ANALYSIS OF DENTAL ANOMALIES IN PATIENTS WITH UNILATERAL CLEFT LIP AND PALATE AT ACADEMIC HOSPITALS IN THE WESTERN CAPE, SOUTH AFRICA.

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ANALYSIS OF DENTAL ANOMALIES IN PATIENTS WITH UNILATERAL CLEFT LIP AND PALATE AT ACADEMIC HOSPITALS IN THE WESTERN CAPE SOUTH AFRICA.

A mini thesis of Dr Vuyisile Solomon Gomba, student number: 3203262, submitted in partial fulfilment of the requirements for the degree of MDS in the Department of Orthodontics, University of the Western Cape, November 2022.

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KEYWORDS

- **Cleft lip and palate**
- **Dental anomalies**
- **Supernumerary tooth**
- **Tooth Agenesis**
- **Ectopic eruption**
- **Peg shape**
- **Transposition**

ABSTRACT

Objectives

The aim of this study is to determine the type and frequency of dental anomalies associated with patients with non-syndromic complete unilateral cleft lip and palate receiving treatment at Academic hospitals (UWC Oral Health Centre and Red Cross War Memorial Children's Hospital) in the Western Cape. To determine whether there is a relationship between gender and dental anomalies associated with unilateral cleft lip and palate.

Materials and Methods

A retrospective cross-sectional study assessing the hospital records of patients diagnosed with unilateral cleft lip and palate. Panoramic radiographs of 93 patients with unilateral cleft lip and palate (UCLP) aged 8 to 14 years were evaluated. Missing and supernumerary teeth were also quantified on the cleft and noncleft side and in the maxilla and mandible. Ectopic teeth, peg shaped laterals, and Crown and root malformations were quantified. Statistical analysis first comprised description of the frequency and types of dental anomalies. Chi-square analysis was used for comparisons of dental anomalies, in addition to specific dental anomalies in relation to gender.

Results

There were no substantial differences in distribution by gender, of the 93 patients with UCLP, 47 (50.54%) were males and 46 (49.46%) were females. Regarding distribution by cleft side, the left side was more frequently affected (69.9%) in both male and female patients, compared with 30.1% found on the right side. The most affected tooth was the cleft lateral, which was missing in 35.48% of the participants, while the non-cleft lateral was absent in only 3.23% and bilateral laterals were missing in 10.75%. Supernumerary teeth were found in 7.53% of the participating individuals and the most affected tooth was the cleft lateral. The cleft

lateral was judged to be peg shaped in 27.96% of the individuals and additional 3.23% showed other malformations. The lateral was positioned distally to the cleft in 50.54%, mesial to the cleft in 13.98%.

Conclusions

Almost all individuals with unilateral cleft lip and palate were found to have at least one dental anomaly. Agenesis was the most common dental anomaly in this overall study sample. Most of the dental anomalies are found in the cleft area. The suggestion therefore is that the effect on the dentition of the cleft disturbance is well localized.

DECLARATION

I **Vuyisile Solomon Gomba** hereby declare that *Analysis of dental anomalies in Patients with Unilateral Cleft Lip and Palate at Academic Hospitals in the Western Cape, South Africa* is my own work, that it has not been submitted before for any degree or examination at any university, and that all the sources I used or quoted have been indicated and acknowledged by complete references.

Vuyisile Solomon Gomba

Student no: 3203262

Signed: 

11th Day of November 2022

The work reported in this thesis was carried out in the Department of Orthodontics, Faculty of Dentistry, University of the Western Cape, Tygerberg, South Africa.

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DEDICATION

I would like to dedicate this thesis to my inspiration and source of strength my father Mr. Mhlupheki Gomba and to my most caring, loving, and kindest mother Mrs. Maitumeleng Gomba. Your prayers for me are what sustained me thus far, your constant encouragement for learning and to pursue my dreams have always lifted me up even in the most difficult days. I hope I have made you proud.

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Chapter 1

1. Introduction

Oral clefts are among the most recognized and widespread craniofacial anomalies which occur in about 1:700 live births (WHO, 2006).

The prevalence rate in Africa estimated from hospital-based data ranges from 0.2/1000 live births in Ethiopia (Eshete *et al.* 2017), 0.8/1000 in Uganda (Kesande *et al.* 2014) and 1.7/1000 reported in Kenya (Khan, 1965). A community household survey in South East Ghana found an estimated prevalence of 6.3/1000 people with cleft lip and palate (Agbenorku *et al.* 2011). Hlongwa *et al.* 2019 reported the prevalence of cleft lip and palate was 0.3/1000 live births in the South African public health system, with regional differences from 0.1/1000 to 1.2/1000.

Most reports have indicated that 70% of cases of cleft lip and palate are non-syndromic and that the remaining 30% are associated of structural defects outside the cleft area (Schutte and Murray, 1999; Cobourne, 2004; Lidral *et al.* 2008). According to Dixon *et al.* (2011) non-syndromic clefts affect one out of every 700 live births, with ethnic and geographic differences. Individuals with a non-syndromic cleft lip and palate usually have different dental abnormalities with tooth form, size, and location (da Silva *et al.* 2008). The extent of these dental anomalies varies according to sex, ethnicity, and cleft type. For example, according to Rizell *et al.* (2019) study agenesis was the most prevalent dental anomaly, followed by presence of peg shaped laterals, supernumerary teeth, ectopic eruption, transposition and infraocclusion.

Patients with cleft lip and palate have usually been diagnosed with impaired dental development and tooth eruption, and abnormalities such as hypodontia, supernumerary teeth, hypoplasia, and disorders in tooth size and form (Ranta, 1986). Early report from Bohn in 1950 was among the first to document the occurrence of missing teeth in cleft lip or cleft palate patients.

He examined lateral incisor anomalies in cleft lip and palate cases. 63 patients aged 3 to 7 years were analyzed for lateral incisor abnormalities in terms of their number. The author found that there was a higher prevalence of supernumerary deciduous lateral incisors and a decrease of permanent lateral incisors in the cleft lip group. There was a low prevalence of supernumerary deciduous lateral incisors, compared to the high number of absent permanent lateral incisors in the cleft lip and palate group.

Various forms of dental anomalies are typical in children with cleft lip and/or palate such as supernumerary teeth or missing teeth, microdontia, tooth rotation, hypoplasia, transpositions, and root deviation, generally found on the cleft side (Sá *et al.* 2016). Tsai *et al.* (1998) found that both primary and permanent dentitions can be affected, and that the dental anomalies are more prevalent on the cleft side of the maxilla.

Clefts of the lip may occur due to the distortion of facial development which prevents interaction between the palatine shelves when they swing into the horizontal position. Clefts of the palate may result from the following: failure of the shelves and septum to contact each other because of a lack of growth or because of a disturbance in the mechanism of shelf elevation or failure of the shelves and septum to fuse after contact has been made because the epithelium covering the shelves does not break down or is not resorbed (Nanci, 2018).

Several genetic and environmental factors have been identified as being responsible for the etiology and pathogenesis of clefts, either collectively or through the association of multiple biological pathways (Yaqoob *et al.* 2013). In 2013, Seo *et al.* evaluated 126 Korean non-syndromic cleft patients and investigated three single-nucleotide polymorphisms (SNPs) of Msx1 and ten SNPs of PAX9 and found an association between SNPs of Msx1 and Pax9 genes and the risk of tooth agenesis in non-syndromic cleft patients. They concluded that genetic disturbances of Msx1 and Pax9 genes were associated with tooth agenesis within and outside the cleft area.

1.1 Problem statement

Dental anomalies in patients with clefts have been shown to have a higher prevalence than in patients without cleft. They include variations in tooth number and position, and reduced tooth dimensions, most of which are localized in the area of the cleft defect. Dental anomalies, especially congenitally missing teeth, are an additional complication for the treatment planning of cleft patients. Cleft lip and palate is accompanied by a wide variety of dental anomalies, which also have a long-term impact on the patient's facial anatomy, self-esteem and burden of care. The maxillary lateral incisors are the most susceptible to dental anomalies within the cleft region (Cassolato *et al.* 2009).

When the lateral incisors in cleft area are missing or extracted, replacement is not required in most cases and the space in the cleft area can be closed by orthodontic treatment. When the space of upper lateral incisor in the cleft area remains open, a removable prosthesis, fixed bridge or an implant can be provided depending on the periodontal condition and integrity of the alveolar ridge. However, because of age changes in tooth position, implants in young children are not recommended for the following reasons: the osseointegrated implant becomes ankylosed and cannot change position as compared to the adjacent natural teeth. Progressive infraocclusion may occur after some years due to continuous eruption of adjacent teeth, including in adults and elderly patients. The normal uprighting of maxillary and mandibular incisors that generally occurs from adolescence to adulthood cannot be matched by implant crowns. For these reasons, orthodontic space closure is the recommended treatment result that would reflect a natural dentition over a long period of time.

Both maxillary permanent lateral incisors located mesial and distal of the cleft area are helpful in maintaining surrounding bone and therefore, must be preserved until an alveolar bone graft is performed. Extracting the abnormal lateral incisor around the time of alveolar bone grafting allows the canines to migrate and erupt forward through the grafted area, providing an improved bony environment that facilitates orthodontic and prosthodontic treatment and improves the stability and health of the periodontium (Hinrichs *et al.* 1984).

The treatment of missing lateral incisors involves either maintaining the space for a resin-bonded bridge or a tooth-supported fixed prosthesis, autotransplantation or an implant-supported crown. If space is to be closed, there are two basic treatment options for patients with this problem, firstly, to move the maxillary canines mesial and reshape them to simulate missing or extracted lateral incisor. Secondly, to position the canines in a Class I relationship and restore the missing tooth structure by increasing the size of the peg-shaped lateral incisor or replacing the missing lateral incisor with a fixed or removable prosthesis (Miller, 1987).

The advantage of space closure is that the treatment can be finished at an earlier age and that no artificial material needs to be inserted into the jaw. Supernumerary teeth need to be extracted for the following reasons, to allow orthodontic alignment of the teeth and also if their presence would compromise alveolar bone graft and implant placement.

Knowledge of the occurrence of these anomalies in individuals with cleft lip and palate will assist orthodontists to predict malocclusion and other problems when coping with these cases in the clinic. Orofacial clefts and associated malocclusion contribute significantly to long-term disability in children, as well as considerable emotional and financial burden for affected individuals and families.

