rarely done,the main form of treatment available for children with CL/P in Nigeria is primary surgical repair
Training	There should be specialist training posts for cleft care specialists.	No such training post for cleft specialists but block posting in teaching hospitals that offer cleft care services (n=8;20%).
Audit and Research	Recommendation of 40-50 cases minimum annually, regular consumer audit, multicenter research .	Most hospitals see over 50 children annually, but do not subject themselves to regular consumer audit, few multicenter research.
Support Group/ Outreaches/Follow up	Local Parent Support Group (PSG) .Outreaches and campaign should be done to educate patients and patients should be regularly followed up.	No local Parent Support Group (PSG) for cleft teams in Nigeria. Outreaches and campaign are done for the purpose of health education and recruitment. Patients are not regularly followed up.
Finances	Resources or should be made available for all aspects of cleft acre, to include travel expenses and general dental care.	The only source of funding is from Smile Train and available only for primary surgical repair of cleft of the lip and /or palate.
Record taking	Reccomended records; Studycasts, cephalometric x-rays, photographs, speech .audiometry and patient/parent satisfaction.	Pre and post-treatment photographs;. Detailed clinical notes to include treatment protocol and timing as well as procedures undertaken .
Surgery	Primary surgical repair should be performed by an experienced qualified surgeon and agreed by a team; secondary corrective surgical procedures may be needed later.	Primary surgical repair is the main form of treatment performed by a qualified surgeon not necessarily agreed by a team, (N=40; 80%) Secondary corrective surgical procedures are rarely done
Orthodontics	Orthodontic services should be available when necessary and performed by an experienced orthodontist.	Orthodontic services available in very few hospitals and on fee for service basis. (N=6; 15%)
Speech and Language Therapy	Early assessment of speech and language problems with availability of corrective therapy by an experienced speech and language therapist	Assessment of speech and language problems
Ear Nose and Throat (ENT)	ENT problems should be identified at an early stage and necessary therapy provided.	ENT services provided in (N= 2; 5%) hospitals.
Clinical Genetics/	Early assessment of anomalies	Not available

Paediatric Developmental Medicine	Genetic Counselling for parents and families should then be made available.	
Dental Care	Children with CL/P should receive regular dental care and advice.	Dental care is not routinely offered, but available in few hospitals (n=8;20%) on a fee for service basis
National Register	There should be a national register for accurate recording of birth of children born with cleft conditions and other craniofacial anomalies	There is no national register for children with cleft and other congenital deformities

4.2.2.1 Prenatal diagnosis

Presently, none of the Nigerian hospitals offer prenatal diagnosis of CL/P condition. Ultrasound is not taken for pregnant women in Nigeria to detect genetic or congenital abnormalities such as cleft lip and palate.

4.2.2.2 Neonatal nursing

Neonatal nursing is offered in only very few hospitals (n=6; 15%), majority of the hospitals (n=34; 85%) do not have nurses on their cleft team, but employ their services on the ward to administer post-surgical care.

4.2.2.3 Training

There are no specialist training posts specific for cleft care specialists in Nigeria, rather doctors training to become specialists in surgery are mandated to undergo 3 months of block posting in cleft care. During the 3 month period, trainees are taught, present seminars and acquire surgical skills by assisting during operation of children with CL/P. Eight Teaching hospitals (n=8;20%) were recorded to offer such training posts. Further experience and training in cleft care is said to be acquired during elective postings overseas. The highest qualification of a cleft specialist in Nigeria recorded is the Fellowship of the Postgraduate College of Medical and Dental sciences (Nigeria/West-Africa Postgraduate Medical College).

4.2.2.4 Audit and research

Very few hospitals in Nigeria (n=6; 15%), see over 50 children affected with CL/P on annual basis and do not subject themselves to regular consumer audit and are rarely involved in multicentre research.

4.2.2.5 Support Group/ Outreaches/Follow up

Currently there is no local Parent Support Group (PSG) for cleft teams in Nigeria, nutritional advice is given by coordinators in majority of the hospitals (n=38;95%). Majority of the Hospitals in Nigeria (n=38;95%) now organise outreach programmes, health education and awareness about cleft conditions and treatment are publicised to the community. Recruitment of cleft patients is largely done during such outreach program. In Nigeria, follow up of cleft care patients in all hospitals (n=40;100%) is on short term basis, up to one month, there is no long term follow up, affected children are hardly recalled years later to assess treatment outcome.

4,2.2.6 Finances

Presently in Nigeria 'Donor funding 'is the only source of finance for cleft care and is mainly from charity organisations. Resources are made available only for primary surgical repair and is available in all of the Nigerian hospitals (n=40; 75%).

4.2.2.7 Record taking

In Nigeria, as of now only pre and post-surgical photographs are taken for children with CL/P are taken, Detailed clinical notes to include treatment protocol and timing as well as procedures undertaken are also available in all hospitals (n=40;100%).. Other records such as study models, Cephalometric x-rays, speech, audiometry and patient satisfaction are rarely taken.

4.2.2.8 Surgery

Surgical repair is performed by qualified surgeons that are also cleft specialists in majority of the hospitals (n=35;87.5%). Surgery is offered free for all affected children from poor background and sponsored by one main charity organisation. Surgical repair of the lip is done for affected children, 3-4 months after birth in majority of the hospitals (n=32; 60%), though two hospitals (n=2; 3.7%) reported surgical repair of the lip in babies less than 3 months old. Palatal repair is done at 9 months in (n=15; 28.3%) hospitals and at 18 months in (n=38; 71.7%) hospitals. The rule of '10s' is ensured before primary surgery is performed (i.e. the child must be 10 weeks old with haemoglobin level of 10mg/dl and 10 lbs. weight). The most common surgical technique employed by Nigerian surgeons for lip repair is the rotation-advancement technique (n=39; 97.5%) and straight line for BCLP (n=40;100%).The most

common surgical technique for palatal repair are von Lange beck technique (n=38; 95%) and palatal push back (n=40; 100%).

4.2.2.9 Orthodontics

There are few hospitals (n=6;15%) who offer orthodontic services for children with CLP in Nigerian hospitals. Orthodontic services are available on a fee for service basis. Orthodontic services offered include pre-surgical orthopaedics and fixed appliance treatment; Orthodontists don't give regular feedback to surgeons on growth related disturbances.

4.2.2.10 Speech and Language Therapy

Presently, none of the Nigerian hospitals (n=40;100%) are able to provide early diagnostic assessment for speech problems for children with CLP. This is because there is shortage of speech and language therapists as well as diagnostic equipment in the country.

4.2.2.11 Ear Nose and Throat (ENT**)**

There are very few hospitals (n=2;5%) that provide audiology services for children with CLP in Nigeria. Early diagnostic and counselling assessments in otology are not done routinely for children with CLP in Nigeria.

4.2.2.12 Clinical Genetics/ Paediatric Developmental Medicine

Genetic counselling is not offered in any of the Nigerian hospitals (n=40; 100%), and there is no follow up on growth related problems for children with CL/P in Nigeria, they are not referred to paediatricians to assess their growth.

