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MASTER OF SCIENCE

The Use of 3D Digital Models for the Assessment of Relative Maxillary Arch Constriction in Patients with Cleft Lip and Palate

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MASTER OF DENTAL SCIENCE

THE USE OF 3D DIGITAL MODELS FOR THE ASSESSMENT OF
RELATIVE MAXILLARY ARCH CONSTRICTION IN PATIENTS WITH
CLEFT LIP AND PALATE

Nafeesa Qureshi

2013-2014

University of Dundee

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**THE USE OF 3D DIGITAL MODELS FOR THE
ASSESSMENT OF RELATIVE MAXILLARY ARCH
CONSTRICTION IN PATIENTS WITH CLEFT LIP AND
PALATE**

**A DISSERTATION PRESENTED FOR THE DEGREE OF
MASTER OF DENTAL SCIENCE**

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BDS

MFDS RCSEdin

UNIVERSITY OF DUNDEE

DECEMBER 2014

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DECLARATION

I, Nafeesa Qureshi, declare that the following dissertation is entirely my own work

Signed:

Date:

CERTIFICATE

ABSTRACT

Objective: The aim of this study was to evaluate relative maxillary arch constriction in 5, 10 and 15 year-old patients with surgically treated unilateral cleft lip and palate (UCLP) and isolated cleft palate (CP) during growth. A preliminary investigation of the accuracy of linear measurements derived from digital models in comparison to plaster models was also undertaken.

Design & Setting: Retrospective longitudinal study using plaster and digital study models of patients with UCLP and CP from the Scottish Managed Clinical Network (Cleft Care Scotland) archives.

Materials and methods: Thirty sets of plaster study models of patients with surgically treated UCLP and isolated CP at ages 5, 10 and 15 years were randomly selected from the Cleft Care Scotland archive. The study models were scanned in occlusion using a commercially available desktop 3D Laser scanner (www.NextEngine.com). The models were secured in the correct occlusion using transparent adhesive tape and placed on the scanner turntable for 40 minutes with the scanner operating in Macro mode, resulting in 127 microns accuracy. The two scanned families were superimposed and the resultant mesh points were turned into a final scanned model by ScanStudio HD software. The scanned models were saved on a laptop (www.acer.co.uk) connected to the scanner. The plaster and digital models for each patient were then scored using the 5 year old/GOSLON and modified Huddart Bodenham (MHB, a measure of relative maxillary arch constriction) scoring systems by three observers on two occasions, 3 weeks apart. The inter-canine and inter-molar widths were measured on upper plaster and digital models of the same patients using electronic calipers and MeshLab software, respectively by a single examiner.

Statistics: Two-way ANOVA was used to calculate the statistical significance of the differences between age and group (UCLP and CP) with the 5 year old/GOSLON and MHB data. Weighted Kappa and Kendall's coefficient values were used to determine the level of agreement within and between the observers for the 5 year old/GOSLON and modified Huddart Bodenham (MHB) scoring systems, respectively. The Welch t-

test was used to calculate the significance of the differences between the means of the linear measurements from the plaster and digital models.

Results: There was no statistically significant change in 5 year old/GOSLON/MHB scores with age ($p>0.05$). There was a highly statistically significant change in 5 year old/GOSLON/MHB scores between the UCLP and CP groups ($p<0.05$) using plaster and digital models. Intraobserver reproducibility was very good (0.74 to 0.84) for plaster model scores and was moderate to good (0.46 to 0.79) for digital model scores using the 5 year old and GOSLON indices. Intraobserver reproducibility was high (0.72 to 0.85) for plaster model scores and was moderate to good (0.66 to 0.75) for digital model scores using the MHB index. Interobserver reliability was very good (0.72 to 0.85) for plaster model scores and was moderate to good (0.44 to 0.67) for digital model scores using the 5 year old and GOSLON indices. Interobserver reliability was high (0.84 to 0.86) for plaster model scores and was good (0.68 to 0.71) for digital model scores using the MHB index. The differences between the linear distance measurements made on the upper plaster and digital models were not statistically significant ($P>0.05$) with the mean differences being less than 1mm, and this was also regarded as clinically insignificant.

Conclusions: There was no progressive worsening of relative maxillary arch constriction with growth between ages 5 and 15 years in patients with surgically treated UCLP and isolated CP. Relative maxillary arch constriction was greater in patients with UCLP compared to patients with isolated CP. The reproducibility and reliability of the indices was good to very good using plaster models but only moderate to good agreement for digital models. The linear measurements from the digital and plaster models were similar. These results are preliminary and a larger sample and more accurate scanning technology is required for the verification of digital models as an alternative to plaster models.

CHAPTER ONE: INTRODUCTION

Orofacial clefts are a heterogeneous group of congenital disorders characterised by the presence of fissures on the lip and/or the palate (Lo et al., 2003). Among all cleft types, clefts of the lip and palate (CLP) are the most common. Children born with clefts may exhibit a range of functional and aesthetic co-malformations e.g. feeding problems, hypo/hypernasality and nasal air escape on speech, skeletal mid-face deficiency, a constricted maxillary dental arch, congenitally missing and malformed teeth, hearing loss due to otitis media with effusion (OME), psychological difficulties and an increased risk of dental caries (Mossey et al., 2009).

Primary surgical repair of the lip and palate is one of the earliest and the most significant interventions that is carried out for the management of patients with CLP. There are multiple treatment protocols used by cleft teams worldwide and there is no consensus about the optimal timing and techniques for surgical repair. Maxillary retrusion is the most common long-term complication (Williams et al., 2001).

Isolated cleft palate is the most common type of cleft in Scotland with a mean incidence of 45 births per year (Dobbyn, 2009). Retrusion of the maxilla is prevalent in patients with repaired CP (Yoshida et al., 1992). There is a need for further studies to compare the most common cleft type in Scotland (CP) with the overall most common type of cleft (UCLP) with regards to relative maxillary arch constriction.

Various methods have been used by researchers to measure relative maxillary arch constriction such as cephalometry and radiographs. The traditional and relatively non-invasive method involves the assessment of the dental occlusion using plaster study models. However, plaster models have a number of drawbacks and to overcome them, three dimensional digital models have been introduced. This study will involve the three dimensional digitisation of study models of patients with CLP and will validate their use for the measurement of surgical outcomes. Notably, using an existing 'off the shelf' scanner for the digitisation of plaster models could improve the speed of assessment of treatment outcomes, which could assist in determining the optimal surgical protocol.

Three measurement indices, namely the 5 year old, the modified Huddart Bodenham index (MHB) and GOSLON yardstick were used to assess occlusion on plaster models and their digital (scanned) counterparts. These indices were selected for the following reasons: 1) The GOSLON yardstick is the most commonly used index for the measurement of treatment outcome worldwide, allowing the results to be 'benchmarked' with those from other studies. The 5 year old index is used to assess occlusion in 5 year old patients with surgically treated CLP and allows evaluation of the effect of surgical treatment alone on the maxillary arch. However, both are 5-point categorical indices with the majority of subjects scoring in the second or third categories. 2) Conversely, the MHB index is an ordinal scale index and as a result more precise differences in occlusion can be detected, which enables the determination of more subtle differences between groups. The study will shed further light on the comparison between these indices based on their reproducibility and reliability.

1.1 Normal maxillary growth

The human craniofacial skeleton and its accompanying dental arches go through noticeable changes as they grow, adapt and age. During the transitional dentition, the changes are relatively rapid and become less pronounced once the permanent dentition is established (Carter and McNamara Jr, 1998). So far researchers have addressed the issues that take place in first two decades of life because the rapid changes in growth occur during this period (Moorrees, 1959, YAVUZ and OKTAY, 2005, Dutra et al., 2009). Bishara et al. (1997) studied longitudinally the inter-canine and inter-molar widths over a 45 year span involving two cohorts of normal subjects. The first cohort of subjects was evaluated at ages 6 weeks, 1 year, and 2 years and the second cohort at ages 3, 5, 8, 13, 26 and 45 years. In the former cohort, the anterior and posterior arch widths increased, in both the arches with the primary dentition still erupting. The inter-canine and inter-molar widths maintained the increasing trend between ages 3 and 13 years. Once the permanent dentition had fully erupted upto the second permanent molars, both the arches (U/L) showed a decrease in inter-canine and inter-molar widths.

1.2 Maxillary growth in surgically treated patients with CLP

Patients with surgically treated CLP have abnormalities of dental arch form (severe maxillary arch constriction), malocclusion (class III malocclusion) and facial deformity. Mars and Houston (1990) investigated the effects of surgery on facial growth and morphology in Sri Lankan males (age >13years) with and without UCLP in a case control study. The cases had three separate subgroups; those with completely unrepaired CLP, those who received only lip repair in infancy, those who had lip and palate repaired in infancy. It was concluded that patients who had no surgery had a normal potential for maxillary growth, patients with lip repair only had a nearly normal maxillary growth but maxillary retrusion was common in the subgroup of patients with surgically treated CLP. Lin et al. (2014) compared cranio-facial hard and soft tissues between patients with surgically treated CLP and the normal subjects with skeletal class III malocclusion. All the participants were in their pre-pubertal phase with mixed dentition. After the analysis of CBCT cephalograms, it was found that the patients with UCLP had severe vertical discrepancies and a more hyperdivergent growth pattern. The soft tissue profile was more concave and flattened with a less satisfactory compensatory alteration. In a retrospective study Reiser et al. (2013) evaluated the changes in cleft size and maxillary arch dimensions and related these changes to the surgical interventions performed on patients with UCLP and CP. It was found that the patients with UCLP had wider maxillary arch dimensions than patients with CP during their first year of life, when only lip closure was undertaken. After the closure of hard palate, the transverse growth was decreased in patients with UCLP. At age 5, the patients with UCLP had similar maxillary arch dimensions compared to patients with isolated CP.

1.3: Measurement of linear dimensions:

Plaster study models are the essential part of any dental practice and these are required for research. For medico-legal reasons the dental professionals are required to make and keep records accurately as part of confidential patient record for eleven years for adults and twenty five years for children. Storage of these models poses a huge problem in terms of space and cost. Various methods have been used to archive

the plaster models into three dimensional digital models i.e., Stereophotogrammetry, digital photographs, holography etc. In present study Laser scanner from NextEngine will be used to digitise patients' plaster study models. Many researchers believed that comparing the linear dimensions of plaster study models with digital models could validate the use of the digital models. Many research studies have validated the use of three dimensional digital models in patients with and without CLP so far (Bell et al., 2003, Santoro et al., 2003, Okunami et al., 2007, Keating et al., 2008, Asquith et al., 2007, Asquith and McIntyre, 2010). The present study will shed some light on the linear dimensional comparisons between plaster and digital models of patients with surgically treated UCLP and CP.

CHAPTER TWO: LITERATURE REVIEW

2.1 Background

There are lifelong implications for patients with clefts, resulting in considerable disruption to their lives and adverse psychological consequences for themselves and their families. Patients with clefts require complex long-term surgical and non-surgical treatment which is specific to the cleft sub-phenotype. This treatment is ideally carried out by a multidisciplinary team approach involving a Paediatrician, Cleft Nurse Specialist, Cleft Surgeon, Maxillofacial Surgeon, Anaesthetist, Paediatric Dentist, General Dental Practitioner, Restorative Dentist, Otolaryngologist, Geneticist, Speech and Language Therapist, Orthodontist, Respiratory Physician and a Psychologist (Shaw et al., 1996, Mossey et al., 2009, de Ladeira and Alonso, 2012). For the treatment of rare facial clefts, the services of Neurosurgeon and ophthalmologist may also be required (de Ladeira and Alonso, 2012). The rehabilitation process requires the multidisciplinary team to have a relationship of reciprocity, mutuality and dialogue; with the collaboration among the rehabilitation team in the different areas being fundamental to the success of the care for patients (Freitas et al., 2012). This combined care should make it possible to provide long-term follow-up for the children with CLP throughout their development to adulthood and achieve the treatment goals and objectives listed in the Table 1.

Treatment goal	Treatment objective
Integrity of the primary and secondary palate	Improve feeding Improve speech
Normal facial aesthetics, functional speech and hearing, Good occlusion with normal masticatory function	Improve hearing Improve facial appearance
Airway patency	Reduce the morbidity and negative psychological impact of the cleft and associated deformity
Good dental and periodontal health	
Optimal psychosocial wellbeing	

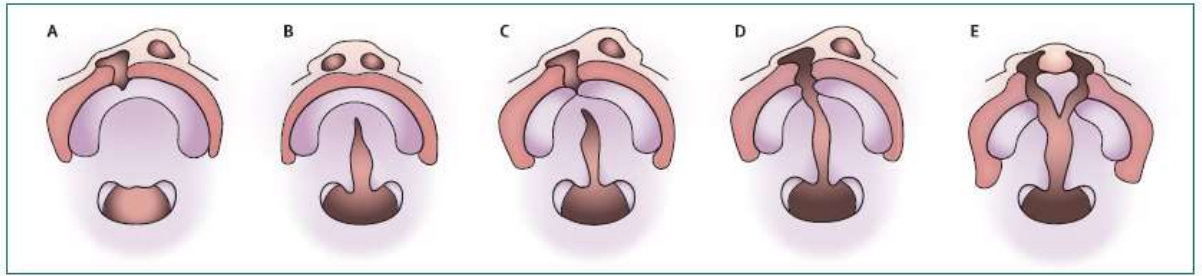
Table 1: Treatment goals and objectives for cleft related care

If these treatment goals and objectives are achieved, they maximise the chances of a child with CLP growing up and developing normally within their social environment. However, the worldwide standard of care for CLP patients continues to remain a cause for concern (WHO, 2002). This is mainly due to little improvement in care of orofacial clefts in different parts of the world over many decades. There is a need for strategic collaboration between different countries to compare and contrast different treatment protocols and surgical methodologies to generate more consistent and predictable results (Mossey and Little, 2009)

2.2 Epidemiology

Craniofacial anomalies are the fourth most common congenital birth defects in newborns (Merritt, 2005) and among all craniofacial abnormalities, clefts of the lip and palate are the most common (Vanderas, 1987). This heterogeneous group of disorders affects the face and / or the oral cavity (Lo et al., 2003). The incidence of orofacial clefts on a worldwide level is approximately 1 in every 600 new births (Mossey and Little, 2002) however, under reporting is a common problem in both developed and developing countries. There is also considerable variation in the frequency of orofacial clefting across the world. CLP usually occurs as an isolated phenotypic feature but can also occur as part of a wider series of birth defects, anomalies and syndromes (Lee et al., 2008).

Non-syndromic orofacial clefts include cleft lip, cleft lip and palate, and isolated cleft palate (Mossey et al., 2009). These disorders affect the lips, hard palate, soft palate, the alveolar ridge area and teeth. Some of the common syndromes associated with orofacial clefts are described in the next section.



*Figure 1: Diagrammatic representation of various Non-syndromic orofacial clefts (A) Cleft lip and alveolus. (B) Cleft palate. (C) Incomplete unilateral cleft lip and palate. (D) Complete unilateral cleft lip and palate. (E) Complete bilateral cleft lip and palate
Reproduced from (Mossey et al., 2009)*

2.3 Embryological background of lip and palate and developmental pathogenesis

2.3.1 Development of the face and palate:

To determine the pathogenesis of orofacial clefting, it is necessary to understand the embryological processes involved in the formation of the face, lip and palate. It is known that human face, lip and palate are among the areas most likely to develop malformations (Chiego, 2014).

The human face develops early in the gestation period during the fourth to the seventh week. The palatal processes are fully closed by the eighth week of gestation. The face and palate are closely related in their time of development and are commonly affected by malformations (Chiego, 2014).

The development of the craniofacial region is an extremely complex process that requires multiple specialised tissues to integrate, such as the surface ectoderm, neural crest, mesoderm, and pharyngeal endoderm, in order to generate the axial skeleton, nervous system, musculature and connective tissues of the head and face.

To understand the development of the face and related structures requires knowledge of the pharyngeal (branchial) arches. These arches start to form on either side of the foregut. Each pharyngeal arch consists of core of mesenchyme, which is covered

externally by ectoderm and internally by endoderm.