Chapter 2

2.1 Literature review

2.1.1 Prevalence

Congenital abnormalities (CA) account for 2-3% of child mortality and childhood morbidity (WHO, 2003). Roughly 1% of babies are born with these syndromes or multiple anomalies; craniofacial anomalies (CFA) form a part of the disorder. These syndromes consist of several malformations that are considered to have an association either etiologically and /or pathogenetically. Syndromes with features of cleft lip and/or cleft palate are of concern in etiological and pathogenetic causes and are believed to be syndromic in 30% of cleft cases, while non-syndromic clefts make up for the remaining 70% (WHO, 2001).

Oral clefts (OCs) are thus among the most recognized and widespread CFAs make about 1:700 live births. CFAs occurs in 1 out of every 1,600 neonates in the United States of America (USA), rather than cleft lip and palate, which includes jaw deformities, malformed or absent teeth, facial or cranial ossification defects and facial asymmetries. Clefts occur more often among Asians than among Africans (WHO, 2001).

Patients with cleft lip and palate have been commonly reported to have been diagnosed with impaired dental growth and eruption, and dental anomalies such as hypodontia, supernumerary, hypoplasia, and disorders in tooth size and form (Ranta, 1986).

Earlier report by Bohn (1950) was among the first to document the occurrence of congenitally missing teeth in cleft lip (CL) or cleft palate (CP) patients. In cases of cleft lip and cleft palate, he examined lateral incisor anomalies. 63 patients aged 3 to 7 years were clinically and radiographically analyzed with respect to their number for lateral incisor abnormalities. In the group with cleft lip and palate 52% of the absent lateral incisors were observed, preceded by 12% in the cleft lip.

Hlongwa *et al.* (2019) established an epidemiology and clinical profiling of people with cleft lip and/or palate (CLP) using specialist academic hospitals in South Africa's public health sector. The researchers reviewed 699 records of CLP individuals. They found that the predominance of CLP was 0.3 per 1000 live births in the South African public health system, with regional differences from 0.1/1000 to 1.2/1000. The results were the following 35.3% had cleft palate; 34.6% had cleft lip and palate; 19% cleft lip and other 2% had cleft anomalies. 47.5% of the total CLP results were male, and 52.5% were female, and this discrepancy was statistically significant. Most clefts were reported on the left (35.5%) for males and female palate (43.4%), with unilateral cleft lip and palate (53.3%) primarily for males.

Moodley *et al.* (2018) reviewed the records of 383 patients with cleft lip and/or palate which were obtained from the Sefako Makgatho Health Sciences University, Medunsa Oral Health Center, Department of Orthodontics between 2003 and December 2014. 47% of patients had unilateral clefts, 23% had bilateral clefts and 30% had palate clefts. Clefts on the left side accounted for 56.4% and on the right side for 43.58%, making clefts on the left side more prevalent. Clefts of the lip and palate accounted for 45.8%, those affecting the lip for 24.2% and those affecting only the palate for 30%.

Manyama *et al.* (2011) undertook a study to establish the frequency, laterality, sex and geographical distribution of orofacial clefts and their associated congenital anomalies among patients attending Bugando Medical Centre in Mwanza, Tanzania, from 2004 to 2009. A total of 240 orofacial cleft cases were seen during this period. Isolated cleft lip was the most common cleft type followed closely by cleft lip and palate (CLP). This is a departure from the pattern of clefting reported for Caucasian and Asian populations, where CLP or isolated cleft palate is the most common type. The distribution of clefts by side showed a statistically significant preponderance of the left side (43.7%) = 92.4, $p < 0.001$), followed by the right (28.8%) and bilateral sides (18.3%). Unilateral orofacial clefts were significantly more common than bilateral clefts; with the left side being the most common affected side.

Dreise *et al.* (2011) study was to estimate the need for resources for cleft repairs in Uganda by determining the overall incidence of oral-facial clefts and the ratio of isolated cleft lip to isolated cleft palate to cleft lip and palate. A 1-year prospective study was implemented in seven hospitals and health centers with maternity units in and around Kampala. All live babies were examined for cleft lip and/or palate at birth. Over the course of 1 year (February 1, 2008, to January 31, 2009), 26,186 babies were delivered. Nineteen babies had a cleft lip and/or palate, giving an incidence of 0.73 in 1000; 12 of the 19 babies (63.2%) had a cleft lip and palate, six (31.2%) had an isolated cleft lip, and only one (5.3%) had an isolated cleft palate. The ratio of boys to girls was 1.1:1. The incidence of clefts in this study was 0.73 in 1000.

Morrison *et al.* (1985) investigated the incidence of cleft lip and palate in the Western Cape province, South Africa. Using hospital data, they found that between 1983 and 1984; 52 children were born with a cleft, of these 3 were Black, 43 Coloureds and 6 were Caucasians. They hence reported a high incidence in coloureds 1.4/1000 birth.

Eshete *et al.* (2017) investigated the prevalence and incidence of orofacial clefts in Ethiopia. They found that the incidence rate estimated from the total number of affected children during the study period (N=8232) is 0.44/1000 live births. The prevalence rate is 0.20/1000 and this was estimated using the number of total population in 2013 (N= 88,703,914). There is a significant difference in frequency between bilateral CLP (26.9%) versus unilateral CLP (73.1%) (P<0.0001). In Ethiopia, a rate of 1.4 in 1000 live births was reported in a hospital-based study.

2.1.2 Hypodontia

Wong *et al.* (2012) studied the occurrence of the various forms of dental abnormalities in Southern Chinese children with CLP and correlated the findings with a sample of non-cleft children matched by age and gender. The authors found that 57.6% of the children with CLP had hypodontia, 10% of the sample had

hyperdontia, taurodontism accounted for 8.7%, 0.8% a double tooth, 1.30% dens evaginatus, and 42.4% had microdontia in the permanent dentition.

Research undertaken by Rullo *et al.* (2015) to determine the presence of various forms of dental abnormalities in children with cleft lip, unilateral cleft lip palate and bilateral cleft lip and palate. They observed that in 40% of the sample the lateral incisor on the side of the cleft was congenitally absent, while in 4.4% of cases there was second premolar which failed to develop. 30% of patients had supernumerary teeth in the incisor region, while ectopic tooth eruption occurred in 18.9% of the study. Rotation of the lateral or central incisors was observed in 31.1% of the study, while shape abnormality, lateral incisor microdontia and enamel hypoplasia were detected in 25.6%, 5.6%, and 18.9% of cleft patients respectively.

Al-Kharboush *et al*. (2015) reported that missing teeth were the most seen in their study constituting 46.5%, 31.6% had microdontia, 10.4% had ectopic eruptions, 9% had supernumerary teeth and with a few patients (2.4%) having macrodontia.

Shetty *et al.* (2013) assessed the occurrence of incisor abnormalities in 113 untreated UCLP patients. Their findings were as follows: 48.7% of lateral incisors on the left-hand side were missing, 22.1% lateral incisors on the right had rotations, and right lateral incisors which were missing made up about 21.2% and central incisors that were rotated recorded a % of 18.6.

In a sample of Jordanian participants, the incidence of dental anomaly in CLP patients was higher than in normal subjects. About 66.7% of patients presented with missing upper lateral incisor as the most often affected tooth. Other results included microdontia (37%), taurodontism (70.5%), transposal or ectopic teeth (30.8%), dilaceration (19.2%), and hypoplasia (30.8%) (Aljamal *et al.* 2010).

Yatabe *et al.* (2013) measured the presence of upper lateral incisor agenesis with and without the Simonart's band in the cleft area in patients with unilateral cleft lip and palate. Those patients with the Simonart's Band had a greater incidence of maxillary lateral incisors further away from the cleft region relative to patients without Simonart's band.

Bartzela *et al.* (2013) performed a study to identify trends of dental agenesis and their relative incidence in patients with complete unilateral cleft lip and palate (CUCLP). Their findings were as follows; 8.7% of patients had missing upper lateral incisor in the non-cleft area. The most prevalent symmetric patterns were the lateral incisors (5.2%) and the second premolars (0.9%) when comparing the mandible and maxilla. Agenesis of a single tooth was detected in 48.7% of patients and absence of tooth further away from the cleft was seen in 20.9% of patients.

The upper lateral incisor in the cleft area was frequently missing (39.1%), followed by the maxillary lateral incisor (8.7%) and the mandibular second premolar (7.8%) in the non-cleft region. Thirteen various types of dental agenesis have been reported. In each of these types, maxillary and/or mandibular second and/or first premolars were involved.

Mikulewicz *et al.* (2014) assessed the incidence of second premolar hypodontia in Polish children with clefts from south-west Poland. The sample of 120 participants (78 boys and 42 girls) with unilateral cleft lip and palate, 82 had left UCLP and 38 had right UCLP. Nineteen subjects (15.8%) had second premolars congenitally missing: 13 males (16.7%) and 6 females (14.3%) respectively. 33(6.9%) teeth on average were missing, the male group accounting for (7.7%) 24, while the remaining 9 (5.4%) were the females. In the upper arch, eighteen congenitally absent second premolars (7.5%) and 15 in the lower arch (6.2%) were found.

Camporesi *et al.* (2010) did a study to determine the predominance of tooth abnormalities in the number, size, and shape of deciduous and permanent maxillary dentition in children affected by unilateral (UCLP) or bilateral (BCLP) cleft of the lip and palate and correlate it with the occurrence of dental anomalies in the non-cleft control group (NCLP). The incidence for missing deciduous lateral incisors in UCLP subjects was 8.1% and 27.9% for permanent lateral incisors. In 5.4% of UCLP subjects and 8.8% of BCLP sample the second premolar was absent.

Baek and Kim (2007) evaluated the differences in the pattern of missing maxillary lateral incisor (MLI) and maxillary second premolar (MSP) in a Korean sample with unilateral cleft lip and alveolus (UCLA) and unilateral cleft lip and palate (UCLP). UCLP patients had 2.98 times more missed MLIs and 1.80 times more missed MSPs than UCLA patients, depending on the association between the congenital missing teeth form and the cleft type. The MLI was congenitally missing more in boys than in girls, although the MSPs showed the opposite tendency. Boys had a higher frequency of congenital missing MLIs and MSPs on the cleft side than girls. Girls, on the noncleft side and both sides, had a higher frequency of congenital missing MLIs and MSPs than boys.