4.2.2.13 Dental Care

Dental services are available for children affected with CL/P in very few hospitals (n=6;15%). Affected children do not receive dental health education and are rarely referred for dental consultation, prevention against dental caries is not done routinely and fluoride supplements are not given routinely.

CHAPTER 5 : DISCUSSION

5.1 BACKGROUND

The purpose of this study was to report on treatment outcome in Nigerian children affected with CL/P and who had surgical repair done previously. In reporting outcome, it was also necessary to report on the current status of cleft care as offered in Nigerian hospitals. Treatment outcome has been determined using two validated indices; the 5 year old index (Attack et al., 1997) and the Modified Huddart/Bodenham system. While measurable standards for cleft care services has been used to determine standards of cleft care in Nigeria (Shaw et al., 2001; WHO, 2002; 2003). The results have been presented in chapter 4, this chapter will discuss on relevant findings.

5.2 TREATMENT OUTCOME IN NIGERIAN CHILDREN AFFECTED WITH CL/P.

Dental arch relationships of 5 year old children born with complete unilateral cleft lip and palate (UCLP) were assessed and measured to determine treatment outcome, Of all the cleft subtypes, UCLP is said to be the most homogenous and occurs with sufficient frequency that reasonably sized samples can be gathered (Shaw et al., 1992; Bearn et al., 2001; Shaw et al., 2015). It also represents a group of cleft disorders that require all the skills of a multidisciplinary team to achieve excellent outcome.

One of the main aims of treatment for these group of children is the achievement of a near normal growth and development of the maxilla and surrounding structures as possible. The scarring that results from the primary surgical repair of the cleft may have a detrimental effect on maxillary growth, it has been suggested that delaying the surgical repair of the palate reduces the adverse effect on maxillary growth. However, delaying the repair of the palate may result in adverse effects on speech development (Bardach et al., 1984). Therefore when assessing the clinical outcome, the question of maxillary development and speech are two key areas of concern. Assessment of dental arch relationship is one way of assessing maxillary development from study models. Dental study models provide a precise and durable record of the palate, teeth, dental arches and the occlusion. They are routinely collected in orthodontics as part of the treatment record, and similarly can be collected in children with cleft lip and palate at key treatment stages.

The study design was cross-sectional, sample was taken from a cleft population, purposely selected from one major cleft care centre in Nigeria. The advantages of this centre as a study

population was that it allowed sampling of patients from 3 geo-political zones in the North of Nigeria; (North -East, North-North and North-South) giving a good representation of affected children from the Northern part of the country. Further, it allowed a diverse sample of participants from different background to be selected, this is because patients from both secondary and primary health care centres are seen in this centre.

There has been much literature on treatment outcome in children with UCLP, and much data has been published, many of such studies have been studies conducted in other countries such as Europe (Shaw et al., 1992; Mars et al., 1992; Attack et al., 1997, Mars et al., 2006, Williams et al., 2001, Bearn et al., 2001; Sandy et al., 2001; Semb,2011),Sweden (Brattstrom, 1991;Lilja et al., 2006), Scandinavia (Fride et al.,1991), America (Trotman et al., 1996, Flinn et al.,2006;Long et al., 2011), Germany (Swennen et al., 2002; Noveraz et al., 2014) Japan (Susami et al., 2006;Ozawa et al., 2011;) and India (Rengit, 2009).

This study for the first time, reports dento-facial treatment outcome in a Nigerian cleft population, the results shows that treatment outcome is of good quality (2.3). The study was conducted in line with the Helsinki's declaration (World Medical Association, 2013). The chapter continues by using relevant themes for discussion ;

5.2.1 Sample size

.One of the limitations of the study, was the small sample size. The use of small sample size in cleft research have been reported as a limitation because of the low incidence of CL/P in any population (Adeyemi and Akintububo, 2015). Sample size in studies may range from one participant for example in case presentation to much higher numbers of hundreds in collaborative studies .Previous studies that has assessed dental arch relationships in children with UCLP used a range of sample size from between 24 -136 study casts (Brattstrom, 1991,Fride et al.,1991;Shaw et al., 1992; Mars et al., 1992; Trotman et al., 1996,Attack et al., 1997; Williams et al., 2001, Bearn et al., 2001; Sandy et al., 2001; Suzuki et al., 2007; Long et al., 2011; Ozawa et al., 2011;koszelj etal.,2012;Dogan et al.,2014).

5.2.2 Determining outcome at the age of 5 years

The advantage of measuring outcome at the age of 5 years is that it serves as early predictor of treatment outcome, it is timely and relevant and provide a means to reduce the length of research studies without increasing the sample size (Roberts et al., 1991). Further the development of early markers of treatment outcome is relevant, especially in countries where

standards are perceived to be below the best European standards of cleft care (Shaw et al., 2015). Though the impact of surgery on the soft and hard tissue in children with cleft lip and palate is said to be obvious months after treatment, it is best to measure treatment outcome after age of 18 years, usually when growth is complete (Semb, 1991; Semb, 2013). Most studies have measured treatment outcome at between 10-12 years, though measurement of treatment outcome is possible at any age, once the aims and objectives are clearly stated. Since previous studies (Attack et al., 1997; Mars et al., 1992; Suzuki et al., 2007; Long et al., 2011; Ozawa et al., 2011; Britton et al., 2014; Heliovaara et al., 2013; Karsten et al., 2013; Mostled et al., 2013) have successfully reported outcomes in 5 year old children and have confirmed the use of a reliable and valid yardstick, it seems reasonable to use the same age group for this study.

The problem is that small caseloads by an individual surgeon make statistical analysis meaningless. It has been suggested that larger centres could act as reference norm against which smaller centres could measure their success (Shaw et al., 1992).

5.2.3 Dental Study Models

Fifty-five (18 Nigerian and 37 European) dental models of children affected with CL/P were assessed and used to report treatment outcome. The use of dental models can be justified, since they are known to be exact replica of the jaws and can give accurate measurements (Hunter and Priest, 1960). Further, taking of impressions to reproduce dental models are minimally invasive procedures that can be tolerated by young children. Many studies have successfully used dental models to determine dental arch relationships (Shaw et al., 1992; Mars et al., 1992; Attack et al., 1997; Williams et al., 2001, Bearn et al., 2001; Sandy et al., 2005; Mars et al, 2006; Susami et al., 2006; Long et al., 2011; Ozawa et al., 2011; koszelj etal., 2012; Dogan et al., 2014). Dental arch relationships are said to be reliable indicators of facial growth and predictors of treatment outcome (Harthorn et al., 1990; Ozawa et al., 2011; koszelj et al., 2012; Noveraz et al., 2014)).There are other ways of determining dental arch relationships apart from using dental models, such as using cephalometic x-rays and photographs (Shaw et al., 1992; Mostled et al., 1992; Asher Mc-Dade et al., 1992;Daskalogiannakis et al., 2011;Bartzela et al., 2012).Though use of cephalometric x-rays has been reported to be limited in patients with abnormalities such as cleft, because some land marks may be difficult to identify because of the distortion of the maxillary structures (Mostled et al., 1992). While the use of photographs may produce error especially if they are

not well taken (Ash Mcdade et al., 1992). Dental models are scored using validated indices to generate mean scores.