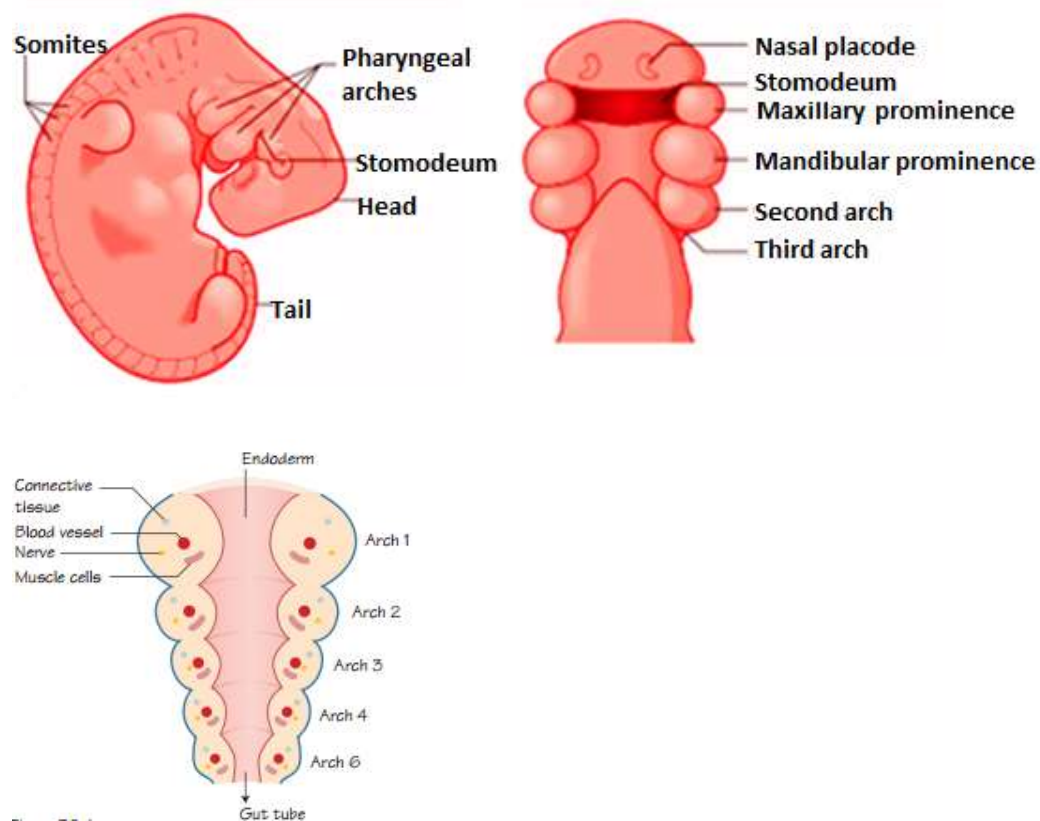


Figure 2: Diagrammatic representation of Pharyngeal arches adapted from (Webster and De Wreede, 2012)

The ectoderm invaginates around the stomodeum (primitive mouth) by the fourth week of the development of the embryo whilst the face develops from the tissues surrounding the primitive mouth. The stomodeum forms the centre of the face early in development

The mesenchyme that forms the core of the pharyngeal arches is derived from the paraxial mesoderm, lateral plate mesoderm, and the neural crest cells. The mesoderm contributes to the development of the musculature that develops in each particular arch while the neural crest cells contribute to the development of the skeletal portion of each arch.

At the early stages of embryonic development, a series of small buds of tissue called the facial primordia form around the stomodeum. The facial primordia are made up mainly of neural crest cells that migrate from the cranial crest.

The upper jaw develops from the following five main buds of tissue: a single median frontonasal process (prominence), the two lateral nasal prominences on both sides, and, flanking these, the two maxillary processes. The lower jaw develops from the paired mandibular primordia and the mandibular processes. Paired maxillary and mandibular processes are derivatives of the first pair of pharyngeal arches. Each of these prominences are formed by the proliferation of the neural crest cells that migrate into the arches from the neural crest during the fourth week of gestation.

The neural crest cells differentiate into the connective tissue components, including bone, cartilage, and ligaments in the facial and oral regions. The muscle cells are derived from a separate cell lineage and originate from the paraxial mesoderm and migrate into the facial primordia.

The frontal portion of the frontonasal process forms the forehead, whereas the nasal element of the frontonasal process forms the dorsal border of the stomodeum and nose.

In summary, the derivatives of the prominences are as follows:

- Frontonasal process - Forehead and the apex of the nose
- Lateral nasal process - Sides (alae) of the nose
- Medial nasal process - Nasal septum
- Maxillary process - Upper cheek region and most of the upper lip
- Mandibular process - lower lip, chin and lower cheek regions
- Mesenchyme in the facial prominences – Muscular derivatives and various bones.

2.3.2 Early Development of the Face

The development of face occurs mainly between the fourth and eighth weeks of the embryonic life.

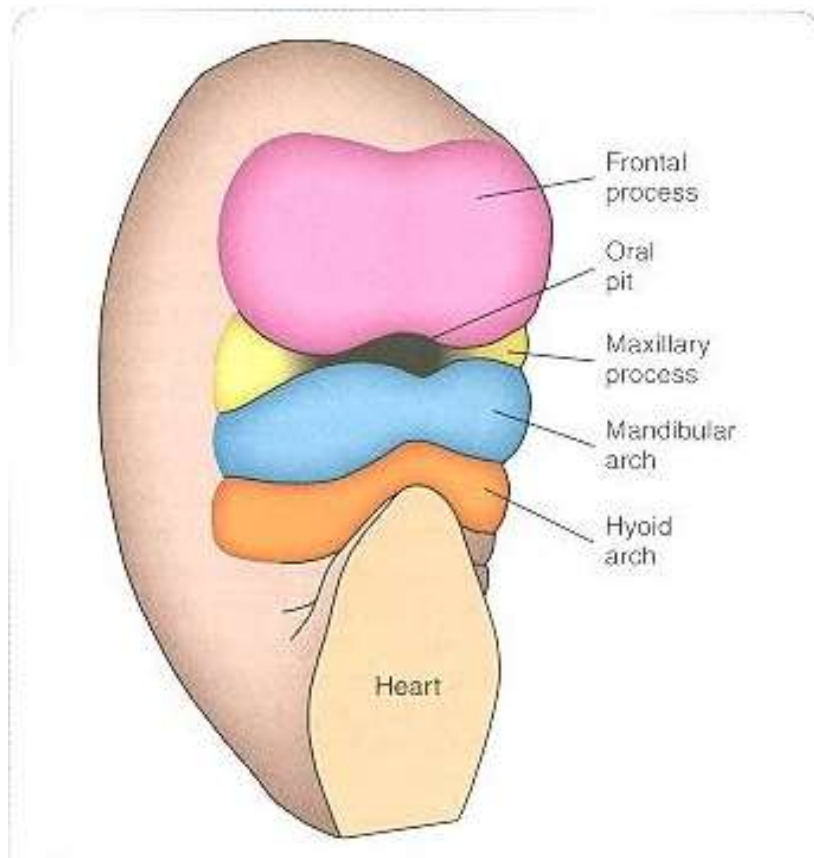


Figure 3: Human face during the fourth week of embryonic life. Adapted from (Chiego, 2014)

By the 4th week of development the primordia of the face appear at the cephalic end of the embryo. Two nasal placodes cap the bulbous frontal prominence. Three paired branchial arches have formed by this stage. The first arches split into maxillary and mandibular processes. In the midline, the mandibular arch appears, but is constricted. The second arch is called the hyoid arch. Between the first arches and frontal process, the buccopharyngeal membrane becomes fenestrated and by this stage is known as the oral pit or stomodeum.

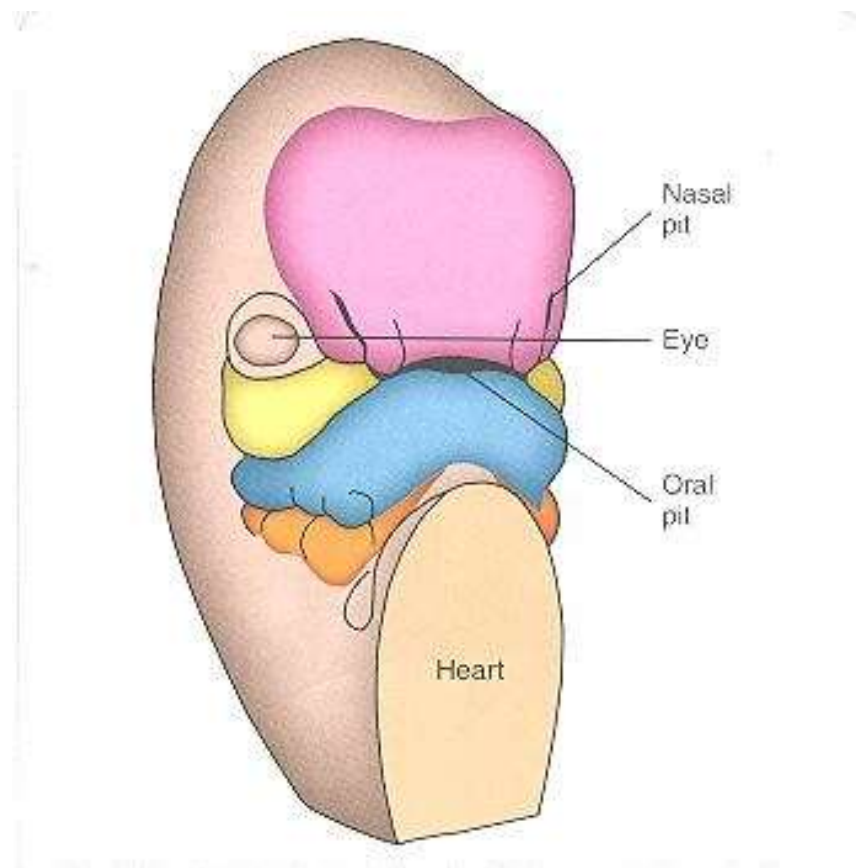


Figure 4: Human face during the fifth week of embryonic development. Adapted from (Chiego, 2014)

By the 5th week of development, nasal pits develop in the nasal placodes, whilst the rims of the placodes differentiate into medial and lateral nasal prominences. The frontal process at this stage becomes the frontonasal process. The lens vesicles invaginate and close within the optic discs. The mesenchyme from the mandibular arch fills in across the midline. Throughout the 5th week, the mandibular arch loses the midline constriction. The caudal end of the medial nasal prominences begin to fuse with the maxillary prominences.

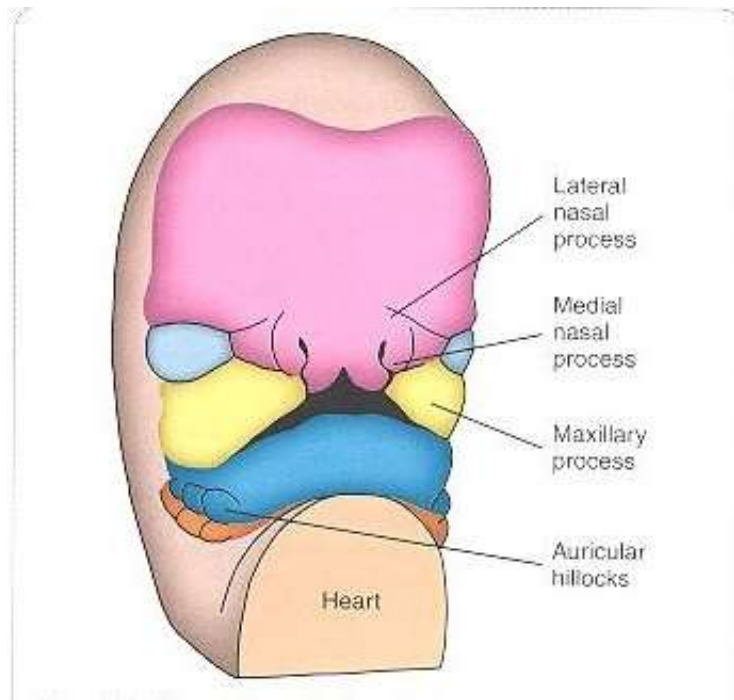


Figure 5: Human face during the sixth week of embryonic development. Adapted from (Chiego, 2014)

At the beginning of the 6th week of development the lateral parts of the face expand, resulting in the widening of the face. Growing and shifting subectodermal mesenchyme smooth out the furrows between prominences and arches, resulting in the second arch increasing in size. Six auricular hillocks, which will become the pinna of the ears, appear on the mandibular and hyoid arches. At the end of the sixth week of embryonic development the medial and lateral nasal processes fuse. The maxillary processes begin the formation of the upper jaw. The approximation of the medial nasal process in the midline forms the nasal septum. At this stage if there is any aberration in the fusion of the medial nasal and maxillary processes, a bilateral cleft lip will result.

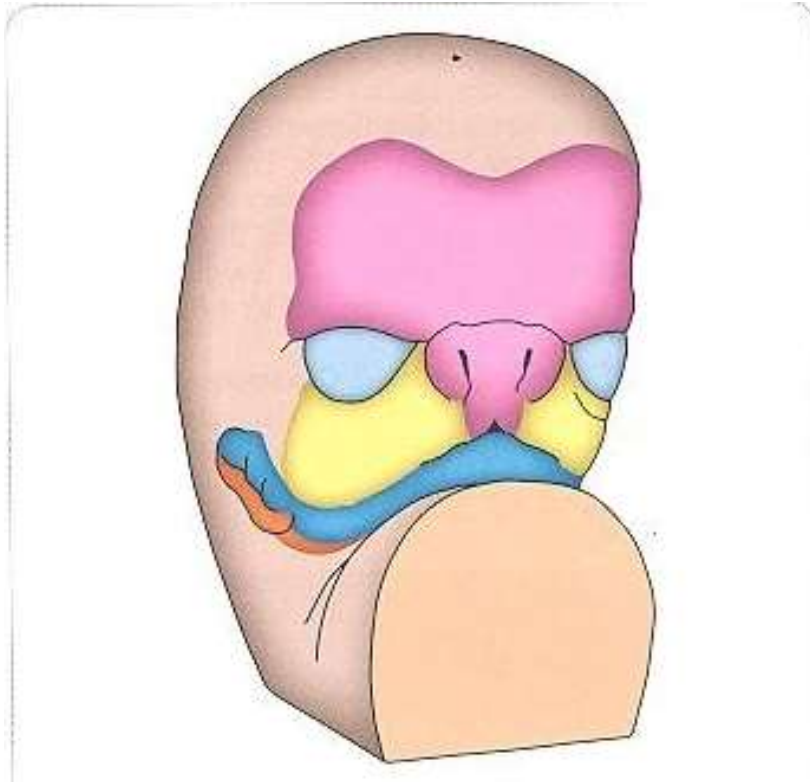


Figure 6: Human face during the sixth week of embryonic development. Adapted from (Chiego, 2014)

At the beginning of the 7th week of embryonic development, the face has a more human appearance. The tip of the nose is elevated between the medial nasal prominences and is visible in profile. The eyelids also become prominent and the pinna of the ear takes shape. By the end of the 7th week of development the pattern of facial features take on a human appearance. However, the facial proportions develop later during the foetal period. The fusion of the medial nasal processes forms the central axis of the nose and the philtrum of the lip. The danger of developing a cleft lip has passed at this stage.

2.3.3 Final Development of the Face

From the start of the 8th week of embryonic development, the facial development occurs slowly and consists mainly of changes in the proportion and relative positions of the facial components and this continues till birth. During the early embryonic stage, the nose is flat and the mandible remains underdeveloped, but both obtain their characteristic form while facial development is being completed. Expansion of the brain creates the prominence of the forehead, the eyes drift medially, and the external

aspects of the ears rise. In conclusion the development of a human face is a dynamic process which takes place in multiple steps.

2.3.4 Development of the Palate

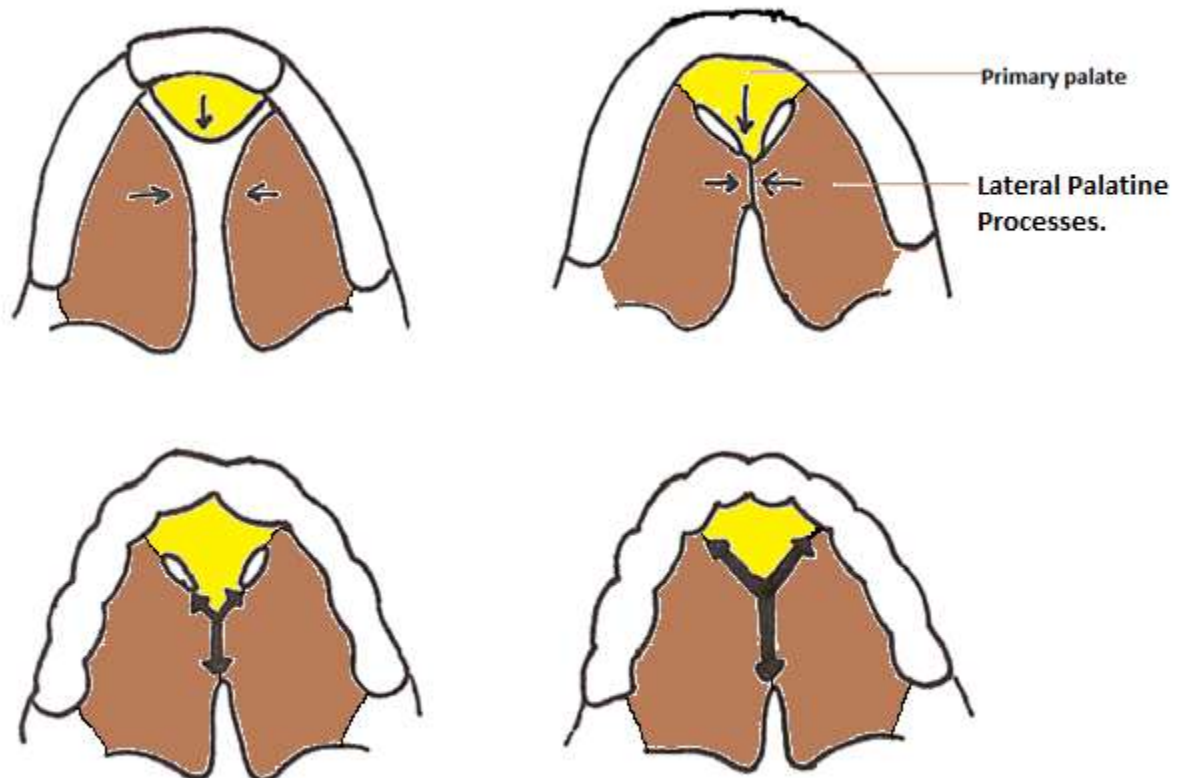


Figure 7: Diagrammatic representation of the formation of the palate

The palate separates the oral and nasal cavities and is divided into the hard and soft palate. The palatal development takes place from the anterior wedge shaped medial part and two lateral palatine processes. The medial part is known as the primary palate as it develops first. The primary palate develops at the same time as the external face. The secondary palate that forms from the lateral palatine processes develops next from the maxillary tissue laterally and grows towards the midline.