Dewinter *et al*. (2003) evaluated the presence of dental abnormalities and examined the periodontal condition of the teeth adjacent and in the cleft area of 75 individuals with UCLP before, during, or after a long-lasting orthodontic and surgical treatment. Their study indicated 58.6% of the patients had missing lateral incisors, of those teeth found outside the cleft area such as the second premolar and the lateral incisor made up 27.2%. They therefore discovered that 32% of the central incisors in the cleft area had crown deformities.

Ribeiro *et al.* (2003) reported that 42 patients (20.7%) presented with hypodontia outside the cleft area and in 32 patients on the non-cleft side (15.7%). The lateral incisor on the cleft side was missing in about 101 patients (49.8%), while there was an absence of same tooth on the non-cleft side in 22 patients (10.9%). The maxillary second premolar was the most commonly missing tooth, followed by the mandibular second premolar, maxillary first premolar, and mandibular first premolar.

2.1.3 Supernumerary teeth

Rizell *et al.* (2019) reported that 16.9% of the participants had supernumerary teeth. Aljamal *et al.* (2010) study found similar results compared to Rizell *et al.* (2019) in that 16.7% of patients had supernumerary teeth, whereas a study by

Rullo *et al.* (2015) reported a higher prevalence rate that 30% of patients had supernumerary teeth in the incisor region.

2.1.4 Lateral incisor position and size

Rizell *et al.* (2019) studied dental abnormalities in 448 children born with UCLP at age eight, which was used in the Scandcleft Randomized Clinical Trials. Missing teeth were observed in 52.6%. In 43.7% of the cases there was absence of the lateral incisor on the cleft side and 45% were peg shaped laterals. The incidence of ectopic eruption was 14.6%, mostly involving the first maxillary molars, while 4.3% of the individuals had transposition of teeth. Furthermore, 7.2% of the participants had one or more primary molars which were infraoccluded.

Tsai *et al.* (1998) noted a variation in the prevalence of maxillary lateral incisors throughout the primary and permanent dentition. In the primary dentition, the lateral incisor was located distal to the alveolar cleft (82.4%), followed by the absence of the cleft side maxillary lateral incisor (9.9%), the presence of one tooth on each side of the alveolar cleft (5.5%), and the lateral incisor was located mesial to the alveolar cleft in 2.2% of the sample.

The absence of the maxillary lateral incisor on the cleft side (51.8%) was the most common pattern in the permanent dentition, followed by lateral incisor located distal to the alveolar cleft (46%), lateral incisor located mesial to the alveolar cleft (1.5%), and the presence of one tooth on each side of the alveolar cleft (0.7%).

Ribeiro *et al.* (2003) conducted a study to establish the incidence of the permanent lateral maxillary incisor and the occurrence of hypodontia outside of the cleft region, in a given population with complete unilateral cleft lip and palate. The permanent lateral incisor on the cleft side was found in 102 cases (50.2%) and was more frequently located on the distal (76.5%) side of the cleft than the mesial (23.5%). Pertaining to the shape of the tooth, 92.2% were found to be conical in comparison to its antimere.

Menezes and Vieira (2008) examined the radiographic and medical records of 146 participants in the CLP. At least one dental anomaly was observed outside the cleft region in 47 (32.19%) subjects, and subjects with complete CLP had more dental abnormalities than those with partial CLP. The prevalence of mandibular premolar abnormalities was found to be higher in patients with cleft palate than in patients with CLP, the most affected being upper lateral incisors and premolars. In unilateral CLP cases, on the non-cleft side 12.5% had dental defects of the maxillary lateral incisor.

2.2 Rationale for study

The incidence of dental anomalies is significantly increased in children with cleft lip and palate compared to the general population. These conditions increase the need for multiple health care services throughout the lifespan of affected individuals. For example, additional surgeries, dental treatments, speech therapy, and psychosocial services are commonly needed and recommended throughout childhood, adolescence and can extend into early adulthood. The burden of care for children with cleft lip and palate can create a financial and emotional burden on affected families.

The mean length of orthodontic treatment for unilateral cleft lip and palate patients varies from 18-30 months and this can be explained by many factors such as the complexity of treatment related to the number of dental anomalies, the requirement of two or three courses of orthodontic treatment and the long distance that these patients often have to travel between their place of residence and the treatment center and this often results in many missed appointments due to access, travel and financial reasons, thereby extending the treatment time.

Patients with unilateral cleft lip and palate also have an unfavourable facial growth pattern which increases the orthodontic burden of care. Orthodontic treatment time is greatly increased in patients with unilateral cleft lip and palate compared to the general population, in which orthodontic treatment may require up to 3 stages of intervention, despite the existence of dental anomalies. These

include the interceptive orthodontic treatment in preparation for an alveolar bone graft, the comprehensive orthodontic treatment at about 12-13 years of age and a course of orthodontic treatment in preparation for orthognathic surgery for those with adverse facial growth.

The prevalence of dental anomalies in patients with unilateral cleft lip and palate in the Western Cape province of South Africa has not been reported. Access to such information is essential for successful orthodontic treatment planning. This information adds to the understanding of the complexity of orthodontic treatment which can also extend treatment times, thus adding to the overall burden of care.

The burden of care for patients born with a cleft include surgical, orthodontic, dental, speech therapy, hearing, psychosocial and as a consequence financial. It is important for all care providers to take every effort to reduce this burden from all these aspects. Therefore, this study seeks to provide data on the prevalence of dental anomalies associated with unilateral cleft lip and palate to assist efficient successful orthodontic diagnosis and treatment planning.

2.3 Study question

What are the different types and frequencies of dental anomalies associated with patients with unilateral cleft lip and palate receiving treatment at Academic hospitals (UWC Oral Health Center and Red Cross War Memorial Children's Hospital)?

2.4 Aim of study

The aim of this study was to determine the type and frequency of dental anomalies associated with patients with non-syndromic complete unilateral cleft lip and palate receiving treatment at Academic hospitals (UWC Oral Health Center and Red Cross War Memorial Children's Hospital).

2.5 Study objectives

2.5.1 To determine the different dental anomalies associated with unilateral cleft lip and palate.

2.5.2 To determine whether there is a relationship between gender and dental anomalies associated with unilateral cleft lip and palate.

2.5.3 To determine the frequency of each dental anomaly associated with unilateral cleft lip and palate.

Chapter 3

3 Methodology

3.1 Study design

This was a retrospective cross-sectional study from 01 January 2010 to 31 December 2021 assessing the hospital records of patients diagnosed with unilateral cleft lip and palate.

3.2 Study and setting

The study population comprised of clinical records of all the patients with UCLP who have attended at Academic hospitals (UWC Oral Health Center and Red Cross War Memorial Children's Hospital) from 01 January 2010 to 31 December 2021. Clinical records of 93 patients with UCLP were evaluated during the study including hospital files, panoramic and anterior occlusal radiographs were considered adequate.

3.3 Sampling method and sample size

We employed a convenience sampling method of the patient records of all the patients who were treated at UWC Oral Health Centre and Red Cross War Memorial Children's Hospital during the study period. Sample size estimation was not calculated as we planned to include all the subjects who satisfied the inclusion criteria.

3.4 Materials and Methods

A retrospective cross-sectional study was conducted at UWC Oral Health Center and Red Cross War Memorial Children's Hospital located in the Western Cape province, South Africa, to assess the hospital records of patients diagnosed with unilateral cleft lip and palate from 01 January 2010 to 31 December 2021. Most patients with orofacial clefts in the surrounding regions are usually referred to

these hospitals as it is they are two of the three centres that offer surgical expertise to repair orofacial clefts in the Western Cape province. Cleft lip and palate records were obtained from patient files in the Hospital's Departments of Orthodontics, Plastic Surgery and Medical Records. Because individuals with CLP attend these centers frequently over time, care was required to register each person just once to avoid duplications.

Patient file notes are usually written by medical officers and Plastic surgeons from the time of hospital admission to discharge. Age at presentation, sex, region of origin, type and laterality of the cleft were recorded. Additionally, presence of associated congenital anomalies or syndromes was recorded. Cleft lip and palate cases that lacked some of the above information (e.g., type of cleft, laterality) were excluded.

3.4.1 Inclusion criteria

All the patient records which satisfied the following inclusion criteria were included in this study:

- Patients must be aged between 8 and 14 years
- Selected patient records must be of good quality and acceptable diagnostic standards. Distortions are acceptable if the examination criteria are readable.
- Digital or scanned panoramic and anterior occlusal radiographs were used.

3.4.2 Exclusion criteria

- Syndromic patients
- Patients with an incomplete unilateral cleft lip and palate
- Patients with a Simonart's band
- Incomplete records
- Poor quality radiographs

Ninety-three individuals (47 males and 46 females) with a mean age of 11.2 years (8-14yrs) were finally included in this part of the project. Digital or scanned panoramic radiographs from the cleft area, taken at eight years of age to avoid the possibility that the tooth germs of the second premolars had not developed yet., were collected from the participating hospitals. The radiographs were obtained prior to orthodontic treatment and bone grafting. Occlusal radiographs were not obtained for all patients with UCLP in the present sample; as a result, they were thus eliminated from the analysis.

Parameters evaluated in all panoramic radiographs included the presence or absence of permanent lateral incisors, agenesis and supernumeraries of permanent teeth, shape and position of cleft lateral, ectopic eruption, transposition, infraocclusion, evident crown or root malformations and presence of agenesis in the mandible.

The evaluation was performed separately by 2 operators namely the researcher and an orthodontist with more than 20 years of experience evaluated all the dental panoramic radiographs. In the case of a divergence in outcomes, the operators re-examined and discussed the panoramic radiographs until they reached an agreement. Overall inter-observer agreement was calculated by comparing the findings of the 2 operators.