5.2.4 Quality of surgery.

Children affected with CL/P undergo surgical repair early in life, the quality of surgical repair have been known to determine treatment outcome (Shaw et al.,1992). The average mean score is a measure of scores distribution and for the Nigerian sample was 2.3, indicating that Nigerian children affected with CL/P receive good quality surgery, this may be dependent on the surgical technique, treatment protocol and the skill of the surgeon. The European sample had a mean score of 2.9. The surgical protocol and timing for both the European and Nigerian study models also compared favourably as both cleft population had lip and palate repair done at 3 and 18 months respectively and both used the Millard and von langenbeck's surgical technique for lip and palate repair respectively. The differences in mean scores between the Nigerian and European samples may be due to ethnic differences. Nigerians are said to have a bi-maxillary profile with a tendency towards class II malocclusion and increased overjet (Isiekwe et al., 2014). It is possible that the Nigerian children had a measure of overjet that would reduce the influence of maxillary growth disturbance. Measurement of over jet serves as a good predictor of treatment outcome (Bearn et al., 2001).

The cleft surgery was performed by one surgeon, using the Millard technique for lip repair and von-Langebeck for the palatal repair. The results of this study indicates that the current surgical protocol, timing and technique for cleft lip and palate repair in the selected Nigerian is good. All surgeries was performed by one surgeon, which was of great advantage, the surgical skill of the operator is believed to influence the post -surgical quality of treatment and is a significant predictor of treatment outcome (Delvin, 1990; Bardach et al., 1990; Pauline and Thilander 1991, Ross, 1990; Shaw et al., 1992). The good skills exhibited by the surgeon may be directly related to years of experience and the volume of care received at the hospital, in the main stream of comptemporary cleft surgery. To validate the findings of this study and its applicability to other cleft outcome studies, it was necessary to compare findings with those from other studies.

5.2.5 Similar Outcome Studies

The findings in this study is similar, as well as in agreement with a previous study on early surgical outcomes in Scotland, where 50% of study models assessed were said to be in the

good category-grades 1&2 (Clark et al., 2007). Both studies used the retrospective cross-sectional design and surgical outcome of only one surgeon was examined in both cases. In the British sample, 30 study casts were assessed, while in the Nigerian study, 18 study casts of 5 year old children were assessed. Lack of co-operation in this age group was reported as the main reason for obtaining a small sample size in the British sample, both sample however allowed for statistical analysis and can be said to be reliable.

Multicentre studies involving 10 Scandinavian cleft care centers (Scandcleft) have also reported on dental arch relationships in 5 year old children with UCLP. Study casts of 445 affected children from Scandinavian countries were assessed to report on treatment outcome (Heliovaara et al., 2013).

Other studies on dental arch relationships have also reported on mean scores in 5 years old in different populations. In Japan, Suzuki and colleagues reported a mean score of 2.95 among 5 year old Japanese children, indicating fair treatment outcome in that selected population (Suzuki et al., 2005). One study in America (Flinn et al.,2005) compared outcomes in 5 year olds from three centres(A,B,C) using the 5 year old index and reported mean scores of 2.0, 2.1&2.4 from each of the centres respectively. The percentage distribution of scores showed that over 50% of study models (Centre A; 72%, Centre B; 63% 7 Centre C; 59%) were assessed to be of good outcome. The findings in this study just like that of the American study have also demonstrated good treatment outcomes.

In reporting the 5 year old index as a useful tool to assess UCLP outcomes, Attack and colleagues assessed 54 study models of 5 year olds, out of which 13-18% were said to be of poor outcome; grades 4 &5(Attack et al., 1997). This is contrary to what obtained in this study, as none of the study models (0%) were assessed to be of poor outcome. The level of agreement between assesors is important and should be recorded to determine the reliability of such studies.

5.2.6 Inter-rater and Intra-rater agreement

The intra and inter-rater agreement aims at producing either qualitative or quantitative measurements under the same experimental conditions and is an indication of the reliability of the study (Gwet,2014).There was good inter-rater agreement ($k>0.60$) and very good intra-ratter agreement (0.85) during the assessment of the Nigerian and European samples with both the 5 year old index and the MH/B system. This is agreement with another study

that assessed treatment outcome in children affected with CL/P, moderate, good and very good intra and inter-rater agreement was reported (Gray& Mossey,2005) .The Modified Huddart /Bondeham system (M/HB)system allows quantitative scoring of dental models and agreement by assessors.

5.2.7 Modified Huddart/Bondeham System

In this study, the Modified Huddart/Bondeham system was also used to assess treatment outcome. The MH/B system is said to be objective with relative simplicity and requires no clinical experience for scoring (Gray and Mossey, 2005). This was of great advantage for the PI (AA) who had no experience in scoring, the weighted kappa statistics showed very good (0.85) and moderate (0.60) inter and intra examiner agreement respectively with the use of this index, confirming further the validity and reliability of the study. Another advantage of the MH/B system is its versatility in that it can be applied to models of any cleft sub-group and age, though in this study it was applied to 5 year olds only. One great benefit of the MH/B system over other treatment outcome indices is its ability to be very sensitive to changes, it is an ordinal continuous scale unlike the 5 year old index which is categorical. This allows changes as little as 0.5 that can enable differentiation between categories of cleft groups to be detected (Mosey and Gray, 2005), thereby allowing differences between ratters and samples to be statistically detected.

5.3 Other Treatment Outcome Studies in the Field of Cleft.

Several studies have reported on treatment outcome in children affected with CL/P in different populations.

5.3.1 The Eurocleft Study

In Europe, the Eurocleft studies (Asher-McDade et al., 1992; Mars et al., 1992; Mølsted et al., 1992a; Shaw et al., 1992b) was a concerted effort of six European centres to measure outcomes in cleft patients aged 8-11 years. These patients were followed up longitudinally in five of the initial participating centres, till the patients were more skeletally matured (age 12 to 17 years).Years later, further series of reports on five of the Eurocleft centres were published (Shaw et al., 2005; Semb et al., 2005a,b; Brattström et al., 2005; Mølsted et al., 2005). All patients used for the study were affected with UCLP and had been treated previously Details of the history were obtained from the clinical notes and the following outcome measures were assessed; Craniofacial form and soft tissue profile; Dental arch

relationships, Nasolabial appearance and speech (Grunwell et al., 2000). The general findings of the Euro cleft study was evident, important differences in treatment success at different centres were reported; Treatment success appeared to be associated with well organised centres of excellence; outcomes were consistency in all the centre over the time; treatment were judged to be cost effective and simple,

there was no association between the amount of treatment and the final result; there was a strong association between outcome and patient/parent satisfaction.