As the face grows in the sagittal dimension, the primary palate is too short to provide adequate separation between the nasal cavities and the oral cavity. The secondary palate develops to further separate these cavities. During the 7th and 8th week of embryonic development, the medial walls of the maxillary processes produce a pair of thin medial extensions, called the palatal processes. Initially, these grow predominantly in a downward direction and parallel to the lateral surfaces of the

tongue. By the beginning of the eighth week however, the tongue begins to contract and moves out of the way; this process is known as palatal shelf elevation and takes place very rapidly. The palatal shelves have a final growth surge until they contact in the midline and fuse with each other and with the primary palate; this is known as palatine shelf closure. In addition, the lower jaw drops as it grows downward and forward. The fused palatal processes form the secondary palate together with the primary palate to give rise to the definitive palate. The palatal processes also fuse with the overlying nasal septum in the midline of the face. This results in complete separation of the oral and nasal cavities as far posteriorly as the nasopharynx where both cavities communicate in the pharynx (Chiego, 2014, Sadler, 2011).

2.3.5 Developmental Pathogenesis:

Craniofacial abnormalities can be caused by failures of neural crest cell migration. The development of lip and palate involves a complex series of events that require close coordination of the processes for growth, cell migration, differentiation and apoptosis (Mossey et al., 2009). Clefts occur between the 4th and 12th weeks of intrauterine development, the period during which the embryonic development of the face and palate takes place. Cleft lip results from the failure of the maxillary and the medial nasal processes to merge on one or both sides.

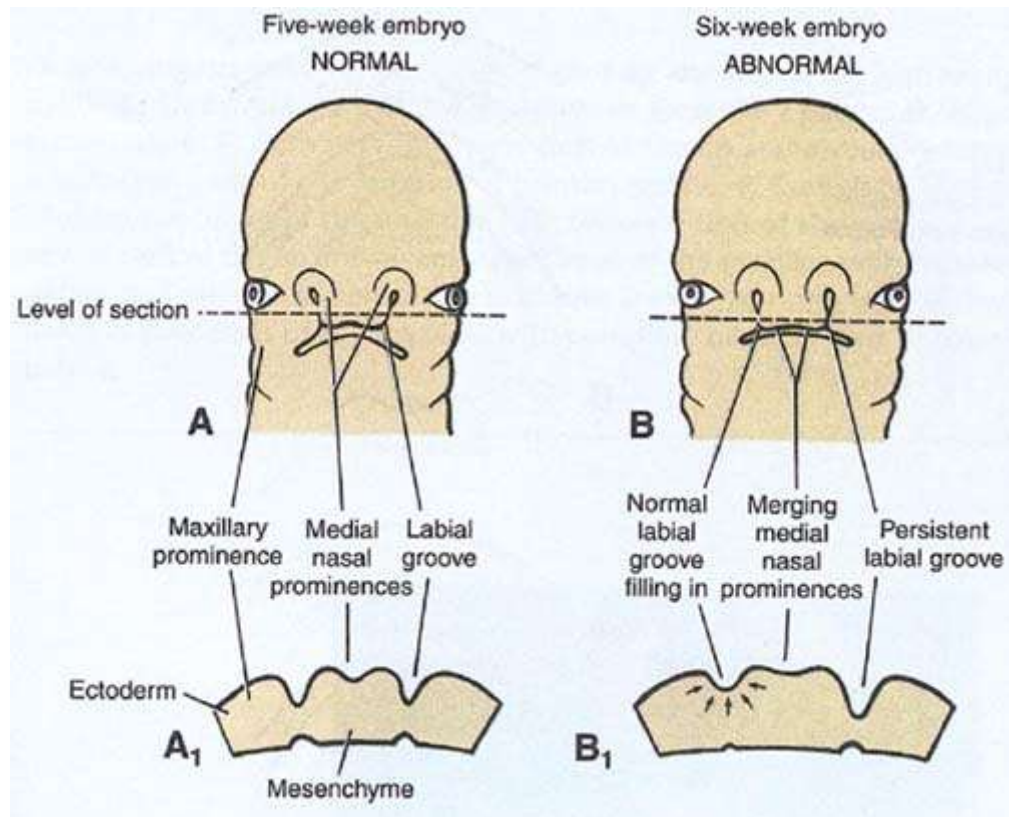


Figure 8: The embryologic origins of a unilateral cleft lip. (A) Normal embryo at 5 weeks gestation. (B) Embryo at 6 weeks with a persistent labial groove on the left side. (A₁) Horizontal cross-section illustrating the grooves between the maxillary prominences and the medial nasal prominences that are merging. (B₁) Horizontal cross-section; arrows point to the grooves filling gradually on the right side after the mesenchymal tissue proliferates. Adapted from (Moore et al., 1994)

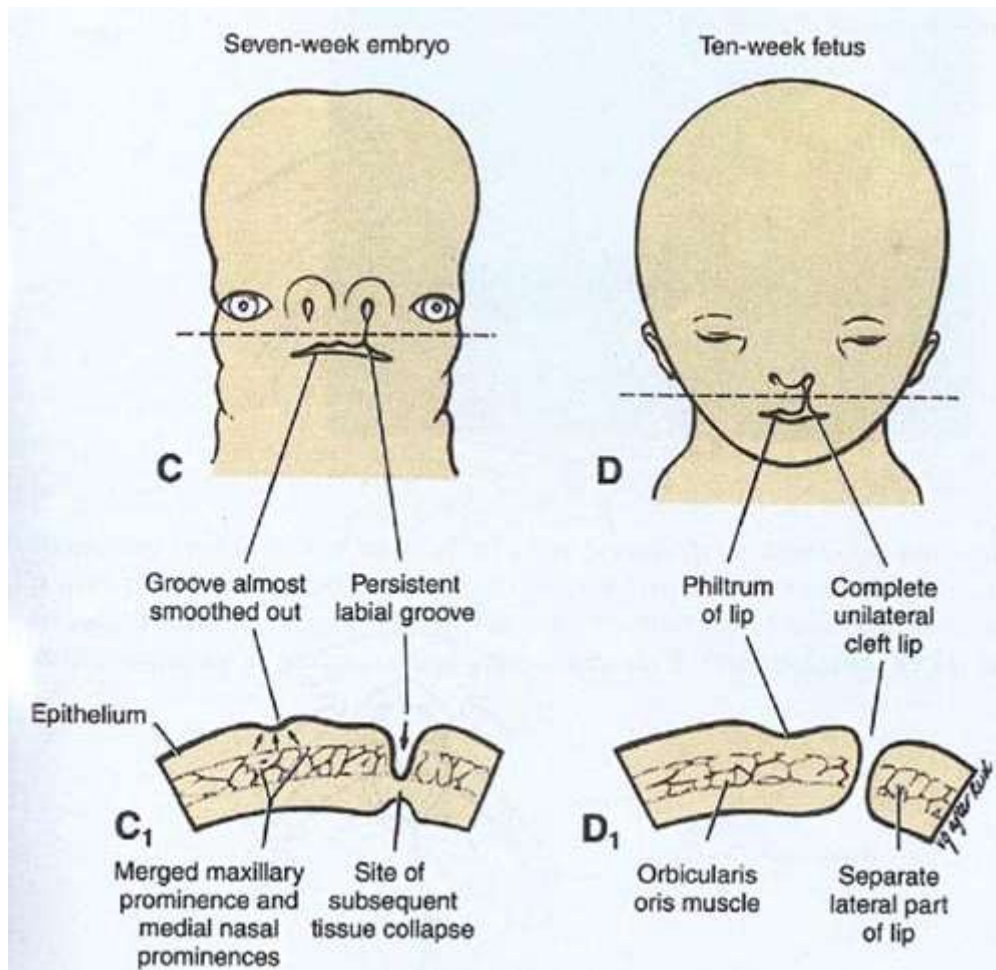


Figure 9: The embryologic origins of a unilateral cleft lip (C) Embryo at 7 weeks. (D) Embryo at 10 weeks with a complete unilateral cleft. (C₁) Horizontal cross-section showing how the merged epithelium on the right, between the prominences, has been almost completely pushed out due to the persistent labial groove. (D₁) Horizontal section shows the persistent labial groove formed from the stretching of the epithelium tissue and breakdown of this tissue, resulting in a complete unilateral cleft lip. Adapted from (Moore et al., 1994)

A cleft palate results from partial or complete failure of the primary and secondary palate to meet and fuse with each other or lateral palatine processes to meet and fuse with each other in the midline. Cleft palate is more often associated with craniofacial syndromes, whereas cleft lip is most often an isolated defect (Merritt, 2005).

A midline cleft lip of the maxilla, historically called a harelip, is a rare condition which involves a notch in the medial nasal tissue that may extend as a cleft into the nose (Chiego, 2014).

2.4 Classification of CLP

There are various classifications that have been devised for the description of cleft lip and palate. A few that have received widespread clinical acceptance are highlighted below.

2.4.1 Davis and Ritchie classification (Davis and Ritchie, 1922)

The Davis and Ritchie classification divides cleft lip and palate into 2 groups, which is subdivided into the extent of the cleft (e.g., 1/3, 1/2), as follows:

- Group I - Clefts anterior to the alveolus (unilateral, median, or bilateral cleft lip)
- Group II - Postalveolar clefts (cleft palate alone, soft palate alone, soft palate and hard palate, or submucous cleft)

2.4.2 Veau Classification (Veau and Borel)

Veau proposed a method of classification that categorised clefts into four categories:

- Group I – Defects of the soft palate only
- Group II – Defects involving the hard palate and soft palate
- Group III – Defects involving the soft palate to the alveolus, usually involving the lip
- Group IV – Complete bilateral clefts of lip and palate

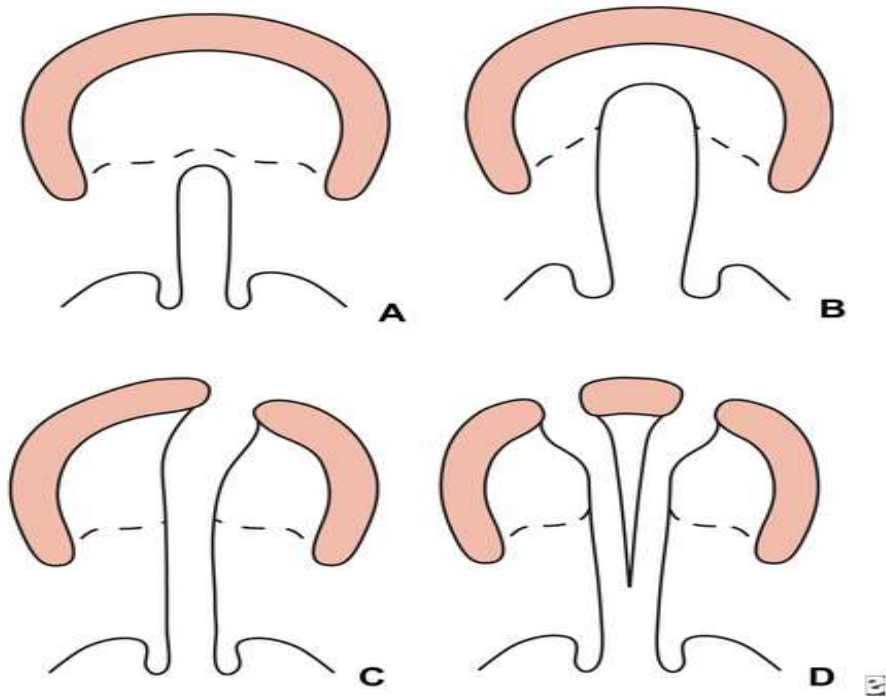


Figure 10: Diagrammatic representation of the Veau classification.

This classification is simple and is still in use in some cleft centres today. However, it does not take into account incomplete clefts or clefts of the lip alone.

2.4.3 (Kernahan and Stark, 1958)

This classification is based on the embryological background of clefts and uses the incisive foramen as a boundary, dividing clefts of the primary palate from those of the secondary palate. The primary palate is made up of the lip, alveolus and the palate anterior to the incisive foramen. A complete cleft of the primary palate involves the full thickness of these structures. The secondary palate refers to the hard and soft palate, up to the incisive foramen. This idea was adopted during the development of Kernahan's striped Y symbolic classification (2014, KERNAHAN, 1971).

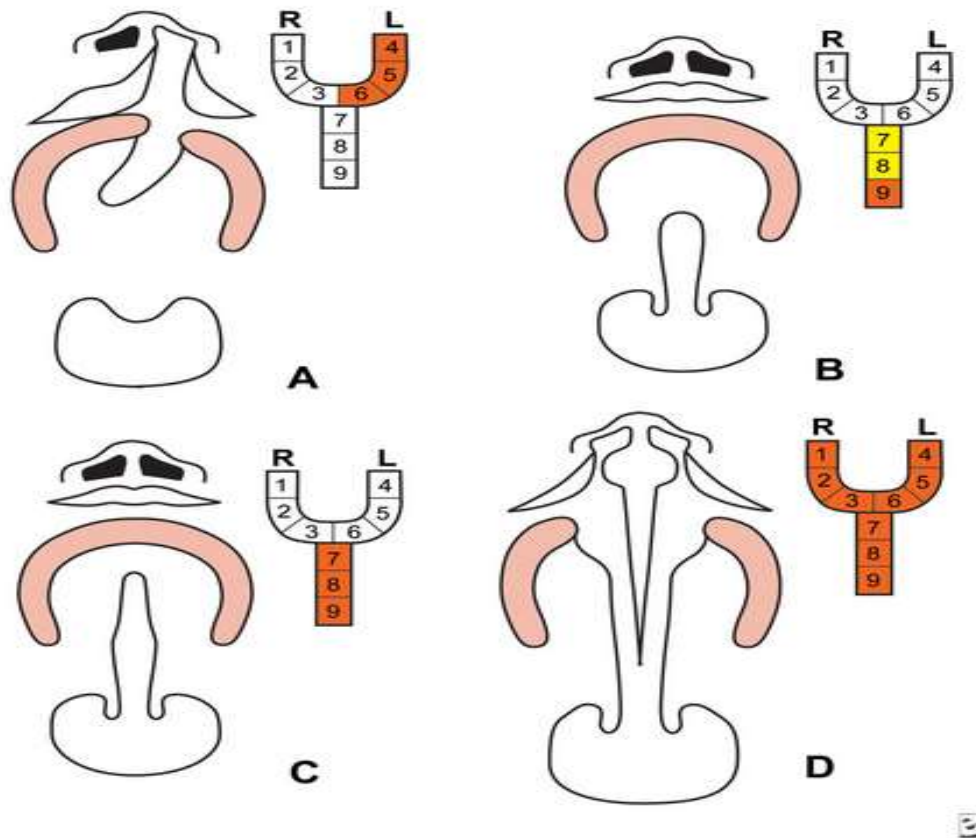


Figure 11: Kernahan and Stark Classification. Areas 1 and 4 represent the right and left sides of the lip, respectively. The alveolus is represented by areas 2 and 5. The hard palate anterior to the incisive foramen is represented by areas 3 and 6. The hard palate posterior to the incisive foramen is represented by areas 7 and 8 and the soft palate by area 9.

2.4.4 International Confederation of Plastic and Reconstructive Surgery classification 1968

This classification system uses an embryonic framework to divide clefts into three groups, with further subdivisions to denote unilateral or bilateral cases, as described below:

Groups	Description
Group I	Defects of the lip or alveolus
Group II	Clefts of the secondary palate (hard palate, soft palate, or both)
Group III	Any combination of clefts involving the primary and secondary palates.

Table 2: International Confederation of Plastic and Reconstructive Surgery classification of CLP

2.4.5 Kriens Classification (Kriens, 1989)

In the United Kingdom, there has been a general move to adopt a simple system of classification of cleft lip and palate, which is easy to use yet has sufficient accuracy for clinical and research purposes. This classification is based on the letters of the Palindrome LAHSHAL, which represent the two sides of the lip (L), alveolus (A), hard palate (H) and the soft palate (S). Upper and lower case letters indicate complete and incomplete clefts, respectively. The Royal College of Surgeons of England adapted the system by omitting one "H" to result in a simpler system, but this removed scope to record bilateral clefts of the hard palate.