The dental anomalies investigated were defined as follows:

a. Abnormalities of tooth number

Diagnoses of hypodontia and supernumerary teeth in the cleft area were established according to the criteria reported by Damante (1972) as cited by da Silva *et al.* (2008). Supernumerary tooth was defined as any additional tooth mesial or distal to the cleft area in the presence of the lateral incisor. Outside of the cleft area, these anomalies were diagnosed according to the criteria of Garvey *et al.* (1999): hypodontia or tooth agenesis was diagnosed when the tooth or tooth bud was absent on radiographs, resulting in a deficient dental developmental series, and a supernumerary tooth was diagnosed based on the identification of an

additional tooth germ or calcification (beyond the normal dental developmental series) on radiographs in any region of the dental arch.

b. Abnormalities of crown morphology

Macrodontia and microdontia refer to teeth that are substantially larger and smaller, respectively, than the average normal size, or larger and smaller, respectively, than the contralateral homolog or a tooth in the sample group from the opposing arch (D'Souza *et al*. 2006). Microdontia also refers to a tooth that does not fill its space in the dental arch or appears small because of the absence of expected shape (D'Souza *et al*. 2006).

Microdontia of maxillary lateral incisor is called as "peg lateral", that exhibit converging mesial and distal surfaces of crown forming a cone like shape. The root on such a tooth is usually shorter than usual (Gupta, 2019).

c. Infraocclusion

Infraocclusion is a condition of tooth eruption insufficiency observed largely in human deciduous molars but noted also in other teeth, including permanent molars (Shalish *et al.* 2010).

Diagnostic criteria - A tooth was recorded in infraocclusion when its occlusal surface is 1 mm or more cervical to the occlusal plane of the fully erupted neighbouring teeth (Kurol, 1981).

d. Ectopic maxillary canines

Ectopic eruption of maxillary canines refers to canines that have erupted, but are displaced, or more importantly are still erupting but show signs of moving in an incorrect direction (Hudson *et al.* 2010).

e. Tooth impaction

Tooth impaction occurs when a tooth is prevented from reaching its normal position in the dental arch. This can be due to hard or soft tissue obstruction and/or an abnormal eruption pattern (e.g., after the expected time of eruption) (Thilander and Myrberg, 1973).

f. Tooth agenesis

A tooth is defined to be congenitally missing if it has not erupted in the oral cavity and is not visible in a radiograph at an age when it would be detected (Cobourne, 2017).

3.5 Data analysis

Statistical analysis first comprised description of the frequencies and types of dental anomalies. Chi-square analysis was used for comparisons of dental anomalies, in addition to specific dental anomalies in relation to gender. A p-value below 0.05 was considered to be statistically significant. The data were analyzed using the Statistical Package for Social Science Version 12.0 for Windows (SPSS, Inc., Chicago, Illinois, U.S.A.).

3.6 Data capturing

Once data capturing was completed, it was thereafter verified. Records of 10% of the patients were reassessed and re-entered at a later stage for reliability testing. Once the data capturing was complete, it was then printed in the form of Excel spread sheets. The data analysis was primarily of a descriptive nature. Categorical data was summarized by frequency counts and % calculations. Continuous data was summarized by sample size, mean, standard deviation, median, interquartile range, minimum and maximum values.

For assessing inter-examiner reliability, 93 radiographs were scored twice by 2 observers. Cohen's kappa was used to test reliability of the scores among the two raters. Intraexaminer reliability for identification of missing teeth and size and shape of lateral incisors was conducted by the primary investigator through random selection of a subsample of 10 patients 1 month after the initial identification. The kappa index and overall agreement for intraobserver consistency in evaluating the dental anomalies, ranging from 1 (perfect agreement) to 0.47 (moderate agreement) (Appendix D).

3.7 Ethical considerations

Ethical permission was granted by the Tygerberg hospital (NHRD Ref: WC_202204_013) and Biomedical Research Ethics Committee (BMREC) of the University of the Western Cape (Ref No. BM20/10/22) (Appendix A).

Chapter 4

4 Results

There were no significant differences in distribution by gender, of the 93 patients with UCLP, 47 (50.54%) were males and 46 (49.46%) were females. Regarding distribution by cleft side, the left side was more frequently affected (69.9%) in both male and female patients, compared with 30.1% found on the right side (Table 1). Figure 1 illustrates the distribution of patients according to gender and age.

Distribution of orofacial clefts according to gender and laterality

		Frequency (%)
Gender	Male	50.54
	Female	49.46
Laterality	Right	30.1
	Left	69.9

Table 1: Distribution of orofacial clefts according to gender and laterality

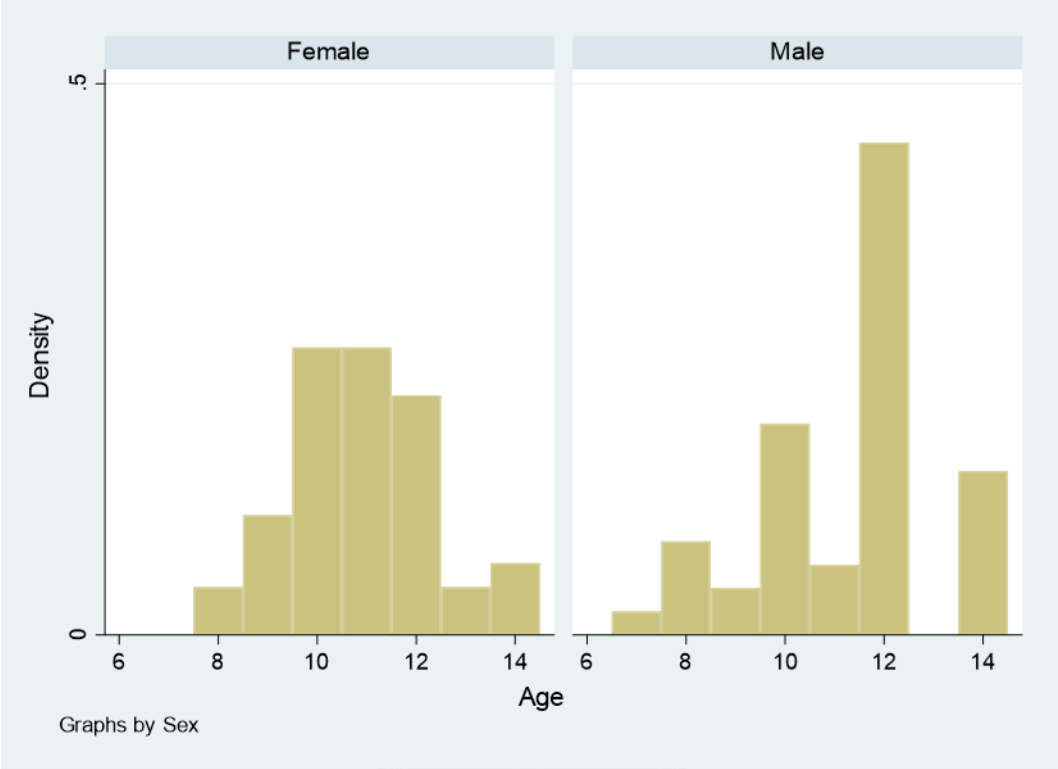

Figure 1: Distribution of patients according to gender and age

Most of the individuals with agenesis were missing only one single tooth (86.06%), but the number of missing teeth ranged from one to as many as four teeth. The most affected tooth was the cleft lateral, which was missing in 35.48% of the participants, while the non-cleft lateral was absent in only 3.23% and bilateral laterals were missing in 10.75% (Table 2). The second premolar was affected in 9.68% of the individuals with a range of one to four maxillary and/or mandibular teeth being absent (Table 2 and 3). Mandibular agenesis was observed in 6.45% of the sample.

Supernumerary teeth were found in 7.53% of the participating individuals and the most affected tooth was the cleft lateral (Table 2), these supernumerary laterals were located on the cleft side. A mesiodens was found in 3.23% of the participants (Table 2).

Numerary anomalies		Number of individuals n = 93	Frequency (%)
Agenesis	Agenesis		
	Cleft lateral	33	35.48
	Non-cleft lateral	3	3.23
	Bilateral maxillary laterals	10	10.75
	Maxillary central	2	2.15
	Maxillary/mandibular second premolar	0	0
	Maxillary/mandibular molar	6	6.45
Supernumeraries	Lateral (only)	7	7.53
	Mesiodens	3	3.23
	Mandibular incisor	0	0
	Lateral + mesiodens + 44	0	0
	Total of individuals with supernumeraries	10	10.76

Table 2: Prevalence of agenesis and supernumerary teeth in individuals with unilateral cleft lip and palate

Agenesis of second premolars		Number of individuals n = 93	Frequency (%)
Number of missing second premolars	One premolar	16	17.2
	Two premolars	5	5.38
	Three premolars	0	
	Four premolars	1	1.08
	Total	22	23.66
Location of second premolar agenesis	Agenesis of maxillary second premolars	9	9.68
	Agenesis of mandibular second premolars	6	6.45
	Agenesis of maxillary and mandibular second premolars		
	Total	15	16.13

Table 3: Agenesis of second premolars (number and location) in individuals with unilateral cleft lip and palate

In our sample, the cleft lateral was judged to be peg shaped in 27.96% of the individuals and additional 2.15% showed other malformations (Table 4). The lateral was positioned distally to the cleft in 50.54%, mesial to the cleft in 13.98%, and on each side of the cleft (i.e., where a supernumerary lateral was present) none was found in any of the cases.

	Number of individuals n = 93	Frequency (%)
Cleft lateral shape		
Peg shaped	26	27.96
Other malformation	2	2.15
Normal	32	34.41
Agenesis	33	35.48
Cleft lateral position		
Distal to cleft	47	50.54
Mesial to cleft	13	13.98
Lateral on each side of the cleft (i.e. present cleft side supernumerary)	0	0
Agenesis	33	35.48

Table 4: Prevalence of variations in shape and position of the permanent cleft lateral in individuals with unilateral cleft lip and palate

The prevalence of maxillary canine ectopia in our study was 16.13%, 3.23% for the first premolar and 1.08% for the second premolar. The prevalence of maxillary lateral incisors was found to be 3.23%. Maxillary canine/first premolar unilateral transposition was present in about 7.53% of the sample (Table 5).