The findings of the Eurocleft study then provided the basis for 'The Clinical Standards Advisory Group (CSAG) study' (Sandy et al., 2001). This was a follow-up to an investigation on the quality of care for children born with complete unilateral cleft lip and palate in the UK (CSAG, 1998).

5.3.2 The Clinical Study Advisory Group (CSAG)

This study was conducted over a 15-month period, a total of 601 out of 647 children participated in the study. Children from two cohorts; 5-year-olds and 12-year-olds were recruited from 50 cleft centres. The Five-year-old Index (Attack et al., 1997) was used to assess dental arch relationship for the 5-year-old group, 239 out of 326(73%) patients had no need for orthodontic treatment or bone graft surgery.

In the 12-year-old group, 218 out of 321(68%) patients who were assessed by the Bergland Index (Bergland et al., 1986) and the Goslon Index (Mars et al., 1987) to rate the effectiveness of secondary alveolar bone grafting and facial growth. Other outcome measures reported were skeletal relationship, general oral health, psychological status, speech, difficulties attending cleft clinics, patient/parent satisfaction. The results confirmed the need for better levels of care and the following recommendations were issued.

The following recommendations were made to the UK Health department;

Reduction of the number of the cleft centres in the UK from 57 to 8-15 in order to optimize public resources; Purchasers and commissioners should ensure specifications and clearly indicate the range of required expertise, the quality standards, and the information for contract monitoring; Trusts and provider units should review their services and ensure that the full range of clinical skills is available. Practitioners and clinicians should agree on a common database for all patients; Information should also be available for comparative audit studies; Royal Colleges and faculties should ensure that training programs for all specialist cleft clinician should be approved only in cleft centres with high volume of high quality clinical experience; The office of National Statistics should ensure that completeness of recording of the records of births of children in the UK should be improved.

5.3.3 Scandcleft

The members of 4 centre Scandinavian countries (Friede etal., 1991) and the six centre international study (Asher Mc-Dade et al., 1992; Mars et al., 1992; Mostled et al., 1992; Shaw et al., 1992) developed a concerted programme for multi-disciplinary intercentre research in cleft lip and palate. Initiation of randomised controls trials (RCTs) in clefts were started, however such RCTs were regarded as unethical and unmanageable in the past (Berkowitz 1995; Shaw, 1995). Later on the scandcleft project along with 10 European teams initiated RCTs for primary surgery for patients with UCLP (Semb, 2001). Four surgical variations were randomly allocated; short delay in closing hard palate; long delay in closing hard palate; simultaneous soft and hard palate closure; early hard palate closure with vomer flap at the same time as lip closure.

5.3.4 Eurocleft Project

As a follow up to the Scandinavian intercentre study (Friede et al., 1991) and the Eurocleft cohort study, further research was initiated. The Eurocleft Clinical Network Project (1996-2000) and the EUROCRAN (European Collaboration in Craniofacial Anomalies)(Shaw et al., 1996;Shaw et al., 2001).The Eurocleft project identified the following as shortcoming in cleft care services in Europe; consequences of poorly organised care, lack of evidence based practises and uncoordinated cleft research. Advantages of the collaboration were; the creation of a network among European cleft care specialists; development of consensus recommendation on services and a survey of the services offered as of then. The benefit of inter-centre comparisons were evaluated this led to recommendation of the following; Need for routine clinical audit by establishing good practise archives, registries and benchmarks (Shaw, 2005). The Eurocan research program initiated a group of multi-centre randomised clinical trials of variations in surgical techniques and molecular genetic studies.

5.3.5 Americleft

In 2006, Flint and colleagues conducted a retrospective studies on different protocols of primary surgery on 118 patients (Flint et al., 2006). Data from three centres were retrieved (Centre A=41;Centre B=33;Centre C=44), treatment outcome was assessed using the 5 year old index , 2 centres (A&B) showed comparable good scores , while the 3rd centre (C) had a lower score than the others, but showed the least burden of care with only two surgical techniques and pre-surgical orthopaedics.

5.3.6 Japancleft

A similar study to the Eurocleft study was conducted in Japan (Susami et al., 2006).Dental arch relationships of Japanese children with UCLP were compared with those of the Eurocleft study using the GOLSON yardstick. However the sample size was small and the study was retrospective, it was considered a preliminary study.

5.3.7 BIAS IN CLEFT RESEARCH

Bias in cleft research has been reported previously (Shaw et al., 2015). Data collected for this study may have been subjected to bias.

5.3.7.1 Data collection

Comparisons of facial growth data may be unreliable in the presence of inherent differences in facial form among communities ;case mix bias (Shaw et al., 2015). The skill of a more gifted surgeon or clinical team may inflate the apparent effectiveness of the technique used ;Proficiency bias. However, without knowing about all cases in which a particular technique was tried, reliable conclusions cannot be drawn ;Follow-up bias. To minimize bias the study design should be appropriate. Opportunities for non-experimental retrospective comparisons of therapies or protocols of care can arise in several ways: by the coexistence of different therapies at the same centre, by the replacement of one therapy with another, or by collaboration of two or more centres (Shaw et al., 2015).

In this study, data was collected from only one centre, to minimise bias, a multicentre study may have been appropriate. The merits and demerits of single and multicentre studies are discussed further.

5.3.7.2 Single Centre Study

Individual reports of the treatment outcomes at particular centres have been by far the commonest form of presenting outcomes in the literature, though surrounded by fundamental difficulties in making comparisons with reports elsewhere because of the invariable presence of a set of potential biases (WHO, 2002).Several single center studies have been reported previously in cleft literature, contributing much to knowledge and adding to the existing literature. One study conducted in the United Kingdom that determined the impact of cleft size on treatment outcome in children with UCLP (Johnson et al., 2000). Longitudinal

records of 48 children with UCLP were examined initially to determine the severity of the cleft. These children were followed up to 6 years of age and their dental arch relationships were examined, the results showed that the initial cleft size did not have any influence on the quality of treatment outcome. In Belgium, another single center study, assessed long-term velopharyngeal function in children with non-syndromic cleft lip and palate (Vander Poorten et al., 2006). The Leuven protocol was used for surgical repair, the study showed that there was no need for secondary pharyngoplasty in majority of the patients, 84% of the patients did not display VPI related articulation disorders. The conclusion was that velopharyngeal function is excellent in patients treated by this protocol. Another retrospective study in Athens Greece, determined treatment outcome in all children with cleft of the lip and palate seen from 1995-2007 were reviewed. A total of 530 patients were eventually reviewed, majority had very good and excellent treatment outcome. These studies allowed comparative evaluation of different surgical protocols and techniques by one surgeon, hence more likely to be subject to casemix bias because of the diverse background and inherent error in selected sample. Single centre studies are more likely to be subject to reporting and proficiency biases (Semb et al., 1991;Semb and Shaw,1998,Shaw et al., 2015).Therefore the integrity of the researcher is very important, because findings disseminated would depend largely on their judgment.