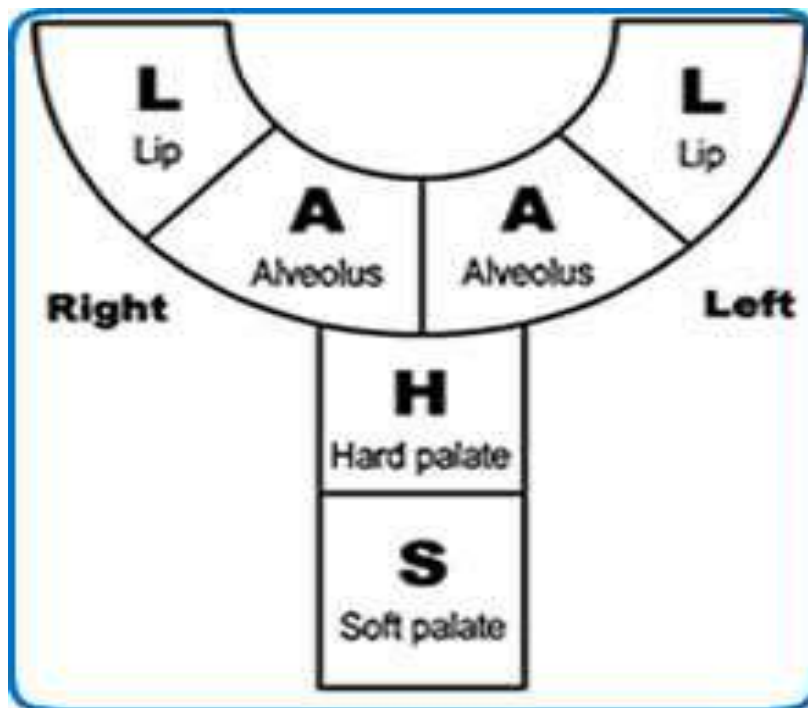


Figure 12: Kriens (1989) classification

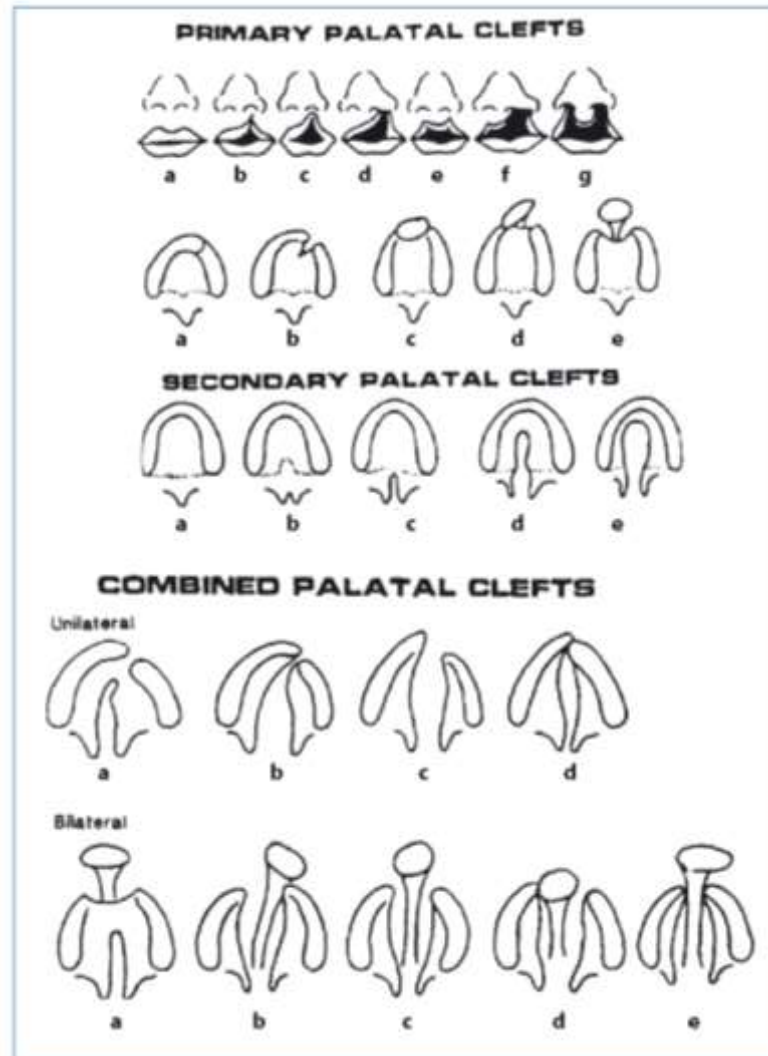


Figure 13: Variations in the types of clefts Adapted from (Berkowitz, 2005)
Variations in the form, size, and extent of clefting in primary, secondary, and combined palatal clefts.

Primary Palatal Clefts (with normal hard palate): Top row: **a** Normal lip; **b–g** the clefts may involve the lip only or may include the alveolus (tooth bearing area) as well. The cleft can extend toward the nostril on one or both sides. Middle row: the cleft of the alveolus can extend to the incisal papilla on one or both sides to any degree. Bilateral alveolar clefts: **c**. incomplete on both sides; **d** incomplete on one side and complete on the opposite side; **e** complete on both sides.

Secondary Palatal Clefts: **a** normal palate; **b** bifid uvula; **c** cleft of soft palate; **d** isolated cleft palate (moderate); **e** isolated cleft palate (extensive).

Combined Palatal Clefts: Unilateral: **a** Isolated CP with cleft lip and alveolus; **b** Incomplete unilateral cleft lip and palate (IUCLP), cleft lip and alveolus are incomplete; **c** Complete unilateral cleft lip and palate (CUCLP); **d** Incomplete unilateral cleft lip and palate (IUCLP). Bilateral: **a** Complete bilateral cleft of lip and alveolus; **b** Bilateral-complete on one side, incomplete on the opposite with complete hard palate cleft; **c** Complete bilateral cleft of the lip and palate; **d** Bilateral incomplete alveolar cleft on one side, complete alveolar cleft on opposite side; **e** Complete bilateral alveolar clefts with both palatal segments attached to the vomer.

2.5 Syndromes associated with clefts in the craniofacial region

In humans, more than three hundred syndromes have been recognised affecting the oral, cardiac, skeletal and other body areas (Gorlin et al., 1990). Fifteen percent of all orofacial clefts are syndromic and cleft lip is the sub-phenotype least associated with syndromes (Mossey and Little, 2002). Around 50% of syndromes are associated with isolated cleft palate and 25% with cleft lip and palate with bilateral CLP almost twice as likely to be associated with syndromes when compared to unilateral CLP (Wyszynski, 2002).

It is important to recognise the syndromes, conditions and anomalies associated with oral clefts to assess the risks faced by the child with these problems. Thorough knowledge of the anomalies associated with orofacial clefts will help determine appropriate treatment and improve the coordination of care provided for children with syndromic clefting.

Pierre Robin sequence is a condition characterised by facial abnormalities and is caused by a sequence of events rather than a specific genetic abnormality, and its exact aetiology is not completely understood. People with this condition demonstrate mandibular hypoplasia, a cleft of the palate and obstruction of the upper airway. The cleft of the palate is thought to occur in the Pierre Robin sequence as the tongue occupies the oronasal cavity preventing the palatal shelves from merging, resulting in a cleft palate. Pierre Robin sequence, can present as part of Stickler syndrome and hence there is an association with isolated cleft palate and Stickler syndrome. Mutations in the COL11A1, COL11A2 and COL2A1 genes have all been identified in this syndrome. Notably, these genes are all involved in the production of type II and type XI collagen, which is disrupted in Stickler syndrome.

Van der Woude syndrome is one of the commonest syndromes associated with clefts of the orofacial region. It is the only orofacial clefting syndrome that presents as either isolated cleft palate or unilateral CLP, whereas other associated syndromes present only with a specific cleft sub-phenotype. It is transmitted as an autosomal dominant trait and is caused by a specific variation in the gene IRF6 (Dixon et al., 2011). It is characterised by lower lip pits, which are on both sides of the vermilion border and are

either oval or transverse in shape. These pits traverse the underlying orbicularis oris muscle and terminate in a blind pouch on the buccal side and communicate with minor salivary glands. The associated features are hypodontia with missing maxillary or mandibular second premolar teeth, absence of maxillary lateral incisors and ankyloglossia. Extraoral manifestations, although rare can include congenital heart defects, accessory nipples, Hirschsprung disease and popliteal web (Venkatesh, 2009, Martelli-Junior et al., 2007).

Treacher Collins Syndrome results from the failure of neural crest cell migration into the first pharyngeal arch. It is characterised by craniofacial abnormalities, including a cleft palate, a small mandible and malformed or absent ears. This syndrome has been linked to a mutation in the *TCOF1 gene* on chromosome number five and can be inherited but can also arise from a random mutation. Facial deformities can vary in severity and usually require surgical intervention. Problems with hearing are also common as there is an anomaly in the formation of the structures in the ear as these are also formed by the first arch.

Velo Cardio Facial Syndrome (VCFS) is an autosomal dominant condition and occurs in approximately one in 2000 live births (Thomas and Graham, 1997). Two genes *COMT* and *TBX1* are associated with VCFS, however many more are yet to be identified. It is known that most of the children who have been diagnosed with VCFS have a microdeletion affecting chromosome 22. The most common features are cleft palate, a cardiac anomaly and a characteristic facial appearance which includes malar flattening, vertical maxillary excess, relative mandibular retrusion, narrow palpebral fissures and small ears. There are also minor learning difficulties and additional consequential educational support needs, speech and feeding problems (Venkatesh, 2009).

Median facial dysplasia is caused by mutations affecting the *ALX1*, *ALX3*, *ALX4* genes. It is characterised by midline facial deficiencies along with unilateral or bilateral cleft lip with or without cleft palate (Noordhoff et al., 1993). Children with this condition have compromised development of the midface resulting in Class III malocclusion and severe maxillary hypoplasia (Venkatesh, 2009).

2.6 Aetiology of CLP

The precise aetiology of CLP remains to be fully determined with progress having been made in identifying environmental factors and genes involved in syndromic CLP. The current evidence suggests it is multifactorial in nature with a genetic predisposition and contributing environmental factors (Dixon et al., 2011).

2.6.1 Genetic factors in the aetiology of CLP:

The genetic factors contributing to CLP have been identified in some syndromic cases, but the understanding of the genetic factors that contribute to non-syndromic CLP cases are still not fully understood. CLP appears to run in families, even though in some cases, there does not appear to be an identifiable syndrome present (Beaty et al., 2011). This could possibly be due to the incomplete genetic understanding of facial and CLP development. Many genes that contribute to the development of clefts in syndromic cases of CLP have been identified such as *SKI/MTHFR*, *TGFB2*, *TGFA*, *MSX1*, *PVRL1*, *TGFB3*, *GABRB3*, *RARA*, *BCL3* (Murray, 2002). Present understanding of the genetic complexities involved in the morphogenesis of the face and oral cavity, together with molecular and cellular processes has been significantly aided by research on animal models in relation to the genes *SHH*, *BMP4*, *FGF10*, *SHOX2* and *MSX1* (Cox, 2004, Dixon et al., 2011). The results of a recent study by Ludwig et al. (2014) strongly support the *FOXE1* locus as a risk factor for non-syndromic orofacial clefts. The research evidence also suggests that this locus is the first conclusive risk factor shared between non syndromic CL/P and isolated CP.

2.6.2 Environmental causes of CLP:

Some of the common environmental factors that have been implicated as an aetiological factor in the CLP are included in the Table 3.

Positive family history	Prior affected infant or parent confers the highest risk
Advancing age	Maternal age (<20 or >39yrs) Increasing paternal age
Teratogenic exposure	Smoking, especially if combined with alcohol Medications taken during the first trimester Benzodiazepines Phenytoin Opiates Penicillin Salicylates Cortisone High dose of vitamin A
Vitamin Deficiency	Folic acid deficiency during gestation.

Table 3: Risk factors for CLP [adapted from (Merritt, 2005)]

Maternal smoking has been associated with an increased risk of the development of CLP. The incidence of CLP is increased by an odds ratio of 1.3 among offspring of mothers who smoke (Little et al., 2004b). The increased risk appears to result from the exposure to maternal smoking during the peri-conceptual period (Little et al., 2004a).

Alcohol is a recognised teratogen and exposure to maternal alcohol consumption has also been suggested as a risk factor for oral clefts, but the evidence has been more inconsistent (Mossey and Little, 2009). Studies also suggest that 'binge' drinking patterns increase this risk of the development of clefts (DeRoo et al., 2008). A systematic review and meta-analysis of the literature carried out by Bell et al. (2003) concluded that there was no significant association between alcohol consumption during pregnancy and orofacial clefts in infants, however the study did not rule out methodological issues as the studies included in the review displayed considerable heterogeneity, significantly limiting the reliability of the results.

A survey by Cheng et al. (2003) carried out in Gansu, People's Republic of China, showed that 0.8 percent of children born to women with rubella infection during pregnancy were born with CLP. Both observational studies and interventional trials using prophylactic folic acid supplements suggested that this can influence the risk of clefting (Boyles et al., 2008, Tolarova, 1982, Tolarova and Harris, 1995, Shaw et al., 1995, Loffredo et al., 2001), whilst other studies have found no association between folic acid food fortification and congenital orofacial clefts (Ray et al., 2003).

Beside nutrients and toxins, other environmental factors have been assessed for possible roles in clefting. These factors include hyperthermia (Shahrukh Hashmi et al., 2010), maternal obesity, stress, ionising radiation, occupational exposures to teratogens and infection (Mossey et al., 2009). However, there is no agreement about the harmful effects of these factors, and there is a need for prospective cohort studies which are large enough to measure effects on a disorder such as clefting.

Environmental influences also indirectly interact with genes to produce clefts of the face and mouth. An example of how environmental factors might be linked to genetics comes from research on mutations in the gene *PHF8* that cause CLP. *PHF8* encodes for a histone lysine demethylase and is involved in epigenetic regulation (Loenarz et al., 2010). The catalytic activity of PHF8 has been shown to depend on molecular oxygen, and the importance of oxygen has been considered important with respect to reports of an increased incidence of CLP in mice that have been exposed to hypoxia during early pregnancy (Millicovsky and Johnston, 1981, Loenarz et al., 2010). In humans, CLP has also been linked to maternal hypoxia, as a result of e.g. maternal smoking (Shi et al., 2008), or some forms of maternal hypertension treatment during early pregnancy (Hurst et al., 1995). In summary, orofacial clefts are multifactorial in their aetiology with influences from both genetic and environmental factors.

2.7 Management of CLP

There is no single protocol (i.e. the timing of the individual measures and procedures) for the management of CLP. Treatment protocols vary between different centres and specialists (Long Jr et al., 2011, Semb et al., 2005, Mossey et al., 2009). The UK clinical standards advisory group (CSAG) identified that many cleft centres had adopted a variety of differing surgical protocols (Bearn et al., 2001). It has been suggested that evidence-based practice should answer uncertainties for the treatment of patients with clefts (Lau and Samman, 2007); however high quality evidence is scarce, i.e. systematic reviews and randomised controlled trials of cleft lip and palate care (WHO, 2002). Multicentre collaboration among treatment providers has the potential to reduce the variability of treatment protocols and ensure that patients with clefts receive evidence-based clinical care (de Ladeira and Alonso, 2012). In recent years orthodontists have been instrumental in the large scale multicentre randomised clinical trials in the assessment of CLP surgical outcomes (Kuijpers-Jagtman, 2006).

The early stages of treatment for individuals with CLP involves primary surgical repair of the lip and palate. There are various surgical techniques for the repair of CLP, however there is no agreement on the optimal surgical techniques for CLP repair (Rohrich et al., 1996). The most commonly used treatment protocol for the management of unilateral cleft lip and palate is shown in Table 4.

Poorly performed surgery carries a high risk of interference with facial growth, dental development and speech (Roberts et al., 1991). The most common and significant potential complication of early closure of CLP is the collapse of the maxillary arch with a resultant anterior and posterior crossbite. The results of a study by Mars and Houston (1990) showed that children with CLP who had no surgery had a potential for normal maxillary growth. Children who underwent lip repair in early infancy showed relatively normal maxillary growth, but restricted growth of the maxilla was common and this has the potential to adversely affect facial growth resulting in maxillary constriction and a Class III malocclusion in children who underwent cleft lip and palate repair in infancy.

Timing	Procedure
After 16 weeks of pregnancy	Cleft lip diagnosis by ultrasound images (palatal clefts are more difficult to image adequately)
Prenatal	Discussion with a cleft surgeon. Consultation with a geneticist if necessary.
Neonatal	If the child has a cleft palate, specialised teats and bottles may be necessary to improve feeding after birth
12 weeks of age	Cleft lip repair
6–12 months of age	Cleft palate repair with intravelar veloplasty, sometimes performed in two stages
5 years	Secondary rhinoplasty

Table 4: The most commonly used treatment protocol for the management of unilateral cleft lip and palate (often based on chronological age). Adapted from (de Ladeira and Alonso, 2012)

The most commonly performed surgical procedures for children with CLP are as follows:

- Surgical repair of the cleft lip
- Surgical repair of the cleft palate
- Revision of the cleft lip
- Surgical closure and bone grafting of the alveolar cleft
- Surgical closure of palatal fistulae
- Palatal lengthening
- Pharyngeal flap
- Pharyngoplasty
- Columellar lengthening
- Cleft lip rhinoplasty and septoplasty
- Lip scar revision

- LeFort I maxillary osteotomy

In addition, orthodontic treatment is very complex and varies according to the severity of the cleft (Evans, 2004).

Orthodontic treatment of a child with CLP is carried out in two stages:

- Surgery-related orthodontics
 - Early management (since birth until the time of surgical closure of the palate)
 - Orthodontics related to alveolar bone grafting
 - Orthodontics in relation to orthognathic surgery
- Cleft-related orthodontics (not associated with surgical procedures)

2.8 Surgical outcome:

Maxillary arch constriction is the most common long-term complication of cleft surgery. The abnormal growth of the maxilla in relation to the mandible is attributed to both an intrinsic developmental deficiency and also to iatrogenic causes such as primary surgery. Some researchers acknowledge the intrinsic deficiency in growth is responsible for the mid-facial retrusion in patients with CLP (Bishara, 1973, Isiekwe and Sowemimo, 1984). However, other researchers have found that surgical intervention is responsible for the restricted growth of the maxilla in relation to the mandible when comparing subjects with CLP who had undergone surgery with those that were un-operated (Ortiz-Monasterio et al., 1966, Bishara et al., 1986, Mars and Houston, 1990, Capelozza Junior et al., 1993, Khanna et al., 2012). Nyström and Ranta (1989) by comparing three year old children with and without clefts, the children with clefts were operated at 0.6 years for cleft lip and 1.8 years for cleft palate. It was concluded that forty percent of children with clefts had crowded maxillary arches as compared to six percent of children without clefts. More precisely children with cleft lip had normal maxillary and mandibular arches, those with UCLP had a small maxillary arch and a nearly normal sized mandible whilst the isolated cleft palate group had both maxillary and mandibular arches that were smaller by a similar amount. Williams et al. (2001) reported part of the findings from the CSAG study involving patients recruited from all parts of the United Kingdom. This study investigated the standards of cleft care in

terms of among others, post-treatment dentofacial dimensions, facial appearance, prognosis of alveolar bone grafts, oral hygiene, speech and hearing outcomes, and the satisfaction of care for patients and parents. The study involved two age cohorts: 239 5 year old and 218 12 year old CLP patients treated in 50 NHS cleft centres throughout the UK. The data reported by Williams et al. (2001) involved plaster study casts, photographs, digitised cephalograms, anterior occlusal radiographs. The study also encompassed a thorough dental examination of the subjects and a parent satisfaction questionnaire based on an earlier version produced by the Royal College of Surgeons of England and a thorough interview with patients to assess their levels of satisfaction with their treatment outcome. Among the series of findings, 40% of 5-year olds had crowded arches and 70-% of 12-year old had a retruded maxilla. Consequently, the study report warned that "A rigorous evaluation of cleft care in the United Kingdom reveals disappointing outcomes". Other aspects of the CSAG study were reported by Bearn et al. (2001) who in addition to detailing the nationwide critical appraisal of UK cleft management, also included comparisons of the UK cleft care centres with other European centres. The study highlighted "the poor outcomes for the fragmented cleft care in the United Kingdom compared with European centres. There is an urgent need for review of structure, organisation and training" (Bearn et al., 2001).