	Eruption disturbances	Number of individuals n = 93	Frequency (%)
Ectopic eruption	Unilateral maxillary molar	0	0
	Bilateral maxillary molars	0	0
	Maxillary central	0	0
	Maxillary lateral	3	3.23
	Maxillary canine	15	16.13
	Mandibular molar	0	0
	Total	18	19.36
Reversible ectopic eruption	Unilateral maxillary molar	0	0
	Bilateral maxillary molar	0	0
	Total	0	0
Transposition	Maxillary canine/first premolar unilateral	7	7.53
	Maxillary canine/first premolar bilateral	0	0
	Other	0	0
	Total	7	7.53
Infraocclusion	One affected tooth	0	0
	Two affected teeth	0	0
	Three affected teeth	0	0
	Four affected teeth	0	0
	Six affected teeth	0	0
	Total	0	0

Table 5: Prevalence of ectopic eruption, transposition, and infraocclusion of primary molars in individuals with unilateral cleft lip and palate

In our investigation, incisal central crown abnormalities were also considered. The results of our investigation showed that cleft central microdontia (2.15%) and central malformation (34.41%). Our study showed 2.15% of the sample, had root abnormalities of the central incisor, while 3.23% of the sample had atypical tooth anatomy outside of the cleft (Table 6).

Other anomalies	Number of individuals n = 93	Frequency (%)
Dental anatomy/development		
Cleft central malformed	32	34.41
Cleft central atypical crown-root angulation	2	2.15
Cleft central microdontia	2	2.15
Atypical tooth anatomy outside cleft area	3	3.23
Position		
Inverted mesiodens or lateral	0	0
Other		
Condylar hypoplasia non-cleft side	0	0
Cleft area odontoma	0	0

Table 6: Prevalence of other dental anomalies in individuals with unilateral cleft lip and palate

Our investigation found that 2.15% of maxillary right first molars, 3.23% of maxillary left first molars, 6.45% of mandibular left first molars, and 4.3% of mandibular right first molars were missing due to extraction.

4.1 Statistical analysis

Statistical analysis first comprised description of the frequencies and types of dental anomalies. Chi-square analysis was used for comparisons of dental anomalies, in addition to specific dental anomalies in relation to gender. All radiographs were scored by two observers. For assessing inter-examiner reliability, 93 radiographs were scored twice by 2 observers. Cohen's kappa was used to test reliability of the scores among the two raters. Inter-examiner reliability scores were high (almost perfect agreement) for the evaluation of cleft

location, agenesis, and central incisor anomalies, lateral incisor anomalies, Cleft lateral position, ectopic eruption, supernumerary teeth, substantial agreement for agenesis of maxillary premolars. The results revealed very high inter-examiner reliability, ranging from 1 (perfect agreement) to 0.64 (substantial agreement).

The kappa index and overall agreement for intraobserver consistency in evaluating the dental anomalies, ranging from 1 (perfect agreement) to 0.47 (moderate agreement) (Appendix D).

Chapter 5

5 Discussion

Given the global prevalence of clefts, it is critical for the multidisciplinary health care team to be familiar with dental abnormalities to provide appropriate orthodontic and surgical treatment to these children and adolescents.

The current study focused its attention on dental anomalies such as agenesis, peg-shaped teeth, ectopic eruption, supernumerary teeth, and crown and root malformations. The study focused on children from 8 to 14 years of age mainly because sometimes premolar tooth buds are not visible on radiographs at younger ages.

The use of panoramic radiographs was superior due to their low dose radiation, low cost and all the teeth can be examined at the same time including maxillary and mandibular jaws and their structure.

Gender differences in the prevalence of oral clefts have been reported previously; in comparison with females, males are more often affected and show more severe clefting (Shapira *et al.* 2018; Christensen, 1999; Cooper et al., 2000).

Al-Kharboush *et al.* (2015) assessed pre-treatment records of 184 subjects with cleft lip and palate and their study indicated an absence of a gender-based difference in the prevalence of dental anomalies, which agrees with the findings of others (Ranta, 1983, 1986; Shapira *et al*. 1999; Ribeiro *et al*. 2003; Al-Kharboush *et al*. 2015). The current study found a minor difference between males and females, with a prevalence of 50.54% and 49.46%, respectively.

5.1 Cleft location

A 2019 study by Hlongwa and co-workers analysed 699 records of treated individuals with CLP and reported left side dominated the prevalence of clefts in unilateral CLP for both genders (Hlongwa *et al*. 2019). Patients exhibited a much

greater frequency on the left side, according to Baek and Kim (2007). Their sample included 90 patients with UCLA and 204 patients with UCLP. Their findings revealed that 67.4% of patients with unilateral cleft lip and palate had left-sided clefts. In our study, we observed that majority of clefts were left sided, which concurred with the findings from previous studies.

5.2 Congenital missing teeth

General population has a prevalence ranging from 0.027% to 10.1% in congenitally missing teeth, depending on race and geographic location (Baek and Kim, 2007). Some researchers indicated that the maxillary lateral incisors are the most frequently missing teeth in the general population, while others reported that it is the mandibular second premolars are the most frequency, followed by the maxillary lateral incisors with 2.2% frequency (Ranta, 1986; Shapira *et al*. 2000; Rullo *et al.* 2015; Tortora *et al*. 2008).

In children with CLP, in both deciduous and permanent dentition, the maxillary lateral incisors were reported to be the most commonly missing teeth in the cleft region and the upper second premolars were reported to be more frequently missing than the normal population. Congenitally missing teeth were seen more unilaterally than bilaterally, but in second premolar teeth; the bilateral absence was found to be 1.5 times more often than unilateral absence (Rullo *et al.* 2015; Lekkas *et al*. 2000; Vieira *et al*. 2008).

A study by Ranta (1986) stated that the maxillary lateral incisor is the most frequently missing tooth in the cleft area, followed by the maxillary second premolars (Ranta, 1986).

A study by Wu and co-workers (2011) reported that the severity of the cleft increases the likelihood of lateral incisor agenesis. Dental agenesis is more common on the left side, independent of cleft side. This could be explained by overlapping etiopathogenetic factors in cases of dental agenesis and clefts.

Individuals with CLP present with substantially more dental anomalies, even outside the cleft area, than do individuals without clefts in different prevalence. Shapira *et al.* (1999; 2000) reported that hypodontia of both the maxillary lateral incisors and second premolars were more common on the left side, which also had a higher frequency of clefting. In our study, the prevalence of missing cleft laterals was 35.48%, non-cleft laterals were 3.23%, bilateral laterals were 10.75%, and second premolars were 9.68%. This finding has been explained by the proximity of the cleft to the lateral incisor region, which may strike and divide the primordial tissue related to the developing lateral incisor field (Shapira *et al.* 1999). Similarly, Baek and Kim (2007) also found considerably lower prevalence of hypodontia in the non-cleft side of these patients.

The interaction of MSX1 and PAX 9 appears to be involved in tooth agenesis in humans. This shows that the same etiological factors may be responsible for both cleft formation and congenital missing teeth in affected children (Seo *et al.* 2013).

Inadequate tissue in the medial nasal and/or maxillary processes during embryological development may result in the absence of lateral incisors mesially and/or distally. The high occurrence of agenesis outside the cleft area, on the other hand, suggests a common genetic basis for hypodontia and clefts (Vieira, 2003).

Rizell *et al.* (2019) study revealed the number of individuals exhibiting agenesis of one single or multiple teeth were 52.6%. In addition, agenesis of premolars was recorded in 18.1% of the participants in the Scandcleft trials, which is considerably higher compared to our findings.

Rizell *et al.* (2021) reported that individuals born with UCLP and agenesis of two or more maxillary teeth had a more unfavorable sagittal intermaxillary relationship at 8 years of age compared to the group with full dentition or agenesis of only one maxillary tooth.

5.3 Mandibular teeth agenesis

According to Bartzela *et al*. (2010), dental problems associated with clefts do not appear to be limited to the maxilla because the frequency of agenesis of lower second premolars was reported three times greater than in the normal population (2.9% - 3.2%). Tooth agenesis was more common in the maxillary lateral incisors and mandibular second premolars. The relationship between quadrants with tooth agenesis and quadrants without tooth agenesis suggests that tooth agenesis is a genetically regulated aberration connected to the orofacial cleft process rather than a disruptive osseous defect. The frequency of mandibular teeth agenesis in our study was 6.45%.

Ranta (1986) suggested that specific surgical treatments, such as early periosteoplasty or prenatal hard palate closure, could contribute to the increased occurrence of tooth agenesis in patients with CLP (Ranta, 1986). Although, other investigations have concluded that surgical interventions have little influence on tooth agenesis (Rullo *et al.* 2015; Wu *et al.* 2011; Bartzela *et al.* 2010).

5.4 Shape and position of cleft lateral

The permanent lateral incisor on the cleft side is also the most malformed tooth in the permanent dentition, frequently exhibiting some degree of size and shape abnormalities (Vichi and Franchi, 1995). It is often observed to be microdontic or peg-shaped (Lai *et al.* 2009; Tan *et al.* 2018). According to Suzuki *et al.* (1992) most cleft-side permanent lateral incisors are conical in shape (Suzuki *et al.* 1992).

The proportion of peg laterals in the general population ranges from 0.66% ~ 4% (Thilander and Myrberg, 1973). Ranta (1982) revealed that the permanent upper lateral incisor on the cleft side is usually aberrant in form.

Slayton *et al.* (2003) studied 120 subjects to determine whether the candidate genes previously studied in subjects with cleft lip, cleft palate, or both are

associated with hypodontia outside the region of the cleft and recorded the lowest proportion (12%) in comparison to our findings (Slayton *et al.* 2003).

The current study found a prevalence of 27.96% peg shaped laterals, 34.41 % of the cleft laterals were normal shape, and other abnormalities such as abnormal shape accounting for 2.15 %, however Rizell *et al.* (2019) found that the cleft lateral was peg shaped in more than 45% of the individuals in their sample. In 4% of cases, the lateral had other developmental abnormalities. Ribeiro *et al.* (2003), on the other hand, found a substantially larger % of peg-shaped laterals (92.2%).

The lateral incisor bud is frequently disturbed as it approximates the region of the dentoalveolar cleft, leading to alteration in size, shape (peg, conical teeth), time of formation and eruption (Cassolato *et al.* 2009).

5.5 Position of cleft lateral

When the permanent maxillary lateral incisor is present in individuals with CLP, it is commonly found on the distal side of the cleft (Tan *et al.* 2018). This tooth is frequently shown to be delayed in formation and eruption when compared to the antimeric lateral incisor on the non-cleft side (Lai *et al.* 2008; Tan *et al.* 2012; Ribeiro *et al.* 2003). According to our findings, 50.54% of lateral incisors were found distal to the cleft, whereas 13.98% were found mesial to the cleft. Throughout prenatal development, the medial part of the maxillary process delivers material to the distal regions of the future premaxilla. Evidence suggests that incisive sutures develop distally from the location where the medial nasal prominence and the maxillary process fuse (Lisson and Kjaer, 1997). These hypotheses help to explain the common finding of a lateral incisor distal to the alveolar cleft.