5.3.7.3 Intercentre Comparison

Intercentre comparison has the main advantage of obtaining adequate samples, within a specific subtype treated by contrasting treatment modalities (Shaw et al., 2016). Intercentre collaboration may also provide insights into the processes and outcomes of treatment of comparable services elsewhere, the establishment of future goals, and the exchange of evidently successful practices (Shaw et al., 2005).The advantage lies in the fact that patients can be recalled prospectively, which can allow data on outcome to be collected in a standardized way, rigorous planning and execution across centers can ensure recruitment and consistent evaluation, provided the entry requirements for the study are equivalent in all centers (Shaw et al.,1992) .This strategy is extremely valuable in assessing surgical outcome as well as other treatment modalities. One major advantage of inter centre comparisons is the minimization of analysis and reporting bias whereas ingle centre reports usually relies solely on the individual judgement. Limitations of intercentre comparisons lies in the fact that they are unable to distinguish between the influence of a centre's protocol on its outcome nor

between treatment protocol and the influence of the personnel who deliver that protocol (Shaw et al., 2005).

The fact is that ,it is easier to facilitate statistical comparison across the single data set rather than multiset data, there should be sufficient information available to permit statistical comparison. It is difficult to establish the harmful effect of a particular surgical technique due to several other associated factors such as surgical skill, technique, timing sequence and other ancillary care (Shaw et al., 2015).

5.3.7.4 Minimizing bias in Cleft Research

To minimize bias in cleft research, appropriate design of comparison studies must be undertaken (Sackett,2000).The initial accumulation of series of cases may provide insight into the relative effectiveness of a technique, direct comparison with alternative form of treatment is necessary to establish the true effectiveness of the treatment. Table 5.2 shows the different bias that can occur when conducting cleft research.

Table 5.1 Types of Bias in Cleft research

Follow-up bias	Conclusions should not be made without having full details about all the cases, rigorous follow-up should be done for cases that went badly.
Exclusion bias	This is when cases are excluded in retrospect; it may remove any equivalence the comparison group may have had.
Analysis bias	When raters are not blinded to treatment allocation, they may exhibit bias towards a particular technique.
Reporting bias	This occurs when negative or disappointing findings remain unpublished, the results of the technique cannot be accurately compared

5.4 STUDY DESIGNS THAT CAN MINIMIZE BIAS

5.4.1 Prospective Cohort Studies

Many craniofacial surgical interventions are at the introductory phase and experiencing constant modification of both clinical skills as well as the applied population, hence randomized trial may be impossible to carry out at this stage. Establishing prospective cohort studies will enable critical appraisal of different interventions and minimize the biases that may occur with reporting.

5.4.2 Randomized Controlled Trials

RCTs allow for comparison of therapies both scientifically and ethically, prognostic factors tend to be balanced among treatment groups. Patients are usually registered before treatment which is done with a clearly defined protocol, followed up is then done prospectively, in such a case it is less likely for data to be missing, because the potential loss to follow-up and late exclusion is reduced. There is the need to formalize treatment protocol at the onset, by an ethical review board or funding agency, which increases the likelihood of consistent record collection and impartial analysis. RCTs can yield biased results especially if the randomization procedure is not strictly followed while RCTs with insufficient cases may give misleading results.

The need for RCTs in cleft research had been identified many years ago by Spriestersbach and colleagues (Spriestersbach et al., 1973) who identified the need for prospective research in cleft management. However RCTs in cleft research are not popular (Roberts et al., 1991).

5.4.4 Systematic Review Studies

Systematic reviews help to establish whether scientific findings are consistent and generalizable across populations, settings, and treatment variations. The explicit methods used in systematic reviews limit bias and improve reliability and accuracy of conclusions(Shaw et al 2015) . Meta-analysis, is a statistical method to summarize the results of independent trials, which can provide more precise estimates of the effects of health care than results derived from individual studies.

The Cochrane Collaboration is an international organization established to prepare, maintain, and promote the accessibility of systematic reviews of the effects of health care interventions, as well as RCTs in the field of CL/P can be completed and reported, it is a primary source of review (www.cochrane.org).

5.6 THE CURRENT STANDARDS OF CLEFT CARE IN NIGERIA

Generally speaking, minimum standards of care should be applicable to cleft teams as guidelines or code of practise, to enable them develop their own ethos and approach towards cleft care. Services such as prenatal diagnosis, genetic counselling are starting point in the treatment of children born with CL/P, this is because parents that are well informed ahead of their children's condition are better able to cope. Prenatal diagnosis is considered important for affected Nigerian families because of the associated stigma that may impact greatly on their quality of life. Early diagnosis may allow affected families to adopt coping strategies and more willing to receive treatment.

Advances in ultrasound technology (Nussbaum et al., 2008) have been made in recent years and it is estimated that globally around 20% of clefts involving the lip are now detected prenatally. Although some surveys have reported detection rates as high as 45% (Johnson and Sandy 2003; Martin 2005b; Cleft Lip and Palate Association 2007; Nussbaum et al.,2008). The capacity to accurately assess the extent of a cleft, particularly one affecting a child's palate, remains limited, as it does for the detection of other potentially associated conditions as well (Aspinall 2002; Johnson and Sandy 2003). Research (Farrimond and Morris 2004; Cadogan et al. 2009) reported the emotional impact of prenatal diagnosis, experiences of ten couples in relation to 'knowing or not knowing' about their child's cleft before birth, with mixed feelings of parents reported. Parents who had prenatal diagnosis felt that having prior knowledge was beneficial, however it is interesting to note that several parents who discovered their child's cleft only at birth would not have chosen to know otherwise (Cadogan et al. 2009).

Comprehensive cleft care is the gold standard for cleft care, but seems an uphill task to achieve in Nigeria, going by reports from previous studies (Olasoji et al., 2011; Oginni et al., 2014), the main form of care offered in Nigeria hospitals still remain primary surgical repair of lip and palate. The importance of comprehensive cleft care has been reported previously (Grunwell et al., 2000;Nolletetal.,2005;Fudalej et al., 2009;KosZelj et al., 2012;Peanchitlertkajorn eral., 2011;Bartzela et al., 2010). This can be seen in the United Kingdom where cleft care services was re organised based on a report of poor treatment outcomes (Shaw et al., 1992). Regional centres were then established to allow for more comprehensive cleft treatment (Murray, 2003). Treatment outcome of children assessed after the re organisation showed much improvement (Hathorn et al., 2006).

Since primary surgical repair is still the main form of care offered in Nigeria, it raises concerns as to the standard and quality of care being offered to children with CL/P in this country. This is because comprehensive cleft care using a multidisciplinary team approach has been recognized as the best option of treatment; it is offered by a range of cleft specialists, following an established protocol (Marcus son et al., 2001; Hodgkinson et al., 2005; Clinical Standards Advisory Group, 1998). Further, advantages of multidisciplinary care include fostering a sense of belonging among specialists, more likely to be more cost-effective and generate better treatment outcome as well as enhance research (Kujipers-Jagtman et al., 2000; Semb, et al., 2011; Semb, 2014).