As UCLP is the most common type of cleft worldwide, most investigators have concentrated on assessing UCLP and findings suggest that maxillary arch constriction is the main problem. Sasaki et al. (2004) investigated relative maxillary arch constriction by performing a cephalometric comparison of patients with UCLP and non-cleft subjects, the finding of the study suggest that the patients with UCLP had an inhibition of growth of the posterior maxilla in a vertical direction for all ages when compared to non-cleft subjects.

The evidence suggesting maxillary constriction to be iatrogenic outweighs the evidence which suggested it to be an inherent growth deficiency.

2.9 Indices for the assessment of malocclusion in patients with cleft lip and palate:

The outcomes of CLP surgery have been consistently measured with reference to the post-surgical maxillary arch constriction. There are a number of methods that have been devised to measure relative maxillary arch constriction and dental arch relationships as a surrogate of surgical outcome. Many of these systems have been described in Pruzansky and Aduss (Pruzansky and Aduss, 1964), Matthews (Matthews et al., 1970), Huddart and Bodenham system (HB) (Huddart and Bodenham, 1972), the GOSLON yardstick (GY) (Mars et al., 1987), the 5-year-old index (Atack et al., 1997a), the modified Huddart/Bodenham index (MHB) (Mossey et al., 2003, Gray and Mossey, 2005) and the EUROCRAN Yardstick (EY) (Oskouei, 2007).

2.9.1 Pruzansky and Aduss (1964)

This index used a descriptive classification of arch form and prevalence of crossbite on study models just before lip repair, followed by lip repair, just before the palatal repair and when the eruption of deciduous teeth is complete.

This index divides the occlusion into six categories:

- 1) No crossbite
- 2) Canine crossbite only
- 3) Buccal crossbite only
- 4) Anterior and buccal crossbite
- 5) Anterior and canine crossbite
- 6) Incisor crossbite only

2.9.2 Matthews (1970)

This index was developed by Matthews et al. (1970) at the Great Ormond Street Hospital for Sick Children, London. The index uses the three group categorisation system in the description of occlusion in CLP patients.

Class A: All segments of the maxilla are in normal occlusion with the mandible.

Class B: (I) Tooth bordering cleft on the lesser segment is in lingual occlusion.

(II) Lesser segment is in lingual occlusion, the greater segment is in normal occlusion.

(III) Good occlusion in Class III occlusion.

Class C: Class III occlusion with some part of the arch collapsed.

2.9.3 Huddart and Bodenham (1972)

This numerical index was developed by Huddart and Bodenham (1972) as an alternative system to the descriptive system of Pruzansky and Aduss, and Matthews. It was observed that their assessment of cases varied between the two observers and also the same observer varied in observations on different days. It was noted that the descriptive classification system required subjective judgement. Also, while describing malocclusions they did not consider their extent and so did not allow cases to be ranked by severity. As a descriptive system, no statistical analysis was possible.

The numerical scoring system was devised for repaired unilateral CLP in the deciduous dentition for children below the age 6. It has five categories for scoring incisors and 3 categories for scoring canines and molars. The maxillary arch is divided into two buccal segments and a labial segment.

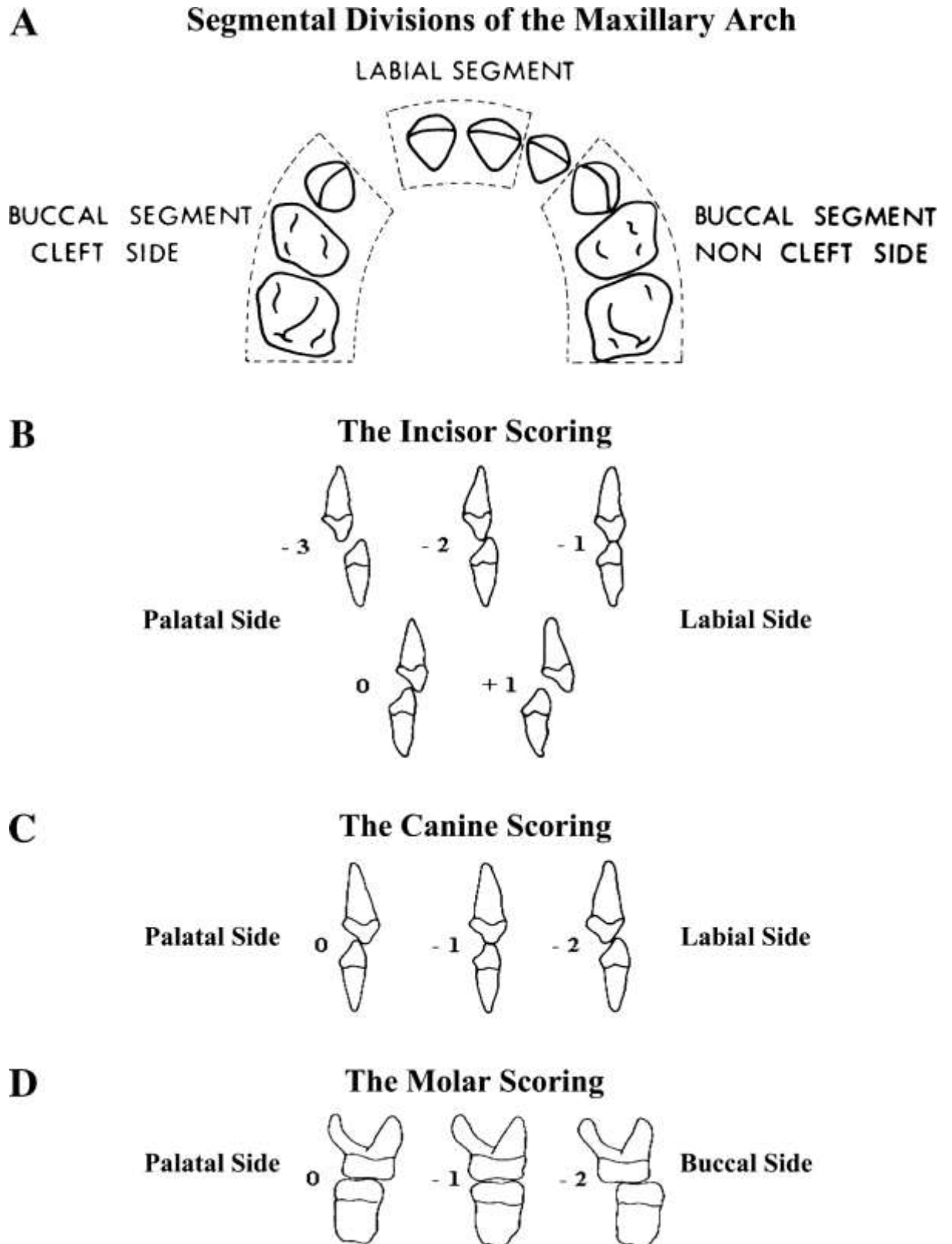


Figure 14: The Huddart/Bodenham system. (a) Segmental divisions of the maxillary arch. (b) Incisor scoring. (c) Canine scoring. (d) Molar scoring. Adapted from (Huddart and Bodenham, 1972)

The Lateral incisors are not assessed as they are frequently missing in patients with CLP. Each maxillary tooth is scored according to its relationship to the corresponding tooth in the mandibular arch. Scores for individual teeth are added to provide a cumulative score for the three segments.

2.9.4 GOSLON YARDSTICK (GY)

The GOSLON (Great Ormond street, London and Oslo) Yardstick (GY) is an Index that allows categorisation of dental relationships in the late mixed and/or early permanent dentition into five distinct categories. Cases are allocated to these categories on a value judgment basis by reference to the calibrated GOSLON models (Mars et al., 1987, 1990).

The GY has the following five grades depending upon the severity of the malocclusion:

- Grade 1 (excellent result)
- Grade 2 (good result)
- Grade 3 (fair result)
- Grade 4 (poor result) and
- Grade 5 (very poor result)

In general, Grades 1 and 2 have occlusions that require simple orthodontic treatment or no orthodontic treatment at all. Grade 3 requires complex orthodontic treatment to correct the malocclusion and in some cases, orthognathic surgery. Individuals in Grades 4 and 5 show very poor dental arch relationships and could only be treated with orthognathic surgery to correct the severe skeletal deviations (Mars et al., 1987). To date the GOSLON yardstick has been the most widely used index in the assessment of surgical outcomes in the world (Altalibi et al., 2013). The major advantage of the GOSLON index is that it considers clinically significant variables in all three planes of space and allows the scoring of models in order of complexity to achieve a favourable outcome.

The major disadvantage of the GY is that it is a subjective ordered categorical index, which does not satisfy the assumptions associated with parametric statistical analysis. The GY requires the operator to be skilled in the use of this index and recalibration is needed to ensure consistency. Ten reference models are required for comparison

during the scoring of study models of patients. All this adds to the complexity of the procedure resulting in the potential for miss-classification.

2.9.5: 5 year-old index

This index was developed to overcome the shortcomings of the GOSLON yardstick (Atack et al., 1997a). It assesses the study models of 5 year olds and allows surgeons to gauge their outcome more accurately so that they can make relevant changes to their clinical skills.

Like the GOSLON yardstick, this is also a categorical index and has 5 Grades:-

Grades	General features	Predicted long-term outcome
1	Positive overjet with average inclined or retroclined incisors. No crossbites and good maxillary arch shape and palatal vault anatomy	Excellent
2	Positive overjet with average inclined or proclined incisors, Unilateral crossbite or crossbite-tendency, +/- open bite tendency around cleft site	Good
3	Edge-to-edge bite with average inclined or proclined incisors; or reverse overjet with retroclined incisors. Unilateral crossbite +/- open bite tendency around cleft site	Fair
4	Reverse overjet with average inclined or proclined incisors, Unilateral crossbite,+/- bilateral crossbite tendency +/- open bite tendency around cleft site	Poor
5	Reverse overjet with proclined incisors, Bilateral crossbite, Poor maxillary arch form and palatal vault anatomy	Very poor

Table 5: Five Grades of 5 year-old index

The major advantage of this index is the excellent and good intra- and inter-observer reliability, respectively. The disadvantages includes the fact that true validation of this index is not possible and it relies on face validity and examiner calibration, therefore making it just as complex to use for scoring surgical outcomes as the GY (Atack et al., 1997b).

2.9.6: Modified Huddart Bodenham (MHB) Scoring System:

This index was adapted from the original Huddart and Bodenham index and modified by Mossey et al (2003) for use in the mixed and early permanent dentitions as compared to the HB system which was designed for use only during the deciduous dentition for patients with unilateral CLP. The index uses a five point scale for each tooth instead of three in the original index.

The modifications of this index are as follows:

- The premolars are scored as deciduous molars.
- If a central incisor is missing the other central incisor is used for scoring.
- If the canine is unerupted, then the canine score is calculated from the midpoint of the maxillary ridge.
- If a premolar is missing, then the score is allocated to the adjacent premolar.
- If none of the premolars are erupted then score is calculated from the midpoint of the maxillary ridge.
- The first molars are not scored before the age of six years and the maximum range of scores is -24 to +8.
- The first molars are scored after the age of six years so the range of scores is -30 to +10.
- To allow comparison of scores between models of under six and other age groups the average of molar scores is taken to ensure the total score is the same in both groups.

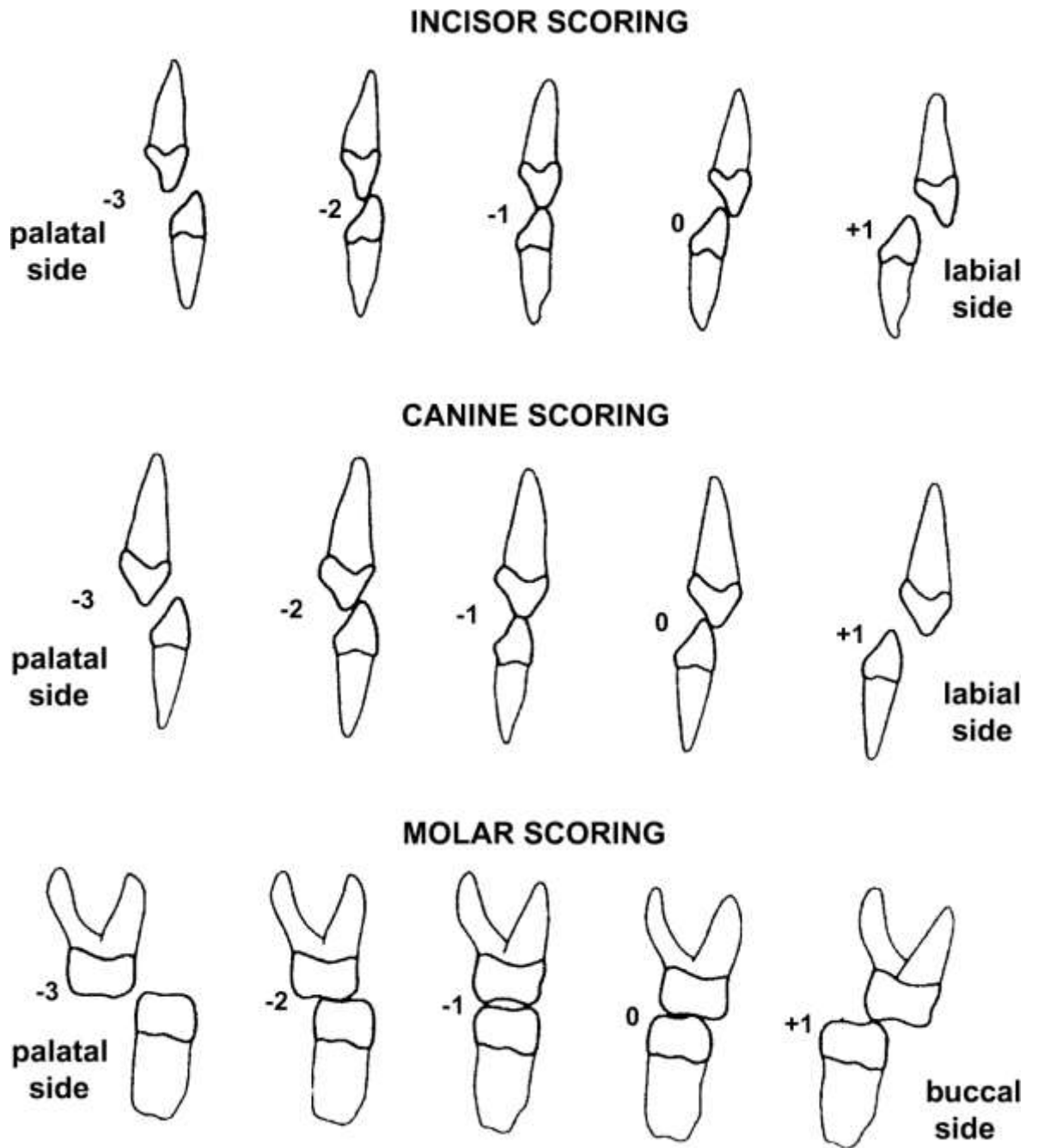


Figure 15: The Modified Huddart Bodenham Index as adapted from (Dobbyn et al., 2011)

Advantages of MHB index :

The study by Gray and Mossey (2005) found the MHB Index to be reliable and objective; it has greater intra- and inter-observer reliability in comparison to the GOSLON and 5 year old indices. The study also showed that the MHB Index is an objective scoring system as neither clinical judgement nor experience is required for scoring. The MHB index is simple and easy to use, no calibration course or reference models are required for scoring when using this index, which eliminates the need to

train examiners and improves the consistency of scoring in multicentre collaborative studies. According to Mars et al. (1987) the more objective the index, the more accurately it can reflect the extent of the interarch discrepancy. The MHB has a high level of reproducibility and user friendliness when compared to other indices (Patel, 2011).

The MHB Index is versatile and sensitive, it can be used for any age and any cleft type (Tothill and Mossey, 2007). The MHB index is a continuous scale measurement and so provides a greater degree of sensitivity and also is appropriate with the assumptions of parametric statistical analysis. The more sensitive the index, the greater its capacity to identify an interarch discrepancy. Therefore, the MHB index has the ability to differentiate severity within the categories that would be identified by the GOSLON or 5-year-old indices (Gray and Mossey, 2005).