The location of the maxillary lateral incisors influences the position of the adjacent teeth. For example, lateral incisors that are mesial to the cleft decrease the amount of space in the premaxillary segment, increasing the likelihood that the central incisors will be rotated. When the lateral incisor is distal to the cleft, it can direct the eruption of the adjacent canine (Lai *et al.* 2009).

According to Tsai *et al.* (1998), following an evaluation of both 91 cases comprising of permanent and primary dentition, found that most primary lateral incisors, as well as their permanent successors, are distal to the cleft. This means that if the permanent lateral incisors are present, they will most likely be located distal to the cleft and the canines will most likely erupt normally.

5.6 Supernumerary teeth

The presence of a supernumerary tooth in the cleft area has been identified as the second most prevalent dental abnormality after agenesis (Ribeiro *et al.* 2003*).* Supernumerary teeth have been found in various populations with frequencies ranging from 0.1 to 3.8% (Baccetti, 1998).

The etiology of the anomaly is not entirely understood. The process of tooth development involves a complicated interaction between epithelial cells and ectomesenchyme derived from neural crest cells through chemical signalling. Disruption in this process results in variation in tooth number, amongst other dental anomalies (Fleming *et al.* 2010).

Supernumerary teeth might be normal in shape or smaller in size, have an aberrant crown or root morphology, and be at a different developmental stage than the rest of the teeth.

Variations in the morphology of supernumerary teeth include normal shape and size, normal shape and reduced size, and conical shape. Supernumerary teeth can be unilateral or bilateral, and they can be found in the anterior maxilla and mandibular premolar areas.

In the general population, supernumerary teeth are found in just 0.1-3.2% of subjects in the permanent dentition (Brook, 1974), and much less often in the primary dentition at 0.2-0.6% (Poyry and Ranta, 1985).

In the population with cleft, supernumerary teeth are common and are found at a rate ranging from 0.3% in the primary dentition and 1.2 - 21.3% in the permanent dentition (Tortora *et al.* 2008; Pegelow *et al.* 2012).

Suzuki *et al*. (2017) surveyed primary and permanent dental anomalies in 1724 patients with CL/P (905 males and 819 females). For primary maxillary dentition, supernumeraries were observed in 17.7% of the participants with cleft lip and/or palate, and 5.7% for permanent maxillary dentition (Suzuki *et al.* 2017).

Another possibility is that the cleft lengthens the precanine region of the oral epithelium, resulting in an expansion of the dental lamina that can develop into a supernumerary tooth. The division of the lateral incisor tooth bud, which is located across the clefted nasopalatal sulcus, might potentially result in the creation of an additional tooth (Kim and Baek, 2006). Non-fusion of the nasal and maxillary fields, as well as a potential post-fusion rupture of the cleft in the lateral incisor area, might result in tooth germ splitting (Mangione *et al.* 2018).

Supernumerary teeth develop because of nonfusion of the nasal and maxillary process and dental epithelia separation (Hovorakova *et al.* 2006). The higher prevalence of supernumerary lateral incisors in patients with CLP is due to the lateral incisor tooth bud's proximity to the cleft, resulting in a greater susceptibility to division or modification of the tooth bud or separation of the epithelial remnants (Lidral and Reising, 2002; Millhon and Stafne, 1941).

The most affected tooth in Scandcleft's study on children with unilateral cleft lip and palate was the cleft lateral (Rizell *et al.* 2019). In our study, supernumerary lateral incisors were identified in 7 (7.53%) of 93 participants, with mesiodens reported in 3.23% of 93 subjects. These findings are consistent with the findings of Tortora *et al*. (2008), who reported that 7.3% of UCLP had supernumerary lateral incisors and 1.2% had supernumerary central incisors. They investigated anomalies in tooth structure, location, and eruption pattern in patients with unilateral and bilateral cleft lip and/or palate. They examined 87 panoramic radiographs of individuals with UCLP and BCLP to find supernumerary teeth.

In contrast to our study, Rizell *et al.* (2019) stated that supernumerary teeth were discovered in 16.9% of the participants, with the cleft lateral being the most often affected tooth. A supernumerary lateral was detected on the cleft side in 14.3% of the patients, on the non-cleft side in 0.5% of the cases, and bilaterally in 0.5% of the individuals, 1.2% of the subjects had a mesiodens.

5.7 Central crown malformations

In our investigation, incisal central crown abnormalities were also considered. Disturbed enamel formation in the permanent teeth is most seen in the central incisor on the cleft side (Maciel *et al*. 2005). Tortora *et al*. (2008) found that in their UCLP sample, the percentage of central incisors with crown malformations on the cleft side was 15.8%, compared to a substantially lower rate (4.9%) outside the cleft side. Brattstrom and McWilliam (1989) study, 59.4% and 19.8% of patients with UCLP had normal central incisors and normal lateral incisors in the cleft area, respectively. Our findings showed that cleft central microdontia (2.15%) and central malformation (34.41%), which is comparable to the findings of Dewinter et al. (2003), who reported a higher percentage of crown abnormalities (32%).

5.8 Root Malformations

Dewinter *et al.* (2003) evaluated the frequency of dental abnormalities and periodontal condition of 75 patients with UCLP before, during, or after orthodontic and surgical treatment. The authors reported 10.6% of the UCLP patients as having root abnormalities of the anterior teeth in the cleft area. A study by Bohn (1963) reported that the root of permanent central incisor on the cleft side was shorter compared to the permanent central incisor on the noncleft side.

Tortora *et al.* (2008) examined central and lateral root abnormalities in 116 Caucasian nonsyndromic patients, 87 patients with UCLP, and 29 patients with BCLP. The central incisor had a root malformation prevalence of 5.1% in the cleft area and 4.9% outside of the cleft area. Interestingly, in their sample, the percentage of lateral incisor root malformations was inverted: 1.2% in the cleft area and 4.9% outside this area (Tortora *et al.* 2008). Our study showed a lower frequency compared to the results published by Tortora *et al.* (2008), in that only 2 individuals, or 2.15% of the sample, had root abnormalities of the central incisor, while 3.23% of the sample had atypical tooth anatomy outside of the cleft.

5.9 Ectopic eruption

Ectopic teeth in the general population ranged between 2 and 6% for maxillary first molars (Bjerklin and Kurol, 1981) and 1 to 2% for permanent canines (Fleming *et al.* 2009).

Ectopic eruption of canines in the cleft region has shown increased angulation, higher vertical position, and are located closer to the midline when compared to the non-cleft side (Holz *et al.* 2018; Westerlund *et al.* 2014). Lai *et al*. (2009) reported ectopic eruption of the maxillary incisors and canines in patients with CLP. Peck and Peck (1995) reported evidence that the maxillary canine-first premolar transposition is genetically controlled.

The lack of space for those teeth induced by maxillary constriction may cause eruption problems for the upper canine. In untreated occlusions in children with a cleft, especially with complete bilateral cleft, the incisive bone tends to move forward, and the maxilla segments close to each other at the time of eruption, which consequently causes maxillary narrowing, mainly in the anterior part (da Silva *et al.* 1998).

Rizell *et al*. (2019) showed that ectopic eruption of canines was only 1.1% in their sample. The prevalence of maxillary canine ectopia in our study was 16.13%, 3.23% for the first premolar and 1.07% for the second premolar. The prevalence of maxillary lateral incisors was found to be 3.23%. We reported no ectopic eruption of molars or maxillary central incisors in our sample.

5.10 Tooth extraction

Our investigation found that 2.15% of maxillary right first molars, 3.23% of maxillary left first molars, 6.45% of mandibular right first molars, and 4.3% of mandibular left first molars were missing. This reflects the lack of oral hygiene and or poor dental practices among this sample.

According to Cobourne *et al.* (2014), the majority of first permanent molars are extracted due to dental caries. Dental caries is still a global public health issue and the major concern to children's oral health today (Hasslöf *et al.* 2017). The existence of a cleft in the oral cavity makes maintaining proper oral hygiene difficult, and children are more likely to acquire dental caries (Hasslöf *et al.* 2017).

The risk of caries in the first permanent molar is complex due to the presence of several characteristics particular to this tooth, such as a distinct morphology and early exposure to an acidic oral environment, resulting in a greater susceptibility to the onset, progression, and subsequent destruction or early loss of the tooth (Morales-Chávez and Mendoza-Hernández, 2019).

A variety of factors have been linked to dental caries, including diet, the presence of cariogenic bacteria, the existence of caries in the primary dentition, socioeconomic status, oral hygiene habits, and the absence of periodic medical dental check-ups (Petcu *et al.* 2016).

When one or more of the risk factors exist, the first permanent molar is affected by carious disease, and if timely treatment is not received, its destruction is precipitated, to the point where it cannot be restored and must be extracted.

As a result, potential mesial migration, supraeruption, premature contacts, dental guidance problems, bone loss, and temporomandibular joint disorders, as well as periodontal disease, are consequences of its decay or loss due to carious lesions (Taboada-Aranza and Rodrguez-Nieto, 2018).

Caries is linked to poor dental hygiene. Given the particular structure of the cleft area, lip scar immobility, and concern of injuring or aggravating alveolar dehiscence, it appears that parents of infants with cleft have difficulties implementing efficient brushing procedures (Haliţchi *et al.* 2017).

According to Batchelor *et al.* (2004) dental caries most usually occurred on the occlusal surface, followed by the buccal and lingual surfaces.

Permanent molars serve as guides for permanent teeth because they influence the development of dental occlusion and contribute in maxillary growth and

physiology of the mandibular apparatus. As a result, permanent molar loss without treatment may disrupt the developing dentition, cause severe malocclusions, and negatively impact oral health (Saber *et al.* 2018).

5.11 Tooth Transposition

Tooth transposition is the positional interchange of two adjacent teeth, or the development or eruption of a tooth in a position occupied normally by a nonadjacent tooth (Peck and Peck, 1993). Several hypotheses have been suggested to explain the phenomena, including a genetic origin, trauma, and interchange of the position of developing tooth buds, lack of deciduous canine root resorption, early loss of primary teeth, and prolonged retention of primary teeth. However, a genetic basis has been identified as the primary etiologic component (Ely *et al.* 2006).