Ideally, comprehensive cleft care should start at birth with the aim of holistic management and complete rehabilitation so that affected children can live a normal life. The general cleft protocol has been described with different stages of treatment with their timing (Sommerlad 1994; Clinical Standards Advisory Group 1998; Hodgkinson et al 2005). Failure to comply with such cleft protocol may result in poor treatment outcome.

Presently, most hospitals in Nigeria are not able to offer comprehensive cleft care; this perhaps might impact negatively on the quality of cleft care being offered. Inability to offer comprehensive cleft care in Nigeria is mainly due to lack of funding
(Olasoji et al., 2011); this is because free sponsorship is only available for primary surgical repair.
The Eurocleft (Shaw et al., 2001) consensus for finances, recommended resources to be available for all aspects of cleft care. Presently, this is not the case in Nigeria, since the government does not finance health care, most parents desiring comprehensive cleft care for their children would have to do so on an 'out of pocket basis'.

There are few hospitals in Nigeria that offer orthodontic treatment to patients with CL/P. The role of the orthodontist in the management of children with cleft lip and palate is important because of the aesthetic and functional consequences associated with CL/P (Lee &Kim, 2003; Hodgkinson et al, 2005). The orthodontist also advices on surgical timing of palatal repair based on his knowledge and experience with growth and development of the jaws.

The usual practise is for children affected with CL/P to be seen in the pre-surgical, post - surgical and adolescent or adult stages. There is weak evidence for pre-surgical orthopaedics for children with CL/P (Shaw et al., 2015), in the post-surgical phase, the orthodontist is concerned with guidance of tooth eruption in the deciduous dentition and the establishment of

an overbite using removable appliances to influence growth and preparation of the maxillary arch prior to secondary alveola bone grafting (Bergland, 1973). In the adolescent or adult stage, treatment is done to establish occlusion and function in the permanent dentition.
Children with CLP in Nigeria are not referred for routine dental check-ups. The importance of dental health education and fluoride therapy is to prevent the development of caries and periodontal problems in affected children, as they are likely to be more prone to caries and poor oral hygiene because of the poor arrangement of their teeth. (Cheng et al, 2007). These children also do not get referred to paediatricians for assessment of developmental growth. This is however important because delay in developmental growth of children with CL/P has been reported previously (Harris and Hullings, 1990; Zarate, 2010), early referral may allow for intervention in growth and improve outcome.

Majority of the cleft teams do not conduct audit of cases seen in their hospital, the consensus at the Euro cleft project was the recommendation of at least 40- 50 cases of affected children per year per centre (Shaw et al; 2001). The importance of audit is to allow for verification of the quality of treatment being offered and unless there is reasonable volume of cases with appropriate records being treated at each centre, it may be difficult reporting meaningful treatment outcome.

This study revealed children born with CL/P are recruited during outreaches and awareness program on oro-facial clefts, a previous Nigerian study reported increase turnout of cleft patients after such outreaches (Onah et al; 2008).

In conclusion, this study determined treatment outcome in Nigerian children affected with CL/P and the current status of cleft care in Nigeria. The quality of surgery offered in a selected cleft care centre is good. There are however shortcomings in cleft care services offered in the country .

WHAT THIS THESIS HAS CONTRIBUTED TO KNOWLEDGE

This thesis reports treatment outcome in a selected Nigerian Cleft population and the current status of cleft care in Nigeria.

Treatment outcome was determined by measuring dento-facial relationships using reliable and validated indices of measure. The mean score was 2.3 for the Nigerian sample indicating good treatment outcome, implying that the treatment protocol and surgery offered in this selected Nigerian hospital is effective.

This thesis has also reported on the current status of cleft care in Nigeria using the WHO standards of cleft care as a yardstick, shortcomings of cleft care in the country has been reported.

The findings of this thesis is novel, being the first time such a study would be carried out in a Nigerian cleft population. The findings would be disseminated through publications in peer reviewed journals, the aim is to further contribute to knowledge and add to the existing literature in the field of cleft. Evidence provided in this study may be used to judge the standards of cleft care presently being offered in the country and may serve as a basis for comparison of standards in other countries.

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APPENDIX 1

QUESTIONNAIRE

Cleft lip and palate care in Nigeria

Serial No __

Dear Specialists,

We are currently collecting data from cleft care specialists in Nigeria on services available for children affected with cleft lip and palate. This questionnaire covers several aspects of cleft care, training of cleft specialists as well as cost of treatment. We will appreciate your response to all the questions in this questionnaire.

Thank you

Principal Investigator

Section A

1. **Name of the hospital:** _____
2. **Location of practise:** City ☐ Rural ☐ Town ☐
3. **Type of practise:** Private ☐ Government P ☐ Second ☐ert ☐
   Religious ☐
4. **Region of practise:** Please tick (✓)
   - South-West ☐
   - South-East ☐
   - South-South ☐
   - North-East ☐
   - North-West ☐
   - North-Central ☐

5. **Professional specialty:** Please tick (✓)
   - Ear, Nose, and Throat surgery ☐
   - General surgery ☐
   - Maxillofacial surgery ☐
   - Paediatric surgery ☐
   - Plastic surgery ☐
   - Orthodontists ☐
   - Speech therapists ☐

Specialists Nurse ☐

Anaesthetist ☐

Specialist Nurse

Others (specify) _____

6. **Highest Qualification:**

First degree

Masters ☐

Doctoral ☐

Fellowship ☐

Others (specify) _____

Section B

Services

7. **Type of Cleft Team**

	Before 2005		After 2005	
Multidisciplinary	Yes ☐	No ☐	Yes ☐	No ☐
Surgical	Yes ☐	No ☐	Yes ☐	No ☐
Individual	Yes	No	Yes	No
Others please specify	_____		_____	

8. **Please tick specialists in your hospital and double tick team co-ordinator**

	Before 2005	After 2005	
Dentist	☐	☐	
Nutritionist	☐	☐	
Orthodontist	☐	☐	
Speech therapist	☐	☐	☐
Pathologists	☐	☐	
General surgeon	☐	☐	
Maxillofacial surgeon	☐	☐	☐
Paediatric surgeon	☐	☐	
Plastic surgeon	☐	☐	
Specialists Nurse	☐	☐	
Anaesthetist	☐	☐	
Geneticist	☐	☐	
Psychologist	☐	☐	
Paediatric developmental Medicine	☐	☐	

ENT Surgeon

9. **Please tick which of the following services that are available in your hospital**

	Before 2005	**After 2005**
Neonatal counselling	☐	
Neonatal Nursing	☐	☐
Surgery	☐	☐
Clinical genetics		☐
ENT	☐	☐
Orthodontic	☐	☐
Dental	☐	☐
Speech therapy		☐
Psychological counselling	☐	
Parent support group	☐	
Social Welfare Officer		

10. **Tick the records you routinely take for patients with cleft lip and palate in your hospital.**