The MHB index has been validated on study models by Mossey et al (2003) and Gray and Mossey (2005). The study by (Mossey et al., 2003) showed that there was a high correlation of the MHB index with the 5-year old and GOSLON indices, which in turn determined that the MHB index measures what it is meant to measure and hence is valid. In a larger study by Gray and Mossey (2005) using a sample of 100 patients, the MHB index was shown to be reliable and objective. In a systematic review, Altalibi et al. (2013) concluded that the contemporary evidence suggests that the MHB Index outperformed the rest of the indices on all the World Health Organization (WHO) criteria. They also recommended that the MHB Index is used to score the malocclusions for all patients with CLP at all ages and to standardise the measurement of the outcomes of patients with CLP to a) facilitate international multicentre studies and b) the optimisation of cleft treatment protocols.

2.9.7: EUROCRAN Yardstick (Oskouei, 2007)

The EUROCRAN Yardstick was developed by the participants within the EUROCRAN project (2000-2004). This index was developed from scoring a mix of 118 cases from different European centres. These cases were scored using the GOSLON Yardstick and 5-Year old indices. The results of this study showed that only one case was graded as 5 and two cases as 1 by all the examiners. Thus, due to a lack of use of the extremes of

scales from 1 to 5, a decision was made to reduce the grades that can be scored from 1 to 4 in antero-posterior, vertical and transverse dimensions, instead of the 5-point grading scale. Additionally, a 3-Grade scale was allocated for rating the palatal form.

This index is a modification of the GOSLON Yardstick and 5-Year old indices and is designed to evaluate surgical outcomes for patients with unilateral CLP. This index is based on study models and the major components are the dental arch relationship in the antero-posterior, vertical and transverse dimensions as well as the palatal form. The index is composed of a set of definitions and a set of study models are used as the reference examples. The reference examples were chosen as well-defined examples of a particular grade and were selected on agreement by experienced users of the EUROCRAN Primary Dentition Yardstick.

The EUROCRAN Yardstick has two elements:-

2.9.7.1 Antero-posterior aspects (four Grades)

Grades	General features in antero-posterior aspects
Grade 1	<p>(a) The apical base relationship is Class I or Class II. The apical base takes priority over the incisor relation in classification. As incisor wear is common in the primary incisors, some judgment is required to establish what the original incisal relationship would have been. The position of the cleft side lateral incisor is to be ignored. Both central incisors have/had positive overjet and overbite. On the deciduous canine Crossbite is allowed.</p> <p>(b) The apical base relationship is Class I or Class II. There is no overbite, but the overjet is markedly increased.</p>
Grade 2	<p>The apical base relationship is Class I. There is no overbite and the overjet is not markedly increased, or a molar crossbite is present on the cleft side. The noncleft incisor is/was in positive overjet and overbite or both incisors are worn down to positive edge to edge. However, if there is a moderate openbite, the case is Grade 3.</p>
Grade 3	<p>The apical base relationship is mild Class III. One or both central incisors are edge to edge or in close anterior crossbite with contact and/or wear. There may be moderate openbite. Crossbite of the molar may be present.</p>
Grade 4	<p>(a) Apical base relationship is Class III. Both central incisors are in anterior crossbite.</p> <p>(b) As Grade 3 but there is marked openbite.</p>

Table 6: Four Grades of the EUROCRAN Yardstick for the antero-posterior aspects

2.9.7.2 Palatal aspect (three Grades)

The position of the cleft-side lateral incisor is to be ignored. The worst feature of the three is indicative of the initial score. This may be modified up or down depending on how good the other features are.

Grades	General features in palatal aspect.
Grade 1	<ul style="list-style-type: none"> ➤ There is good anterior and posterior height ➤ There may be minor surface irregularities (bumps, crevices) ➤ There is no or minor deviation of arch form
Grade 2	<ul style="list-style-type: none"> ➤ There is moderate anterior and posterior height ➤ There are moderate surface irregularities (bumps, crevices) ➤ There is a moderate deviation of arch form (e.g., segmental displacement)
Grade 3	<ul style="list-style-type: none"> ➤ There is a severe reduction in palate height ➤ There are severe surface irregularities (bumps, crevices) ➤ There is a severe deviation of arch form (e.g. hourglass constriction)

Table 7: Three Grades of the EUROCRAN Yardstick for the palatal aspect

2.10 National Managed Clinical Network for Cleft Care in Scotland:

The National Managed Clinical Network (NMCN) for Cleft Services in Scotland (CLEFTSiS, now known as Cleft Care Scotland) is based at Perth Royal Infirmary (PRI). The sphere of activity for the cleft lip and palate network is to support a comprehensive, multi-disciplinary service for the treatment of patients with clefts of the lip and/or palate in Scotland. Network members include a wide variety of clinicians and healthcare professionals involved in the care of patients with CLP from all areas in Scotland. CLEFTSiS aims to co-ordinate and optimise care and outcomes through standard-setting and audit for all patients with CLP. Clinicians involved with the cleft care follow an agreed care pathway and each speciality has an agreed protocol.

CLEFTSiS was commissioned on 1st April 2000 in response to recommendations made by the Clinical Standards Advisory Group (CSAG) report (Sandy et al., 1998) and the Scottish Needs Assessment Programme report on cleft lip and palate (1998). The vision of the network is to offer every patient with a cleft lip, cleft palate or cleft lip and palate a specialist cleft care from diagnosis to adulthood. “The service works with the families of the cleft patients offering the right care, in the right place at the right time to produce the best possible outcome for these patients” (CLEFTSiS, 2014). An ‘Electronic Patient Record’ (EPR) system has been set up to overcome the problems of

accurate and reproducible record keeping. The EPR ensures that all appropriate records are taken at the correct time, so that these records can be analysed and compared with standards in other countries.

The administrative staff based at the Network Office in Perth Royal Infirmary and the clinical staff in each NHS Health Board area have access to a single database containing patient records. Currently, patients from all over Scotland are referred into the main surgical sites of Edinburgh, Glasgow and Aberdeen for their surgery, but these patients attend multidisciplinary clinics and treatment centres as near to their home as possible.

Since its inception in April 2000 to September 2011 there were 1092 cases included in the CLEFTSiS EPR. The database contains study models of patients among a variety of other records (McBride et al. 2013). Of these, 730 cases were non-syndromic with a number of other cases including syndromic clefts, atypical and submucous clefts, still births, abortions, cases with non-cleft velopharyngeal incompetence, Pierre Robin Sequence, or Simonart's bands. The study models have not been digitised yet.

The CLEFTSiS NMCN uses Excelicare software AxSys (2014) to integrate a system of user-friendly and clinically familiar folders for the storage and review of all clinical documents and multimedia items, including images, x-rays, audio and video. The software provides comprehensive EPR functionality with the ability to capture, send, receive, store and merge clinical data into patient folders. Currently there are over 100 users of the system from all clinical specialities. In future, it could be possible for patients to access their own records. As clinical notes and records are collated in one central record, analysis and assessment of patient outcome can be made and it is possible to produce facts, figures, and data analysis for each clinical Specialty.

2.11 Three-dimensional (3D) imaging:

3D imaging and the use of digital study models is increasingly gaining acceptance as an alternative to traditional plaster casts and are widely used for orthodontic diagnosis and record keeping (Wiranto et al., 2013). In Orthodontics, 3D digital study models were introduced more than ten years ago (Fleming et al., 2011). Scanners with a high

resolution can produce digital models which have been shown to be as good as physical plaster models (Brief et al., 2006). 3D printing technology is becoming readily available to reproduce the physical casts from the archived 3D digital data using modern Rapid Prototyping technology (Lin et al., 2001, Asquith and McIntyre, 2012). Asquith and McIntyre (2010) suggested in their study report that digital models could be effectively used to evaluate the relative maxillary arch constriction in patients with CLP using a range of indices and could eventually replace the need for plaster casts in near future.

2.11.1 Advantages of digital models:

Digitisation of dental study models is an evolving development in orthodontics (Fleming et al., 2011). Replacement of plaster models with these new virtual equivalents can benefit orthodontics in a number of ways:

- 1) Instant retrieval of digital patient records, resulting in improved efficiency, and immediate information exchange for referral and consultation. The 3D digital study models can be copied easily and integrated into a patient's electronic file along with other digital records.
- 2) Time saving due to relative ease of digital measurement.
- 3) Enable the easy fabrication of orthognathic surgical designed splints, customized brackets, and indirect bonding systems using CAD-CAM technology
- 4) Exchange of data between clinicians and researchers.
- 5) Virtually no storage of a physical archive required as storage space required for digital models is negligible when compared to conventional plaster models.
- 6) 3D models can be stored, manipulated, and measured using a standard personal computer.
- 7) Retrieval is fast and efficient, and the models can be viewed at multiple locations simultaneously.
- 8) No risk of damage or loss as compared to plaster models, Versatility and financial savings.

2.11.2 Disadvantages of digital models:

- 1) 3D models cannot be held and viewed in the same way as plaster models.
- 2) Considerable time is needed to familiarise with their use.

2.12.1 Review of the literature investigating the use of digital models in Orthodontics:

The search strategy was designed to identify all previous research using digital models in the field of cleft lip and palate and three dimensional imaging. A search was undertaken using PubMed (NLM), Medline, Scopus and Google scholar with a combination of keywords: 'Digital models in Orthodontics'. The list of references and bibliographies of all publications were scanned to find any additional publications of relevance, which were not identified by the electronic search strategy.

Fleming et al. (2011) undertook a systematic review to evaluate the validity of the use of digital models to assess arch length, tooth size, arch width, irregularity index, and crowding versus measurements generated on handheld plaster models with digital callipers in patients with and without malocclusion. They concluded that the use of digital models as an alternative to conventional plaster models may be recommended for study and measurement purposes, although it should be noted that some of the evidence included in this review was of variable quality. Many research studies have been undertaken to validate the use of digital models by comparing them to the plaster models (gold standard), details of some of the studies e.g (Tomassetti et al.2001, Bell et al.2003, Mayers et al.2005, Okunami et al. 2007, Keating et al. 2008, Horton et al. 2010) are given in the Table 8.

Investigators	Characteristics of participants	Study design	Index test/Reference standard	Observers (readings)	Outcome measures
(Tomassetti et al., 2001)	22 subjects; USA, 11 pre- and 11 post-treatment; not more than 3 mm crowding.	Prospective	OrthoCad/Digital callipers	1 (3)	Bolton ratio; Time taken
(Santoro et al., 2003)	20 subjects; USA, permanent dentition; no missing teeth; stable occlusion with 3 occlusal contacts or more.	Prospective, enrolled randomly	OrthoCad/Digital callipers	2 (1)	Tooth size; Overjet; Overbite
(Bell et al., 2003)	22 subjects; UK	Prospective	C3D-builder (Uni. of Glasgow)/Digital callipers	1 (8)	Transverse and sagittal linear measurements
(Quimby et al., 2004)	50 subjects; USA, permanent dentition	Prospective, enrolled consecutively	OrthoCad/Digital callipers	10 (2)	Tooth size; Arch length; Transverse dimensions; Overjet; Overbite; Space available; Space required
(Mayers et al., 2005)	48 subjects; USA, permanent dentition	Prospective, enrolled consecutively	OrthoCad/Digital callipers	1 (2)	Peer Assessment Rating (PAR); score
(Costalos et al., 2005)	48 subjects; USA, permanent dentition; post-treatment; no edentulous space; no	Prospective	OrthoCad/Digital callipers	2 (1)	American Board of Orthodontics (ABO

	malocclusion.				score)
(Stevens et al., 2006)	24 subjects; Canada, complete permanent dentition (from 1st molar to 1st molar) without previous orthodontics, pre-treatment models	Prospective, randomly selected from 225 records; three selected within each of 8 categories of malocclusion	Emodels/Digital callipers	3 (3 and 1)	Peer Assessment Rating (PAR); Bolton ratio
(Mullen et al., 2007)	30 subjects; USA, Pre-treatment; complete permanent dentition (from 1st molar to 1st molar).	Prospective	Emodels/Digital callipers	1 (1)	Bolton ratio; Time taken
(Okunami et al., 2007)	30 subjects; USA, permanent dentition; post-treatment; no malocclusion.	Prospective	OrthoCad/Digital callipers	1 (1)	American Board of Orthodontics (ABO score)
(Redlich et al., 2008)	30 subjects; Israel, mixed and permanent dentition; 10 subjects each with mild, moderate and severe crowding.	Prospective	ConoProbe/Digital callipers	1 (3)	Tooth width; Arch length; Crowding
(Hildebrand et al., 2008)	36 subjects; USA, treated cases; consenting patients; no malocclusion.	Prospective, enrolled randomly	OrthoCad/Digital callipers	1 (1)	American Board of Orthodontics (ABO score)
(Goonewardene et al., 2008)	50 subjects; Australia, permanent dentition erupted including third molars.	Prospective	OrthoCad/Digital callipers	1 (1)	Tooth width; Arch length; Crowding Irregularity
(Keating et al., 2008)	30 subjects; UK	Prospective, enrolled randomly	Easy3D Scan/Digital callipers	1 (2)	Linear dimensions (x, y, z planes)
(Veenema et al., 2009)	30 subjects; Netherlands, pre- and post-treatment; permanent dentition; 5 Class I, 19		Digimodel/Digital callipers	2 (1)	<i>Index of Complexity, Outcome and Need</i>

	Class II div 1, 5 Class II div 2, 1 Class III; 5 treated with extractions.				(ICON score)
(Leifert et al., 2009)	25 subjects; USA, Class I molar relationship, crowding.		OrthoCad/Digital callipers	2 (1)	Crowding
(Watanabe-Kanno et al., 2009)	15 subjects; Brazil, permanent dentition; pre-treatment; 12–18 years.		Cecile3/Digital callipers	2 (1)	Transverse dimensions; Tooth size; Overjet; Overbite
(Horton et al., 2010)	32 subjects; USA, permanent dentition; pre-treatment.	Prospective study	Emodels/Digital callipers		Tooth size/ Time taken

Table 8: List of studies that have evaluated the use of digital models in Orthodontics, adapted from (Fleming et al. 2011)

It has been proposed that the accuracy of digital models is clinically acceptable and in the vast majority of situations digital models can be successfully used for orthodontic records (Zilberman et al., 2003, Rheude et al., 2005). Asquith et al. (2007) undertook a study to examine the accuracy and reproducibility of measurements made on digital models. In this study ten sets of orthodontic study models were scanned using the R250 Orthodontic Study Model Scanner and three-dimensional (3D) images were produced by computer software. Two examiners separately measured 11 parameters on the conventional models and the digital models on two occasions. The parameters included inter-canine and inter-molar width, mesio-distal crown diameter, overjet, arch length, and incisor crown height. The study detected some systematic errors in measurements between plaster models and digital models, but were clinically insignificant, and the level of random errors was not high enough to cause concern for measurements between reference marks. Table 9 shows the studies that were undertaken to validate the use of digital models in patients with cleft lip and palate.

Investigator s	Characteristics of participants	Study design	Index test/Reference standard	Obs erve rs	Outcome measures
Brief et al., 2006	Forty plaster models of newborns up to 8 months of age, Heidelberg Germany	Retrospective	Micromasure 70 three-dimensional Laser scanner	(4)	Arch width measurements
(Oosterkamp et al., 2006)	10 digital models of BCLP patients	Retrospective	LDI-scanner/ digital caliper	2	Linear measurements
Asquith and McIntyre 2012	Thirty sets of study models of 5-year-old patients with unilateral cleft lip and palate UK	Retrospective	R250 Orthodontic Study Model Scanner	2	5-year-olds' and modified Huddart Bodenham indices
Mello et al., 2013	Digital models of ninety-four children aged from 3 to 9 months Brazil	Prospective	3Shape's R700™/ OrthoAnalyzer™	1	intercanine distance
Nicholls et al. 2013	30 dental digital study models of UCLP patients Australia	Retrospective	3M Unitek lava TM	2	GOSLON yardstick
Chawla et al 2012	Plaster and digital Study models of 45 patients born with UCLP, UK	Retrospective	R640 3Shape desktop scanner, (Copenhagen, Denmark)	7	5-year-old index

Table 9: List of studies that have evaluated the use of digital models in cleft care

2.12.2 Review of the literature investigating the use of digital models in cleft care:

Brief et al. (2006) performed a study to “quantify the precision of landmark positioning on digitised casts of patients with unilateral cleft lip and palate”. They concluded that there was significant error in both intra-observer and inter-rater measurements.

Asquith and McIntyre (2012) undertook an observational study involving comparison of digital models and plaster models for the evaluation of dental arch relationships for patients with unilateral cleft lip and palate. In this study thirty plaster models of 5 year old subjects with UCLP were scanned using R250 Orthodontic Study Model Scanner (3Shape A/S, Copenhagen, Denmark). The 5-year olds’ and MHB indices were used for scoring. There was a good intra-observer and inter-observer reproducibility and no statistically significant differences were found between the scores for the digital models when compared to plaster models for both indices. The results revealed that digital models can be as reliable as plaster models for cleft care.

(Mello et al., 2013) carried out a study to evaluate the intercanine distance in newborns with cleft lip and palate using 3D digital models. This study did not validate the use of digital models, but assumed the reliability of their use.

No study has investigated the use of a commercial small object scanner for the production of digital study models, which are significantly less expensive than an Orthodontic model scanner.

The objective of this study is therefore to evaluate the accuracy and reproducibility of the assessment of relative maxillary arch constriction using digital models produced using a commercial small object model scanner.

CHAPTER THREE: HYPOTHESES

Based on current understanding of the development of CLP and accepting relative maxillary arch constriction as a measure of surgical outcome, (accepting the current gap in our understanding of its relation with growth), four research null hypotheses were formulated:

1. Relative maxillary arch constriction in patients with surgically repaired UCLP and CP does not deteriorate progressively with growth.
2. There is no difference in relative maxillary arch constriction in patients with surgically repaired UCLP and CP.
3. The reproducibility and reliability of the 5 year old/GOSLON and MHB indices are not different when assessed on plaster and digital models.
4. There is no difference in linear measurements using plaster and digital models.