The most prevalent type of transposition is certainly maxillary canine-premolar transposition; thus, it has been the one most frequently reported (Shapira and Kuftinec, 1989; Peck and Peck, 1995).

The dental transposition is a subset of ectopic eruption and is a position or order disturbance that affects 0.4% of the general population. However, its occurrence in patients with cleft lip and palate is significantly higher, at approximately 14% (Ribeiro *et al.* 2018; Campbell *et al.* 2014). Rizell *et al.* (2019) study noted a transposition of the maxillary canine and first premolar in 3.4% of the participants. The prevalence of transposition in the current study was 7.53% maxillary canine/first premolars, which is consistent with the findings of the Cassolato *et al.* (2009) study, in which the authors identified ten cases of transposition of the maxillary canine and first premolar in their study (8.6% of cases).

5.12 Infraocclusion

According to Kurol (1981), infraocclusion occurs when the occlusal surface of a tooth is more than 1mm below the occlusal plane of fully erupted adjacent teeth. The prevalence of infraocclusion in the primary dentition ranged between 1.3% to 38.5% (Shalish *et al.* 2010). Ankylosis has been characterized as the most common factor associated with infraocclusion (Kurol, 2002). It has been proposed that ankylosis is a secondary pathology occuring subsequent to other problems, such as deficient eruptive force, infection, deficient vertical alveolar bone growth or traumatic masticatory force producing local periodontal injuries (Proffit and Vig, 1981).

In our study, we found no evidence of infraocclusion, which can be attributed to the premature loss of primary molars. Rizell *et al.* (2019) study found that infraocclusion of one or more primary molars occurred in 7.2% of the 8-year Scandcleft sample.

Table 7: Review of the literature compared with our unilateral cleft lip and palate sample (%)

author	Population	No. of subjects	Cleft lateral missing	central crown abnormalities	3 or 4 transposition	Ectopic eruption	Cleft lateral (peg shaped)	lateral position *wrt* cleft (distal)	Supernumeraries (permanent dentition)	Agenesis of 2nd premolars
Present study	South african	93	35.48%	39.79%	7.53%	16.13%	27.96%	50.54%	10.76%	9.68%
Rizell *et al*.2019	Scandcleft	425	43.8%	5.7%	3.4%	1.1%	44.7%	31.8%	16.9%	17.9%
Tortora *et al.* 2008	Italian	87	48.8%	15.8%	*	*	31.7%	*	7.3%	14.7%
Ribeiro *et al*. 2003	Brazilian	98	49.8%	*	*	*	92.2%	76.5%	6.4%	19.6%
Ranta, 1986	Finnish	109	3.1-10.4%	*	*	*	*	*	20.9%	7.5% to 32.3%
Tan *et al.* 2018	Singaporean	60	43.1%	*	*	*	54.6%	90.9%	21.7%	18.3%
Dewinter *et al.* 2003	Belgium and the Netherlands	75	58.6%	32%	*	*	*	*	10.6%	27.2%
Suzuki *et al.* 2017	Japan	1724	54.8%	*	*	*	21.6%	99.4%	5.7%	12.3%
Cassolato *et al.* 2009	Canadian	116	27%	*	8.6%	*	*	77%	17%	31.9%

*Data not available
wrt- with regards to

Table 7 shows a comparison of the literature with our study findings. The congenital absence of the permanent lateral incisor on the cleft side was observed in 35.48% of our patients with UCLP. This percentage is very similar to other studies reported values ranging from 27% to 58.6%. (Cassolato *et al.* 2009; Tan *et al.* 2018; Rizell *et al*. 2019; Tortora *et al*. 2008; Ribeiro *et al*. 2003; Dewinter *et al.* 2003; Suzuki *et al*. 2017). Ranta (1986) reported the lowest prevalence rate of 3.1-10.4%. These figures show that there are considerable differences between different populations.

The maxillary lateral incisors were the most commonly missing teeth in our study, followed by maxillary or mandibular second premolars. This distribution of missing teeth, particularly on the cleft side and in the mandible, may indicate a common genetic basis for both the cleft and the concomitant anomaly (Al Jamal *et al.* 2010).

Several studies (Tortora *et al*. 2008; Dewinter *et al.* 2003 and Rizell *et al*. 2019) assessed the prevalence of central incisor abnormalities (malformed/shovelling of incisors of atypical tooth anatomy); our study showed a high prevalence (39.79%) with the lowest rate of 5.7% reported by Rizell *et al*. (2019). These findings may support the hypothesis of a genetic link between clefting and incisor shovelling, or they may indicate that local aetiological factors involved in cleft formation have a direct effect on the morphology of the adjacent developing tooth germs (Walker *et al*. 2008).

In our study, the percentage of transposed maxillary canines and first premolars was 7.53%, which is comparable to the 8.6% reported in a study by Cassolato *et al*. (2009). Although the cause of transposition is unknown, the increased prevalence in patients with clefts may be due to a combination of genetic factors, underdevelopment of the maxilla, and severe crowding.

Rizell *et al*. (2019) reported ectopic eruption of maxillary canines in only 1.1% of cases. This is significantly lower than the 16.13% found in our sample. The differences could be attributed to the sample ages which were different from our study. According to the studies above (Tan *et al.* 2018; Suzuki *et al*. 2017; Cassolato *et al.* 2009 and Ribeiro *et al*. 2003), a higher percentage of the lateral incisor is positioned distal to the cleft than mesial to the cleft. This is because hypoplasia is more common in the medial nasal process than in the maxillary process, according to Hovorakova *et al*. (2006).

The prevalence of peg shaped laterals in our study (27.96%), which is closely similar to the Japanese population (21.6%) (Suzuki *et al*. 2017), is low when compared to other studies, which ranged from 31.7% to 92.2%. (Tortora *et al*. 2008; Rizell *et al*. 2019; Tan *et al*. 2018; Ribeiro *et al*. 2003). In our study, we found that supernumerary teeth were common in subjects with CLP (10.76%). Other studies found a higher percentage of supernumerary teeth 21.7% (Tan *et al*. 2018) and 20.9% (Ranta, 1986), while others found a lower percentage of supernumerary teeth 7.3% (Tortora *et al*. 2008), 6.4% (Ribeiro *et al*. 2003) and 5.7% (Suzuki *et al*. 2017). The lower percentages in other studies could be because they are limited to looking only at supernumerary lateral incisors (Suzuki *et al*. 2017; Tortora *et al*. 2008 and Ribeiro *et al*. 2003), and other studies were looking for dental anomalies in the permanent dentition only.

Limitations of the study

Our study is hospital based and captures only patients who presented for orthodontic treatment. This is biased and may not accurately reflect the prevalence and incidence of dental anomalies in patients with unilateral cleft lip and palate. The study sample and size were too small to generalize the findings. Nonetheless it gives a baseline upon which future population studies can be conducted.

Chapter 6

6. Conclusion

Almost all individuals with unilateral cleft lip and palate were found to have at least one dental anomaly. Agenesis was the most common dental anomaly in our study sample. Most of the dental anomalies were found in the cleft area. The suggestion therefore is that the effect of the cleft disturbance on the dentition is well localized.

The management of dental anomalies, which can easily be detected by careful inspection of routine dental diagnostic records, should be taken into consideration in treatment planning of individuals with a cleft. These patients have a greater number of anomalies which can have consequences and implications in terms of aesthetics, periodontal health, restorative care, and orthodontics. This suggests that patients with cleft lip and palate require extensive orthodontic treatment and should be considered as a special group of patients when developing oral health policies.

Chapter 7

7. References

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APPENDIX A: BMREC ETHICAL APPROVAL

UNIVERSITY of the WESTERN CAPE

50 YEARS of hope, action & knowledge

16 March 2021

Dr V Gomba
Orthodontics
Faculty of Dentistry

Ethics Reference Number: BM20/10/22

Project Title: Analysis of dental anomalies in unilateral cleft lip and palate patients in the Western Cape, South Africa

Approval Period: 15 March 2021 – 15 March 2024

I hereby certify that the Biomedical Science Research Ethics Committee of the University of the Western Cape approved the scientific methodology and ethics of the above mentioned research project.

Any amendments, extension or other modifications to the protocol must be submitted to the Ethics Committee for approval.

Please remember to submit a progress report annually by 30 November for the duration of the project.

Permission to conduct the study must be submitted to BMREC for record-keeping.

The Committee must be informed of any serious adverse event and/or termination of the study.

Ms Patricia Josias
Research Ethics Committee Officer
University of the Western Cape

Director: Research Development
University of the Western Cape
Private Bag X 17
Bellville 7535
Republic of South Africa
Tel: +27 21 959 4111
Email: research-ethics@uwc.ac.za

NHREC Registration Number: BMREC-130416-050

FROM HOPE TO ACTION THROUGH KNOWLEDGE.

APPENDIX B: PERMISSION LETTER FROM TYGERBERG HOSPITAL

Western Cape Government

TYGERBERG HOSPITAL
Reference:
Research Projects
Enquiries:
Dr E van der Merwe
Manager: Medical services
Elrike.vandermerwe@westerncape.gov.za | Tel: 021 938 4430

Ethics Reference: BM20/10/22

NHRD Reference: WC_202204_013

Title: ANALYSIS OF DENTAL ANOMALIES IN UNILATERAL CLEFT LIP AND PALATE PATIENTS IN THE WESTERN CAPE, SOUTH AFRICA.

Dear Dr Vuyisile Gomba

PERMISSION TO CONDUCT YOUR RESEARCH AT TYGERBERG HOSPITAL

1. In accordance with the Tygerberg Hospital Health Research Policy and Protocol of **April 2018**, permission is hereby granted for you to conduct the above-mentioned research here at Tygerberg Hospital for a year based on your HREC approval.