	Before 2005	**After 2005**
Photographs		
Before surgery	☐	☐
During surgery	☐	☐
After surgery	☐	
Radiographs	**before 2005**	**After 2005**
Extra oral	☐	☐
Cephalometric	☐	☐
OPG	☐	☐
Intra oral	☐	☐
Peri apical	☐	☐
Occlusal	☐	☐
Study models		
Before Surgery	☐	☐
After surgery	☐	☐
Medical records	☐	☐

Orthodontic care

11. Do the orthodontists have further training in management of children with CL/P?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	☐

12. How many cases of cleft patients do you see for orthodontic treatment on an annual basis?

	Before 2005	**After 2005**
1 – 5	☐	☐
6 – 10	☐	☐
10 – 20	☐	☐
20 and above		

13. Please tick the orthodontic care routinely done in your hospital

	Before 2005	**After 2005**
Pre surgical orthopaedics	☐	☐
Pre bone graft in mixed dentition	☐	☐
Fixed appliances for arch alignment	☐	☐
Others _____		

14. Do the orthodontists give any feedback to surgeons on growth related problems?

Yes ☐ No ☐

Dental care

15. Do you routinely give dental health education to cleft children in your hospital?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

16. Specify which of the specialists give dental health education. _____

17. Do children with cleft have priority access to a paediatric dental consultant?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

18. Do adolescents have priority access to a restorative dentistry consultant? Yes ☐ No ☐

Surgery

19. Average annual number of new cases for the following types of surgical repair

	Before 2005	**After 2005**
Unilateral cleft lip repair	_____	_____
Bilateral cleft lip repair	_____	_____
Cleft palate repair	_____	_____

Alveolar cleft repair	_____	_____
Sub mucus cleft repair	_____	_____
Velopharyngeal insufficiency	_____	_____
Midrace secondary surgery	_____	_____

20. **What is the usual age for the following procedures?**

	Before 2005	**After 2005**
Counselling	_____	_____
Cleft lip repair	_____	_____
Cleft palate repair	_____	_____
Pre surgical orthopaedics	_____	_____
Alveola bone graft	_____	_____
Orthodontic treatment	_____	_____
Dental treatment	_____	_____
Secondary surgery	_____	_____
Speech therapy	_____	_____

21. **What is the usual anaesthetic technique you employ for repair**

General anaesthesia----------

Local Anaesthesia ----------

22 How long do the children stay in the hospital before and after surgery----------

---

22. **Indicate the preferred technique for these procedures:**

a. **Unilateral cleft lip repair**	**Before 2005**	**After 2005**
Millard rotation advancement	☐	☐
Straight line	☐	☐
Triangular flap	☐	☐
Others (specify) _____		_____

b. **Bilateral cleft lip repair**	**Before 2005**	**After 2005**
Millard rotation advancement	☐	☐
Triangular flap	☐	☐
Straight line	☐	☐
Others (specify) _____		_____

c. **Cleft palate repair**	**Before 2005**	**After 2005**
Von Lange beck	☐	☐
		☐
		☐

Palatal pushback ☐

Double-opposing Z-plasty ☐

Vomer flap ☐ ☐

Others (specify) _____ _____

23. Do you routinely repair the nose during cleft lip surgery?

Before 2005		After 2005	
Yes ☐	No ☐	Yes ☐	No ☐

24. Are you mindful of the Nasio labial angle? Yes ☐ No ☐

25. Do you routinely repair the vermilion during lip surgery? Yes ☐ No ☐

26. **Concerning bilateral cleft lip repair, please indicate which of the following methods** **Before 2005** **After 2005**

Both sides done together as a single procedure ☐ ☐

Each side done as a separate procedure ☐

27. **Palatal repair please indicate which of the following methods**

	Before 2005	After 2005
Hard and soft palates are done as single procedure	☐	☐
Hard and soft palates are done as separate procedure	☐	☐

27. How successful are your cleft lip and/or palate repair

Very successful 1out of 10 with fistula formation		**Moderately successful 2-4 out of 10 with fistula**		**Little success 5-9 out of 10 with fistula formation**		**No success 10 out of 10 with fistula formation**	
Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005

What is the usual protocol of care for children with CL/P in your hospital?

		Before 2005	**After 2005**
a.	Neonatal counselling – Pre-surgical orthopaedics – Primary surgery – Secondary surgery – Dental treatment – Orthodontics – Others		

b.	Pre-surgical orthopaedics – Primary surgery – Secondary surgery – Dental treatment – Orthodontics – Others		

Section C: Referral System

28. **What is the usual pattern of referral of children with CL/P?**

	Before 2005	**After 2005**
Children are referred from the neonatal unit of the hospital	☐	☐
Children are self - referred	☐	☐
Children are referred by private practitioners	☐	☐
Children are referred by community health workers	☐	☐

29. **For services that are not available in your hospital which of the following services do you routinely refer children with CL/P for?**

	Before 2005	**After 2005**
Audiology/Otology	☐	☐
Psychology	☐	☐
Speech therapy	☐	☐
Orthodontic treatment	☐	☐
Dental treatment	☐	☐
Pre surgical orthopaedics	☐	☐

30. Do children with cleft lip and palate in your hospital receive Pre-surgical orthopaedics?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

31. What are the types of appliances used during pre-surgical orthopaedics?

Passive method	**before 2005**	**after 2005**
Adhesive tape	☐	☐
Head cap	☐	☐
Lip adhesion	☐	
Active method		
Orthodontic appliances	☐	☐
NAM appliance	☐	☐

Section D – Training

32. Do you have further training as a cleft care specialist?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

33. Is there any formal training for residents in cleft lip palate care in your hospital?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

34. Have you had any form of training in cleft lip and palate in the following areas?

	Before 2005	**After 2005**
Workshop with hands on	☐	☐
Online	☐	☐
Seminars with oral and poster presentations	☐	☐
Conference Attendance	☐	☐
Research	☐	☐
Publications		

35. Please tick the number of publications that you have written in the field of CL/P

	Before 2005	**After 2005**
None	☐	☐
1-5	☐	☐
5-10	☐	☐
10 and above	☐	☐

36. Have you ever assessed web for the following resources on cleft care?

	Before 2005	**After 2005**
Educational and teaching tools	☐	☐
Cleft library	☐	☐
Cleft data base	☐	☐
Other sources, please specify _____		_____

37. Do you collaborate with other cleft specialists in other hospitals?

Before 2005		**After 2005**	
Yes ☐	No ☐	Yes ☐	No ☐

38. How much of clinical experience do you have in the following aspects of cleft care?

Aspect	**Much experience**		**Moderate experience**		**Little experience**		**No experience**	
	Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005

Primary surgical repair								
Secondary alveola bone graft								
Secondary cleft procedures. Please indicate _____ _____								
Ear Nose and Throat services								
Speech therapy								
Orthodontic treatment								
Dental treatment								