CHAPTER FOUR: MATERIALS AND METHOD

This was a pilot study to assess the reproducibility/reliability of scoring relative maxillary arch constriction and dental arch relationships using an ordinal index and an ordered categorical index, respectively on 3D digital models produced using a commercial laser model scanner.

4.1 Ethical approval

Caldicott approval for access to patient data recorded by CLEFTSiS was granted by the Tayside Medical Science Centre (TASC).

Good clinical practice (GCP) training was undertaken before embarking on the project to ensure all investigators possessed a thorough knowledge of data protection and patient confidentiality principles. A password protected laptop computer was used throughout the study to ensure the highest standard of data security was maintained.

As a pilot study, a formal sample size calculation was not required.

4.2 Research questions:

- 1) Are digital models a reliable alternative to plaster models for the assessment of relative maxillary arch constriction in patients with CLP?
- 2) Are there any differences in relative maxillary arch constriction as assessed by 5 year old/GOSLON and MHB indices between patients with UCLP and patients with CP from 5 to 15 years of age?
- 3) Is the intra-observer reproducibility and inter-observer reliability of the 5 year old/GOSLON and MHB indices acceptable when used on both plaster and digital models?

4.3 Materials:

Plaster study models for five patients with UCLP and five patients with isolated CP were randomly selected on the basis of availability of all the three model sets taken at ages 5, 10 and 15 years. The plaster study models for each patient were taken from the entire cohort of patients with CLP stored in the CLEFTSiS study model archive

(CLEFTSiS, 2014). These patients had received comprehensive cleft care, further details are contained in appendices P,Q,R,S.

4.4 Inclusion Criteria

1. 5, 10 and 15 year study models.
2. Non-syndromic complete UCLP or isolated CP.
3. Good quality study models with no defects or fractured teeth.
4. Accurately trimmed such that the models could be bench-articulated to establish the correct occlusion.

4.5 Description of participants

These models belonged to patients with either right or left sided UCLP or isolated cleft palate (CP) of either gender. Each subject was identified by a unique ID printed on the individual models and the box itself. The subjects were divided into two groups:

Group 1: Fifteen sets of plaster models of subjects with UCLP Figure 16

Group 2: Fifteen sets of plaster models of subjects with CP Figure 17



Figure 16: Group 1, Plaster models of patients with UCLP



Figure 17: Group 2, plaster models of patients with Isolated CP

Age	5 years	10 years	15years
Number	10 Subjects	10 Subjects	10 Subjects
Cleft Type	UCLP	CP	
Number	5 Subjects	5 Subjects	
Model Type	Plaster	Digital	
Number	30 U/L * Models	30 U/L * Models	
Examiners	GM	NQ	PM

*Table 10: Data breakdown by age, model and cleft type_*U/L meaning upper/Lower Models*

Each subject had three sets of maxillary (upper) and mandibular (lower) plaster models taken at the ages of 5, 10 and 15 years. There were an equal number of subjects in each cleft type. Master 5 year old and GOSLON yardstick upper and lower dental study models were made available by CLEFTSiS (CLEFTSiS, 2014). The master models were compared with patients' plaster and digital models to determine 5 year and GOSLON scores respectively.

4.6 NextEngine Laser scanner

For this study a desktop Laser 3D scanner manufactured by NextEngine Inc. (Santa Monica, California) was used to scan the study models (NEXTENGINE, 2000).

NextEngine Desktop ScanStudio™ software (NextEngine, Inc. Santa Monica, California) was downloaded on an Acer laptop (www.acer.co.uk) for storing and manipulating the scanned data. This scanner along with NextEngine ScanStudio HD digitises objects. The scanner captures objects in full colour with multi-Laser precision. The scanner captures fine detail to 127 micron precision. A powerful computer was used to visualise and manipulate these data (NextEngine, 2014). The scanner along with ScanStudio HD software produced three dimensional mesh models which could be imported into other software. The scanner was connected to the laptop using a USB cable. The scanner uses a turntable called Autodrive, which rotates and aligns automatically. Because dental models have undercuts, the Multidrive was used as this is a dual axis turntable that further automates the alignment by adding tilt to the rotation. The problem of undercuts present in models was further alleviated by increasing the

number of divisions in ScanStudio HD software. To produce higher resolution for the digital models, the HD speed was adjusted. The configuration of the scanner, Autodrive and Acer laptop computer (www.acer.co.uk) are shown in Figure 18. The specifications of the laptop are detailed in appendix A.



Figure 18: NextEngine scanner

4.7 MeshLab Software

MeshLab is an open source, extensible and portable software. It is ideal for processing and analysing 3 dimensional triangular meshes. The MeshLab software was downloaded and used along with ScanStudio HD for measurement of the digital models (Meshlab, 2014). This software is based on a set of tools for editing, cleaning, healing, inspecting and converting the type of mesh models used in the present study. This software recognises STL files which are imported into MeshLab and analysed and saved. The linear measurements taken from plaster and digital models using digital callipers and MeshLab software were saved in Excel spreadsheets and saved as a csv file. The csv files were imported into the RStudio to be statistically tested.

4.8 Digitisation of plaster study models:

The thirty digital models were produced by scanning the plaster counterparts in occlusion. The digital models were divided into two groups. Each group had subgroups of models for patients at ages 5, 10 and 15 years.

Group 3. Fifteen digital models of five subjects with UCLP (right or left sided) at age of 5, 10 and 15 years.

Group 4. Fifteen digital models of five subjects with Isolated CP at age 5, 10 and 15 years.

The NextEngine laser scanner works on the Multistriple Laser Triangulation Technology (MTL) which involves use of laser lines to scan across the plaster study model Figure 18. The sensor in the scanner captures the laser light reflected from the plaster study model. The trigonometric triangulation is used by the system to calculate the distance between the model and scanner. The point data is converted into a digital model by ScanStudio software.

4.9 Preparation of digital models:



Figure 19: Plaster models in occlusion

For scanning purpose the plaster models were held in occlusion by temporarily occluding the U/L plaster models together using transparent adhesive tape Figure 19. The adhesive tape did not apparently interfere with the functioning of the laser beam. The occluded models were then fixed on the scanner turntable using adhesive clay.

4.10 Scoring Forms:

Scoring forms to record the modified Huddart Bodenham and 5 year old/GOSLON scores were provided to the examiners, together with flow charts and calibrated plaster model sets for both the 5 year old and GOSLON indices for undertaking scoring. These are included in Appendix C.

4.11 Examiners

Three examiners were engaged in this study as follows:

Examiner A- Professor and Honorary Consultant in Orthodontics (PM)

Examiner B- Consultant and Honorary Senior Lecturer in Orthodontics (GM)

Examiner C- Masters Student (NQ)

Examiners A and B had extensive experience of treating patients with cleft lip and palate. Examiner C had no previous experience of treating patients with CLP and was included to test the effect of observer inexperience on scoring.

4.12 Method:

The models were placed on the turntable and secured. The following settings were chosen as shown in Figures 20 and 21.



Figure 20: Screenshot showing the setting used to scan individual sets of models

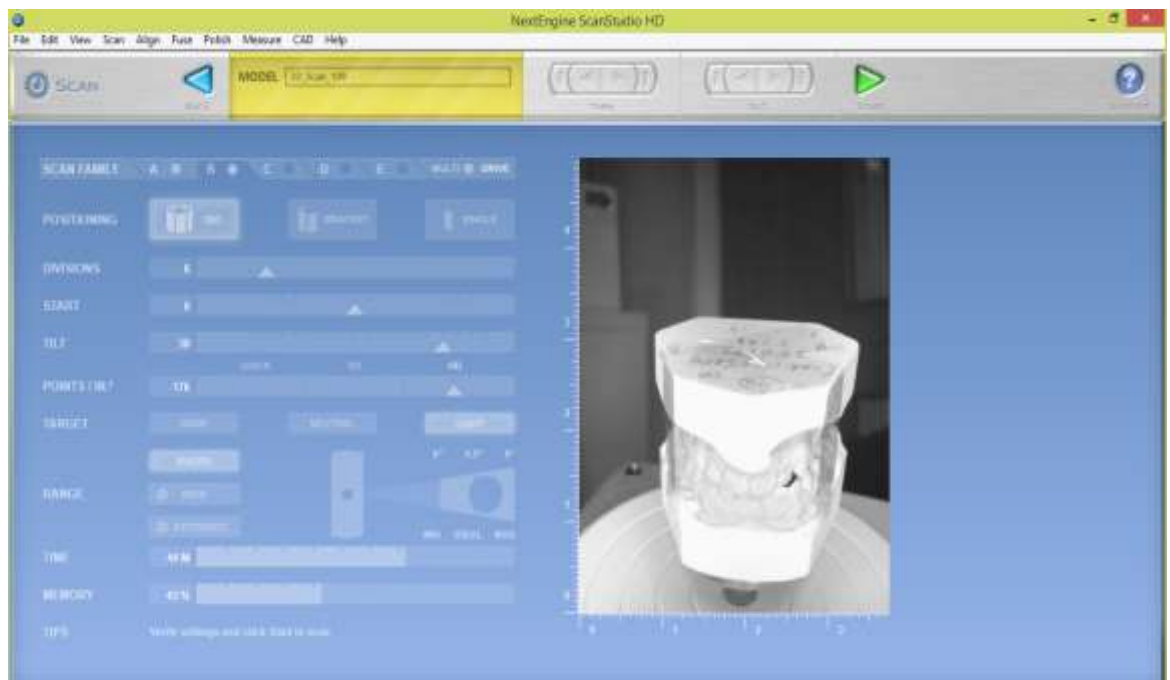


Figure 21: Screenshot showing settings for U/L models of a subject in occlusion

Figures 22 and 23 show 2D screenshots of the anterior view of the final digital models (in occlusion).

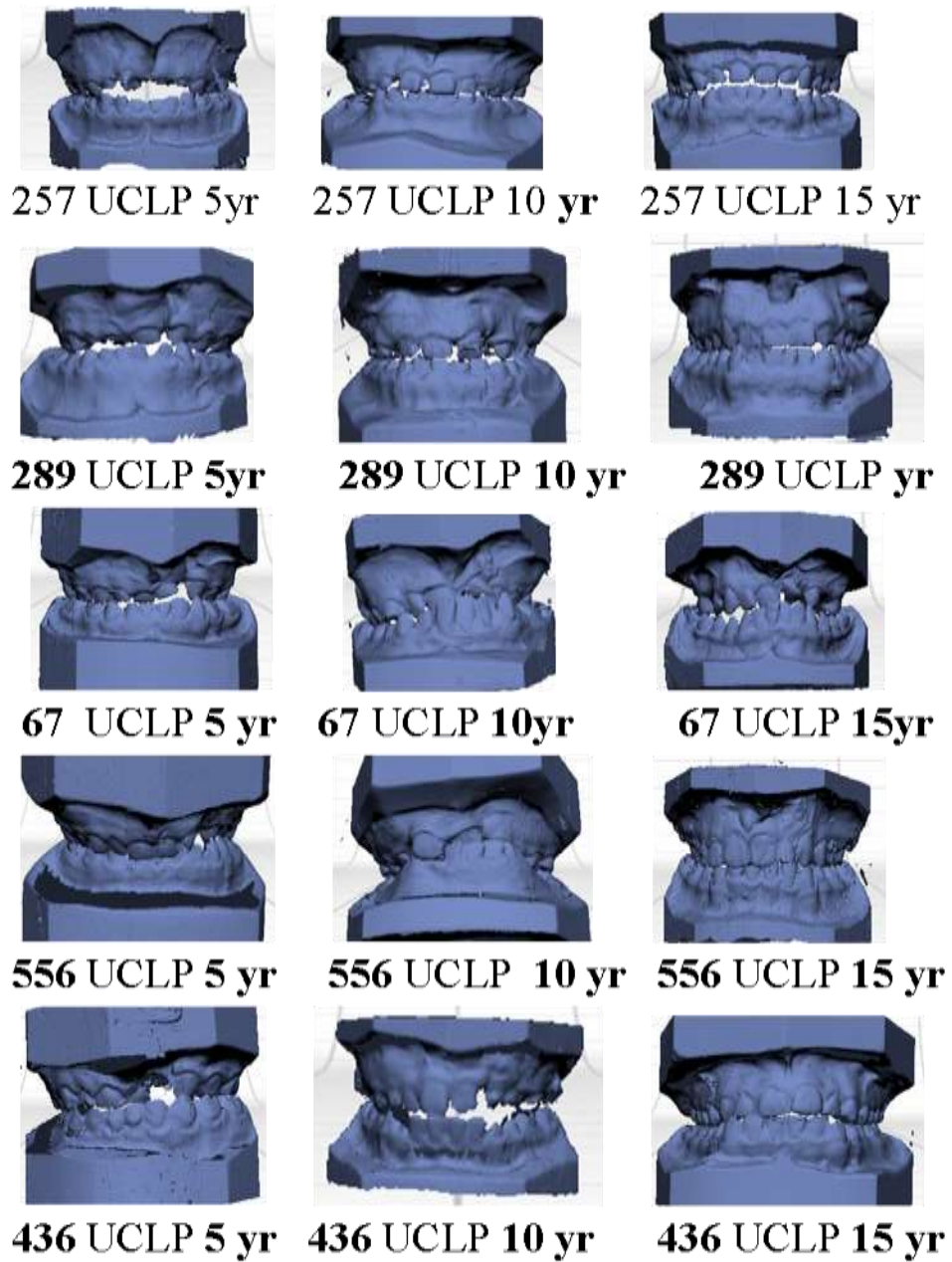


Figure 22: Series of scanned digital models of patients with UCLP

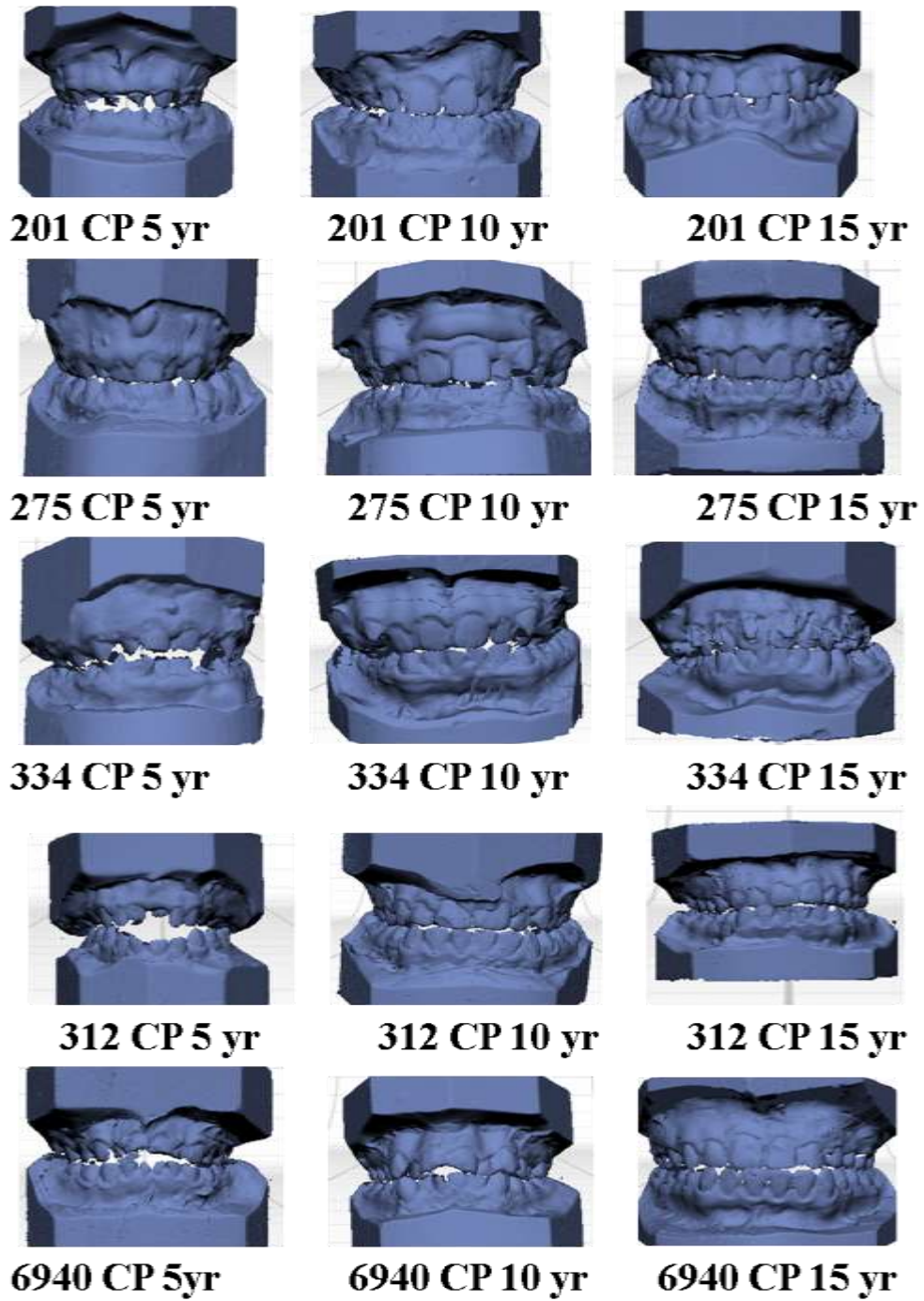


Figure 23: Series of scanned digital models of patients with CP

4.13 Description of scoring forms:

4.13.1 Modified Huddart Bodenham (MHB)

The following information was provided;

- An instruction sheet - which gave instructions on how to carry out the scoring.
- A scoring form with a list of “exceptions to the rule” provided.
- A pictorial flowchart of the MHB index.