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

DR E VAN DER MERWE

MANAGER: MEDICAL SERVICES

Date: 19/04/2022

APPENDIX C: DATA COLLECTION SHEET

Annexure C

DATA COLLECTION SHEET

Patient ID:	**Gender**	**Male**		**Female**		**Age**	
Unilateral cleft lip and palate	**Right**			**Left**			
Agenesis	Cleft lateral	Non cleft lateral	Bilateral laterals	Maxillary central	Second premolar	Molar	Mandibular agenesis
Agenesis of 2nd premolars	One premolar		Two premolars		Three premolars		Four premolars
Supernumeraries	Lateral	Mesiodens		Mandibular incisor		Lateral+mesiodens	
Cleft lateral shape	Normal	Peg shaped		Other malformation		Agenesis	
Cleft lateral position	Distal to cleft		Mesial to cleft		One lateral on each side of cleft		
Ectopic eruption	Unilateral maxillary molar	Bilateral maxillary molars		Maxillary central	Maxillary lateral	Canine	Mandibular molar
Reversible ectopic eruption	Unilateral maxillary molar				Bilateral maxillary molar		
Transposition	Maxillary canine/1st premolar unilateral			Maxillary canine/1st premolar bilateral			Other
Infraocclusion	One affected tooth	Two affected teeth	Three affected teeth	Four affected teeth		Six affected teeth	
Other anomalies							
Dental anatomy/ development	Cleft central malformed		Cleft central atypical crown-root angulation		Cleft central microdontia	Atypical tooth anatomy outside cleft area	
Position	Inverted Mesiodens or lateral						
Other	Condylar hypoplasia non cleft side				Cleft area odontoma		

APPENDIX D: INTER-RATER AND INTRA-RATER RELIABILITY SCORES

Inter-rater reliability

```
kappaetc UCLP Position_code UCLPPositionR2_code

Interrater agreement                            Number of subjects =      10
                                                  Ratings per subject =       2
                                       Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.     t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .        .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .        .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .        .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .        .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .        .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .        .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc Cleft lateral missing R1_code Cleft lateral missing R2_code

Interrater agreement                            Number of subjects =      10
                                                  Ratings per subject =       2
                                       Number of rating categories =       3
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.     t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .        .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .        .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .        .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .        .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .        .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .        .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

kappaetc MxSecondpremolarR1_code MxSecondpremolarR2_code

Interrater agreement                            Number of subjects =      20
                                                  Ratings per subject =       2
                                       Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.     t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  0.9500    0.0500  19.00   0.000     0.8453     1.0000
Brennan and Prediger |  0.9000    0.1000   9.00   0.000     0.6907     1.0000
Cohen/Conger's Kappa |  0.6429    0.3336   1.93   0.069    -0.0554     1.0000
    Scott/Fleiss' Pi |  0.6396    0.3421   1.87   0.077    -0.0764     1.0000
           Gwet's AC |  0.9419    0.0609  15.47   0.000     0.8145     1.0000
Krippendorff's Alpha |  0.6486    0.3421   1.90   0.073    -0.0674     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

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. kappaetc MandibularagenesisR1_code MandibularagenesisR2_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =       2
                                        Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  0.9000    0.1000   9.00   0.000     0.6738     1.0000
Brennan and Prediger |  0.8000    0.2000   4.00   0.003     0.3476     1.0000
Cohen/Conger's Kappa |  0.7368    0.2539   2.90   0.018     0.1625     1.0000
    Scott/Fleiss' Pi |  0.7333    0.2640   2.78   0.021     0.1361     1.0000
           Gwet's AC |  0.8400    0.1705   4.93   0.001     0.4544     1.0000
Krippendorff's Alpha |  0.7467    0.2640   2.83   0.020     0.1494     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc MdtoothagenesisR1_code MdtoothagenesisR2_code

Interrater agreement                              Number of subjects =       2
                                                  Ratings per subject =       2
                                        Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

kappaetc centralabnormalitiesR1_code centralabnormalitiesR2_code

Interrater agreement                              Number of subjects =      20
                                                  Ratings per subject =       2
                                        Number of rating categories =       3
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  0.9000    0.0688  13.08   0.000     0.7559     1.0000
Brennan and Prediger |  0.8500    0.1032   8.23   0.000     0.6339     1.0000
Cohen/Conger's Kappa |  0.8131    0.1305   6.23   0.000     0.5399     1.0000
    Scott/Fleiss' Pi |  0.8131    0.1305   6.23   0.000     0.5399     1.0000
           Gwet's AC |  0.8635    0.0939   9.19   0.000     0.6668     1.0000
Krippendorff's Alpha |  0.8178    0.1305   6.26   0.000     0.5445     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.
. kappaetc or4transpositionR1_code or4transpositionR2_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =       2
                                        Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.
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. kappaetc EctopiceruptionR2_code EctopiceruptionR1_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =      2
                                        Number of rating categories =      2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc lateralpositionwrtcleftR1_code lateralpositionwrtcleftR2_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =      2
                                        Number of rating categories =      2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc CleftlateralshapeR1_code CleftlateralshapeR2_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =      2
                                        Number of rating categories =      2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc Agenesisof2ndpremolarsR1_code Agenesisof2ndpremolarsR2_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =      2
                                        Number of rating categories =      2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.
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for inter-examiner reliability, Cohen's kappa ranged between 0.6429 to 1.00. Indicating moderate to excellent reliability (accuracy).

INTRA-RATER reliability

```
kappaetc UCLPPositionR1a_code UCLPPositionR1b_code

Interrater agreement                            Number of subjects =      10
                                                Ratings per subject =       2
                                      Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc  CleftlateralmissingR1a_code CleftlateralmissingR1b_code

Interrater agreement                            Number of subjects =      10
                                                Ratings per subject =       2
                                      Number of rating categories =       3
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.


kappaetc MandibularagenesisR1a_code MandibularagenesisR1b_code

Interrater agreement                            Number of subjects =      10
                                                Ratings per subject =       2
                                      Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc MdtoothagenesisR1a_code MdtoothagenesisR1b_code

Interrater agreement                            Number of subjects =       2
                                                Ratings per subject =       2
                                      Number of rating categories =       2
```

```
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc centralabnormalitiesR1a_code centralabnormalitiesR1b_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =       2
                                       Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  0.5000    0.1667   3.00   0.015     0.1230     0.8770
Brennan and Prediger |  0.0000    0.3333   0.00   1.000    -0.7541     0.7541
Cohen/Conger's Kappa |  0.0741    0.2724   0.27   0.792    -0.5422     0.6904
    Scott/Fleiss' Pi | -0.0101    0.3332  -0.03   0.976    -0.7640     0.7438
           Gwet's AC |  0.0099    0.3396   0.03   0.977    -0.7583     0.7781
Krippendorff's Alpha |  0.0404    0.3332   0.12   0.906    -0.7135     0.7943
------------------------------------------------------------------------------

. kappaetc or4transpositionR1a_code or4transpositionR1b_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =       2
                                       Number of rating categories =       2
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

.
kappaetc EctopiceruptionR1a_code EctopiceruptionR1b_code

Interrater agreement                              Number of subjects =      20
                                                  Ratings per subject =       2
                                       Number of rating categories =       4
------------------------------------------------------------------------------
                     |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
---------------------+--------------------------------------------------------
   % Agreement |  0.7000    0.1051   6.66   0.000     0.4800     0.9200
Brennan and Prediger |  0.6000    0.1402   4.28   0.000     0.3066     0.8934
Cohen/Conger's Kappa |  0.4690    0.1677   2.80   0.012     0.1179     0.8201
    Scott/Fleiss' Pi |  0.4655    0.1713   2.72   0.014     0.1069     0.8240
           Gwet's AC |  0.6310    0.1330   4.74   0.000     0.3526     0.9093
Krippendorff's Alpha |  0.4788    0.1713   2.80   0.012     0.1203     0.8374
------------------------------------------------------------------------------

. kappaetc lateralpositionwrtcleftR1a_code lateralpositionwrtcleftR1b_code

Interrater agreement                              Number of subjects =      10
                                                  Ratings per subject =       2
                                       Number of rating categories =       2
------------------------------------------------------------------------------
```

```
                    |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
--------------------+------------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
--------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

. kappaetc CleftlateralshapeR1a_code CleftlateralshapeR1b_code

Interrater agreement                            Number of subjects =      10
                                                Ratings per subject =       2
                                          Number of rating categories =       2
--------------------------------------------------------------------------------
                    |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
--------------------+------------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
--------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.

.kappaetc Agenesisof2ndpremolarsR1a_code Agenesisof2ndpremolarsR1b_code

Interrater agreement                            Number of subjects =      20
                                                Ratings per subject =       2
                                          Number of rating categories =       3
--------------------------------------------------------------------------------
                    |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
--------------------+------------------------------------------------------------
   % Agreement |  0.7000    0.1051   6.66   0.000     0.4800     0.9200
Brennan and Prediger |  0.5500    0.1577   3.49   0.002     0.2199     0.8801
Cohen/Conger's Kappa |  0.4667    0.1760   2.65   0.016     0.0983     0.8350
    Scott/Fleiss' Pi |  0.4582    0.1842   2.49   0.022     0.0727     0.8438
           Gwet's AC |  0.5851    0.1511   3.87   0.001     0.2688     0.9015
Krippendorff's Alpha |  0.4718    0.1842   2.56   0.019     0.0863     0.8573
--------------------------------------------------------------------------------

kappaetc MxSecondpremolarR1a_code MxSecondpremolarR1b_code

Interrater agreement                            Number of subjects =      10
                                                Ratings per subject =       2
                                          Number of rating categories =       2
--------------------------------------------------------------------------------
                    |   Coef.  Std. Err.    t    P>|t|   [95% Conf. Interval]
--------------------+------------------------------------------------------------
   % Agreement |  1.0000    0.0000      .       .     1.0000     1.0000
Brennan and Prediger |  1.0000    0.0000      .       .     1.0000     1.0000
Cohen/Conger's Kappa |  1.0000    0.0000      .       .     1.0000     1.0000
    Scott/Fleiss' Pi |  1.0000    0.0000      .       .     1.0000     1.0000
           Gwet's AC |  1.0000    0.0000      .       .     1.0000     1.0000
Krippendorff's Alpha |  1.0000    0.0000      .       .     1.0000     1.0000
--------------------------------------------------------------------------------
Confidence intervals are clipped at the upper limit.
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UNIVERSITY of the WESTERN CAPE

APPENDIX E: TURN-IT-IN REPORT

09 June 2023

Dear Sir/Madam

RE: TURN-IT-IN REPORT FOR DR VS GOMBA

I hope this document finds you well.

The turn-it-in report for Dr Gomba's mini-thesis had a score of 17%. The analysis revealed that similarity was found in the terminology used in orthodontics and description of cleft lip and palate anomalies.

I hope you will find this in order.

Kind regards

VS Gomba

TURN-IT-IN REPORT (MINITHESIS 2023)