Section E – Counseling

39. Do you have formal counseling services for the parents of children with CL/P?

Before 2005				**After 2005**			
Yes	☐	No	☐	Yes	☐	No	☐

40. Please tick the following specialists involved in counseling

	Before 2005	**After 2005**
Nurse	☐	☐
Dentist	☐	☐
Nutritionist	☐	☐
Orthodontist		
Speech therapist	☐	
Pathologists	☐	
General surgeon	☐	☐
Maxillofacial surgeon	☐	☐
Paediatric surgeon	☐	☐
Plastic surgeon	☐	☐
Orthodontist	☐	☐
Speech therapist	☐	☐
Specialists Nurse	☐	☐
Anaesthetist	☐	☐
Geneticist	☐	☐
Psychologist	☐	

Section F – Feeding of Cleft babies

41. What advice do you give to parents of babies with cleft on feeding?

	Before 2005	After 2005
Advice mothers to encourage breast sucking in Unilateral cleft	☐	☐
Advice mothers to encourage breast sucking in Bilateral cleft	☐	
Advice mothers to squeeze breast milk into teat bottle before giving	☐	☐
Advice mothers to use baby milk formula only in long teat bottle	☐	☐
Advice mothers to feed using cup and spoon	☐	☐

Others----------

Section G – Cleft funding

42. What is the usual source of funding for cleft patients in your hospital?

	Before 2005	After 2005
Government	☐	☐
Private	☐	☐
Charity	☐	☐

43. Which of the following procedures are funded in your hospital?

	Free		**Part payment**		**Payment**	
	Befor 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005
Surgery						
Consultation fees						
Surgical procedure						
Drugs						
Accommodation expenses for parents						
Transport						
Hospital admission fees for child						
Orthodontics						
Speech therapy						
ENT services						
Alveola bone graft						
Secondary surgical procedures						
Other ancillary care(specify)						

44. **Cost of treatment in dollars**

	<100		**100 – 200**		**200 – 300**		**>300**	
	Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005	Before 2005	After 2005
Surgery								
Alveola bone grafting								
Secondary surgery								
ENT services								
Orthodontic services								
Speech therapy								

Cleft charities

45. Is your hospital affiliated with any charity organization?

Before 2005			**After 2005**		
Yes	☐	No ☐	Yes	☐	No ☐

46. Please tick the type of relationship your hospital has with charity organizations.

	Before 2005	**After 2005**
Partnership	☐	☐
Full sponsorship	☐	☐
Cleft Missions	☐	☐
Outreach programs	☐	☐

Audit and Research

Have you ever carried out an audit of cleft treatment in your hospital?

Before 2005			**After 2005**		
Yes	☐	No ☐	Yes	☐	No ☐

47. How many cases were seen and audited

	Before 2005	**After 2005**
1-10	☐	☐
11-20	☐	☐
21-30	☐	☐

30 and above ☐

48. Have you ever conducted clinical trials study using cleft patients?

Before 2005 **After 2005**

Yes ☐ No ☐ Yes ☐ No ☐

49. Have you ever conducted any form of research using cleft patients in your hospital?

Before 2005 **After 2005**

Yes ☐ No ☐ Yes ☐ No ☐

50. Overall would you say charity organizations have had an impact on cleft care in Nigeria?

Before 2005 **After 2005**

Yes ☐ No ☐ Not sure ☐ Yes ☐ No ☐ Not sure ☐

51. Do you recall children with CL/P after treatment Yes ☐ No ☐

52. How soon do you recall after treatment

7-14days ☐

14-28 days ☐

1-3 months ☐

Every 6 months ☐

Yearly ☐

Outcome

53. How do you assess outcome of surgery for cleft lip patients----------

54 Challenges

What are the challenges with cleft care in your Centre?----------

55. Patients satisfaction-Tell us how you measure patients' satisfaction----------

Appendix 2: Copy of Consent form

Consent form for parents

Name of Researchers: Abigail Adeyemi, William Shaw, Akintububo Benedict

	Please initial all boxes
1. I confirm that I have read or listened to and understood the participant information sheet (version 2: June 2013).	
2. I have had the opportunity to ask questions about the study and have had them answered satisfactorily.	
3. I understand that my participation in the study is voluntary and that I can withdraw at any time, without giving a reason and without my child's medical care or legal rights being affected.	
4. I agree to be interviewed this may be recorded and transcribed ,also impressions of my upper and lower jaws will be taken.	
5. I understand and agree that anonymised quotations from my interview can be used in reports, publications or conferences to illustrate the findings of the research study.	
6. I understand that relevant sections of data collected during the study may be looked at by responsible individuals from the University of Manchester,where it is relevant to my taking part in the research. I give permission for these individuals to have access to this data.	
7. I agree to take part in the study.	

Name of participant	Date	Signature
Name of person taking consent	Date	Signature

Appendix 3 : Copy of ethical clearance letter, University of Manchester

Secretary to Research Ethics Committee 5

Faculty Office - Devonshire House

Tel: 0161 275 0288

Email: jared.ruff@manchester.ac.uk

Dr Abigail Adeyemi

School of Dentistry

17th September 2014

Dear Dr Adeyemi

Research Ethics Committee 5 (Flagged Humanities) - Project Ref 14266

Adeyemi: Cleft lip and palate care in Nigeria (ref 14266)

I am writing to thank you for submitting the requested changes and clarification to the original material which was reviewed by UREC 5 in your absence on 28th July 2014. This letter formally confirms approval for the above project and that no further changes are required to the documentation submitted to the committee.

This approval is effective for a period of five years and if the project continues beyond that period it must be submitted for review. It is the Committee's practice to warn investigators that they should not depart from the agreed protocol without seeking the approval of the Committee, as any significant deviation could invalidate the insurance arrangements and constitute research misconduct. We also ask that any information sheet should carry a University logo or other indication of where it came from, and that, in accordance with University policy, any data carrying personal identifiers must be encrypted when not held on a university computer or kept as a hard copy in a location which is accessible only to those involved with the research.

Finally, I would be grateful if you could complete and return the attached form at the end of the project.

I hope the research goes well.

Yours sincerely

Jared Ruff
Senior Research Manager
Faculty of Humanities and Secretary to UREC 5 (Flagged Humanities)
0161 275 0288 Jared.ruff@manchester.ac.uk

FEDERAL MEDICAL CENTRE, GOMBE

Ashaka Road, Gombe
P.M.B. 0037

Tel: 072-223410,
223064

Fax: 072-223909

Our Ref: _____ Date: 11 June, 2014

Abigail Adeyeni
University Dental Hospital,
Higher Cambridge Street
Manchester M15 6FH
United Kingdom

Sir,

ETHICAL CLEARANCE

I am directed to inform you that your application and proposal titled **'OUTCOME ASSESSMENT IN UNILATERAL CLEFT LIP AND PALATE (UCLP).''** Submitted to the Hospital Research and Ethics Committee, have been duely reviewed and approved.

On behalf of the committee, I wish you a successful execution of the research.

Thank you,

B.A. Sambo Mrs. (JP,CLN, AD)
Secretary R&EC