4.13.2 GOSLON Yardstick (GY)

The following information was provided;

- An instruction sheet- which provided information how to carry out the scoring.
- The 10 sets of 5 year old and ten master GOSLON yardstick calibrated models for comparison of the CLP models (plaster and digital).

The plaster models of the five year-olds were scored first, followed by the ten year-olds and finally the fifteen year-olds for both the UCLP and CP groups; thus minimising memory bias. Similarly the digital models were scored in the same manner.

Each Examiner scored the sixty (30 plaster and 30 digital) model sets during the first round of scoring Figure 24. The duration of scoring was approximately two to three hours long. A final checklist was enclosed at the end to ensure that all the relevant sections were completed for the correct subject ID. Scoring forms were separated for the plaster and digital models to avoid any errors.



Figure 24: Scoring in progress

Instructions for carrying out the scoring method, together with scoring forms were provided for each examiner. Each examiner participated in the second round of scoring approximately three weeks later to avoid any memory bias. This allowed calculation of intra-observer reproducibility. The models were scored in the same order. The data were entered into a Microsoft Excel sheet (Redmond, California). All three Examiner data sets were then entered under PM1, PM2, GM1, GM2, NQ1 and NQ2 columns respectively.

4.14 Measurements of arch dimension

Linear measurements i.e., inter-canine and inter-molar distances were measured on plaster Figure 25 and digital Figure 26 models using digital callipers Figure 27 and MeshLab software Figure 28-30 respectively. The specification of the digital calliper is detailed in Appendix B. The Welch two sample t-test was used to calculate the statistical significance of differences between plaster and digital measurements. The mean differences of 2mm was used as a threshold value, anything above this value would be considered as clinically significant (McIntyre and Mossey, 2002). Any type of

measurements can be taken from the surface of digital dental models such as linear, angular, area and volumetric measurements or a combination of these (Foong, 2010).

INTER-CANINE AND INTER-MOLAR MEASUREMENTS



Figure 25: Upper plaster models of a subject with CP at 5, 10 and 15 years



Figure 26: Upper digital models of the same subject at 5, 10 and 15 years



Figure 27: Caliper used to measure inter-molar and inter-canine distance on plaster models

The variations in the landmark identification in present study was minimised by selecting the points on the plaster and digital models before any measurements were taken. In a previous study by Asquith et al. (2007) on digital models, prior landmark positioning was undertaken. Ten sets of plaster models were digitised by the Arius 3D Foundation system. The accuracy and inter-examiner reproducibility was evaluated by measuring eleven parameters and comparing those to the plaster models. The lower inter-canine and overjet measurements were most accurate. The process of randomisation and replication of measurements are important methods to avoid bias and random errors respectively (Houston, 1983).

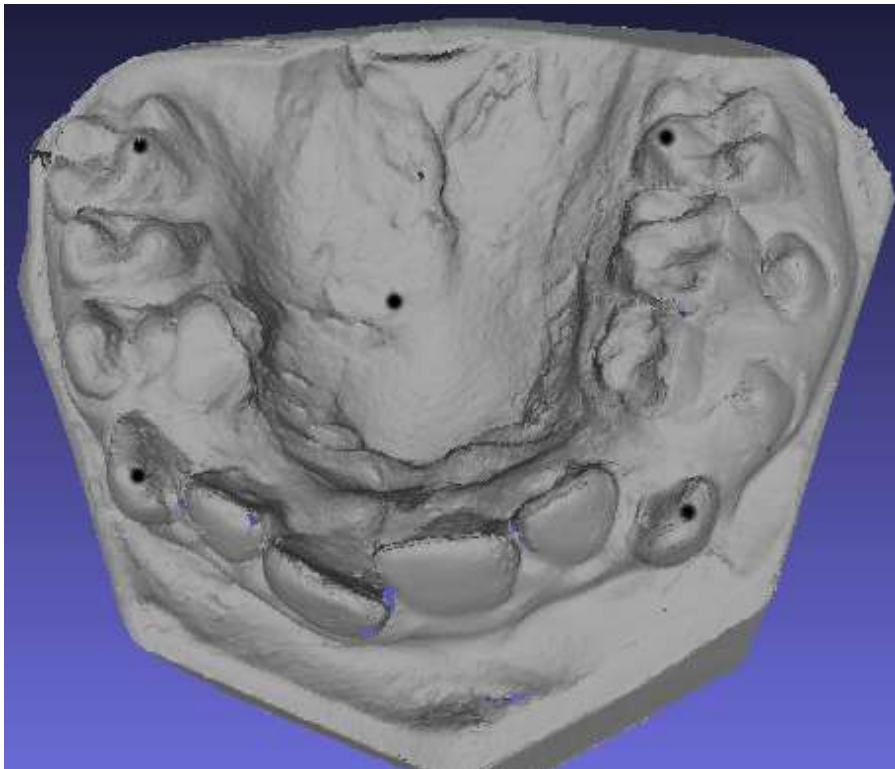


Figure 28: upper digital model with reference points

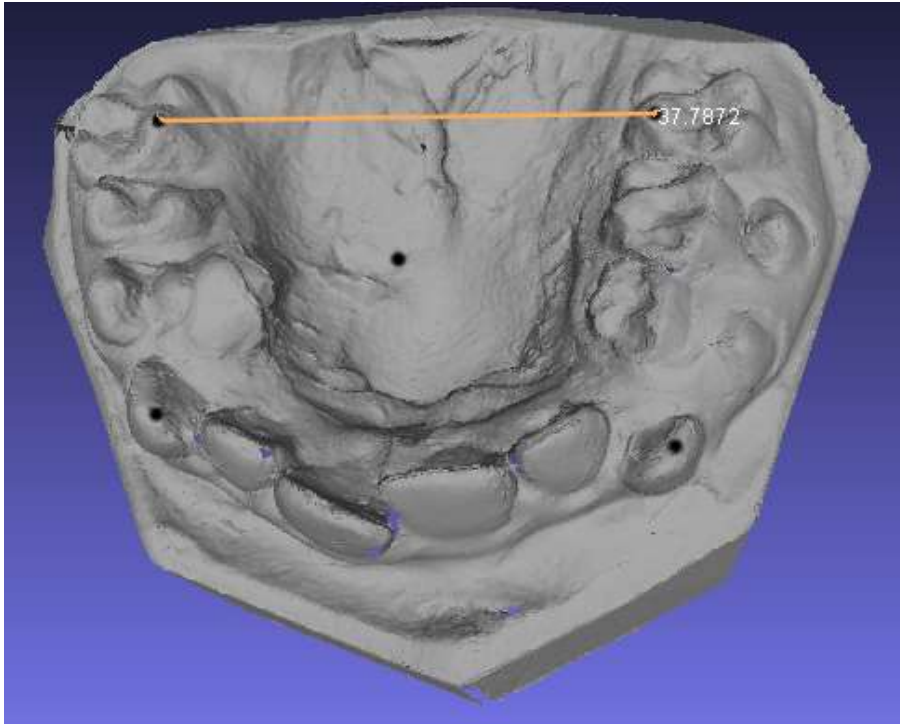


Figure 29: The inter-molar distance measured (orange line connecting the two reference points on the mesio-palatal cusp tips)

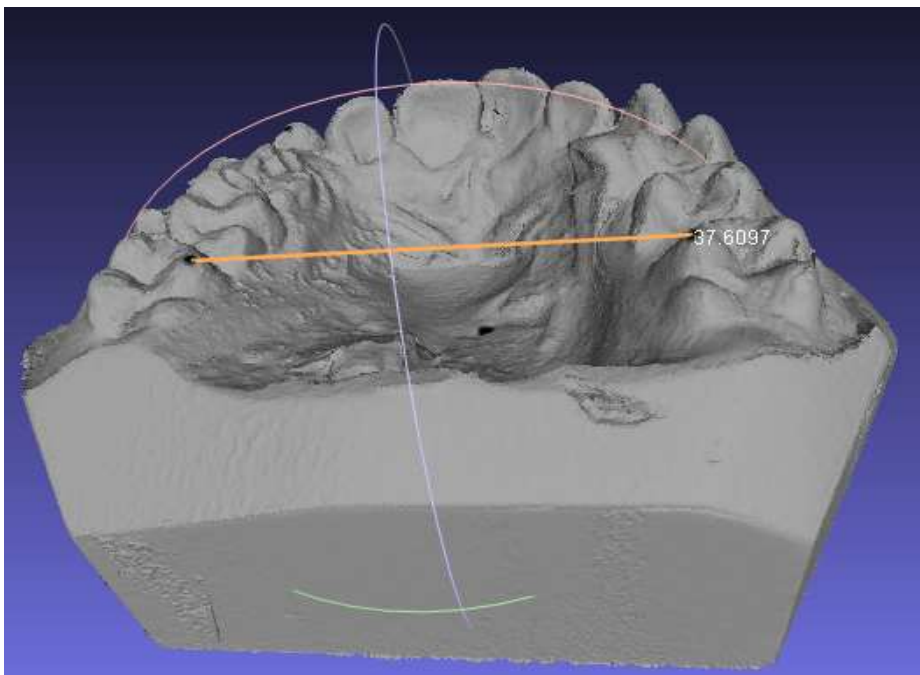


Figure 30: Distal view of the digital model showing reference line (orange)

4.15 Statistical analysis

All statistical analysis was undertaken using R studio (RStudio, 2014), which has the following advantages:

- It is powerful (Compared with SPSS and Microsoft excel).
- It is free.
- Extensive support documentation.
- It is current (being updated by professionals worldwide on a daily basis).
- Simple to learn.

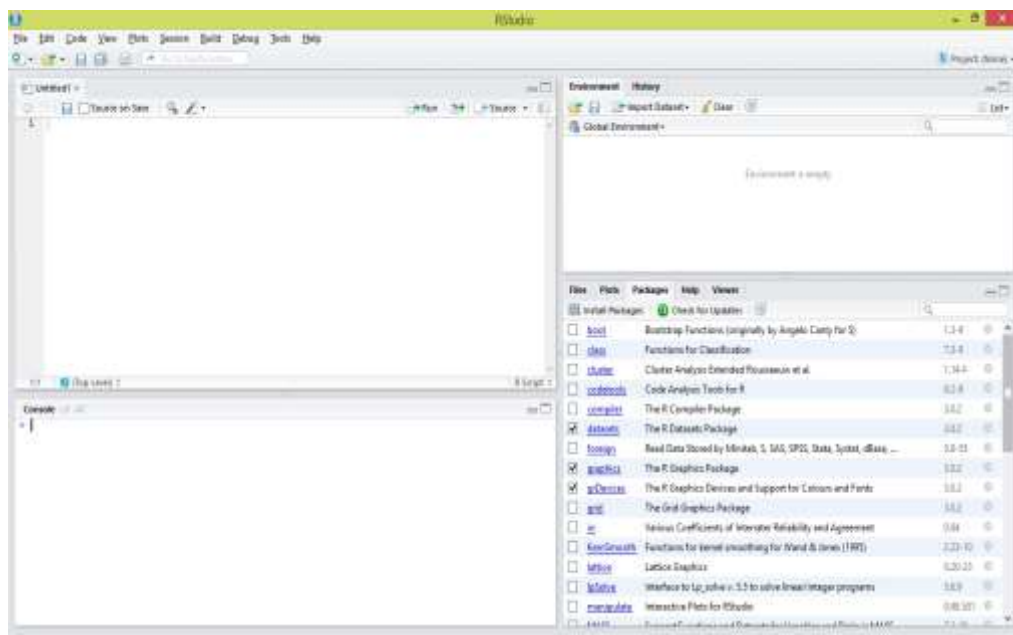


Figure 31: Generic R console is shown in the picture

Excel 2013 (Microsoft, Redmond, California) was used to organise the raw data. The final data sets were saved as csv files. These files were imported into R-console to be statistically analysed. The packages “irr” and “IpSolve” were used in conjunction with R Studio as these have codes for dentistry-related statistics. The R-console is shown in Figure 31. Following tests were run on these datasets:

- Analysis of variance statistical test (ANOVA)
- Intra-rater reproducibility
- Inter-rater reliability
- Welch t-test

A two way ANOVA was used to assess the difference in relative maxillary arch constriction with growth between the UCLP and CP groups. Weighted kappa and Kendall's correlation coefficient were calculated to determine intra-rater reproducibility and inter-rater reliability, respectively. The Welch t-test was used to compare linear measurements made of plaster and digital models.

Weighted Kappa was used with the categorical data to determine intra-rater reproducibility and inter-rater reliability. This takes into account the magnitude of the difference between scores (i.e., scores of 2 and 3 are relatively close, but 2 and 5 are a long way apart). The categorical data in the present study were 5 year old and GOSLON scores obtained from the plaster and digital models. The values were categorised according to the Altman method (Altman, 1990):

0.81 – 1.00 Very good
0.61 – 0.80 Good
0.41 – 0.60 Moderate
0.21 – 0.40 Fair
< 0.20 Poor

Kendall's correlation coefficient measures the association between the two data sets of same examiner or the data sets of different examiners. It is an estimate of intra-rater reproducibility and inter-rater reliability of ordinal data, i.e. MHB scores in the present study. The values range from -1 to +1, can be interpreted as mentioned below:

-1 - Strong negative correlation

+1 - Strong positive correlation

CHAPTER 5: RESULTS

5.1 Hypothesis 1: Relative maxillary arch constriction in patients with surgically treated UCLP and CP does not deteriorate progressively with growth.

The boxplots and histograms in Figures 32, 33 and 34, 35 illustrate the pattern of the relative maxillary arch constriction in 5, 10 and 15 years old patients with surgically repaired CP and UCLP. The boxplots in Figures 32 and 33 are constructed using combined mean 5 year old/GOSLON and MHB scores by all the three examiners on y axes. The median (black line) of each boxplot depicts the centrality of the data representing the relative maxillary arch constriction. The histograms in Figures 34 and 35, where the bars are constructed using mean 5 year old/GOSLON and mean MHB scores by each examiner individually plotted on y axes depict the relative maxillary arch constriction for each examiner individually for every patient.

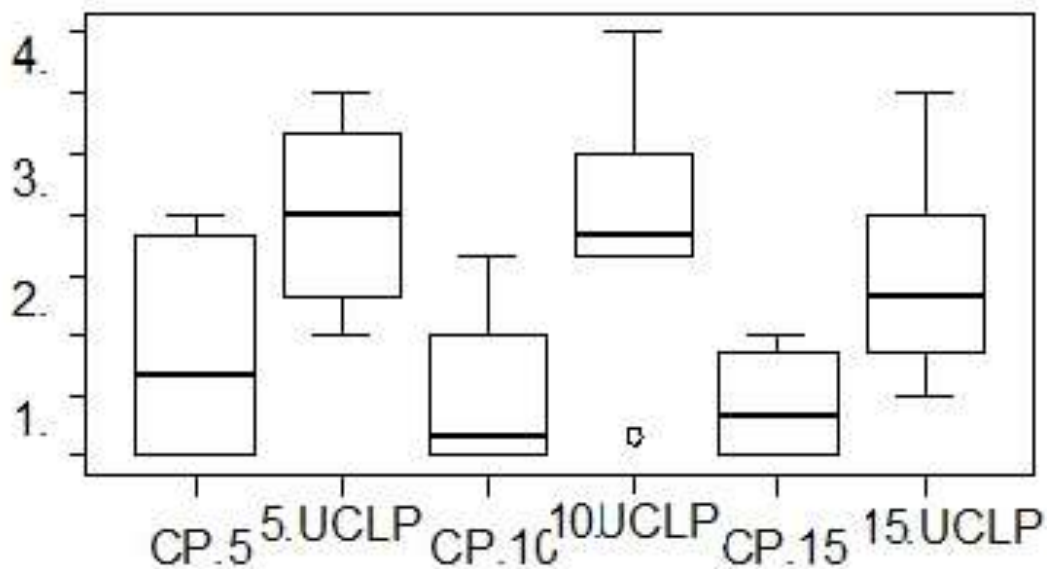


Figure 32: Relationship between combined mean plaster 5 year old/GOSLON scores (y axis) with age and group¹

¹ The black bold line in the middle of the each boxplot is the median of the data set representing relative maxillary arch constriction

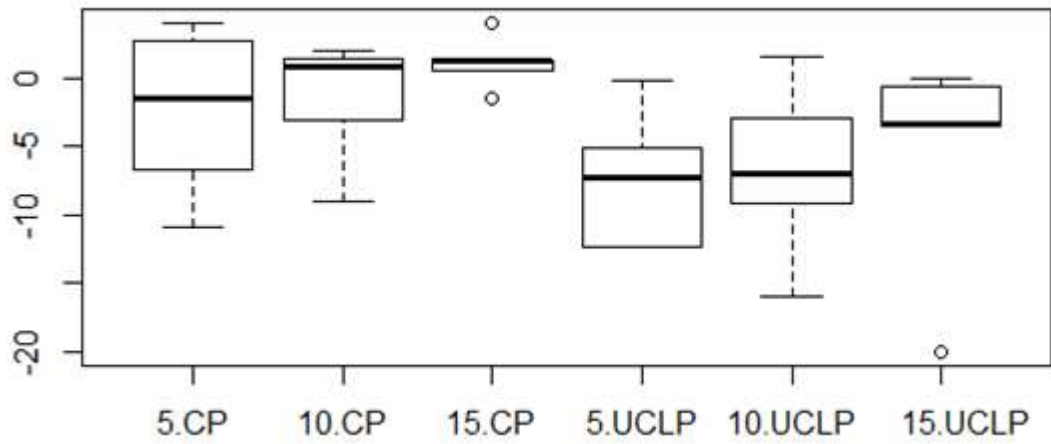


Figure 33: Relationship between combined mean plaster MHB scores (y axis) with age and group¹

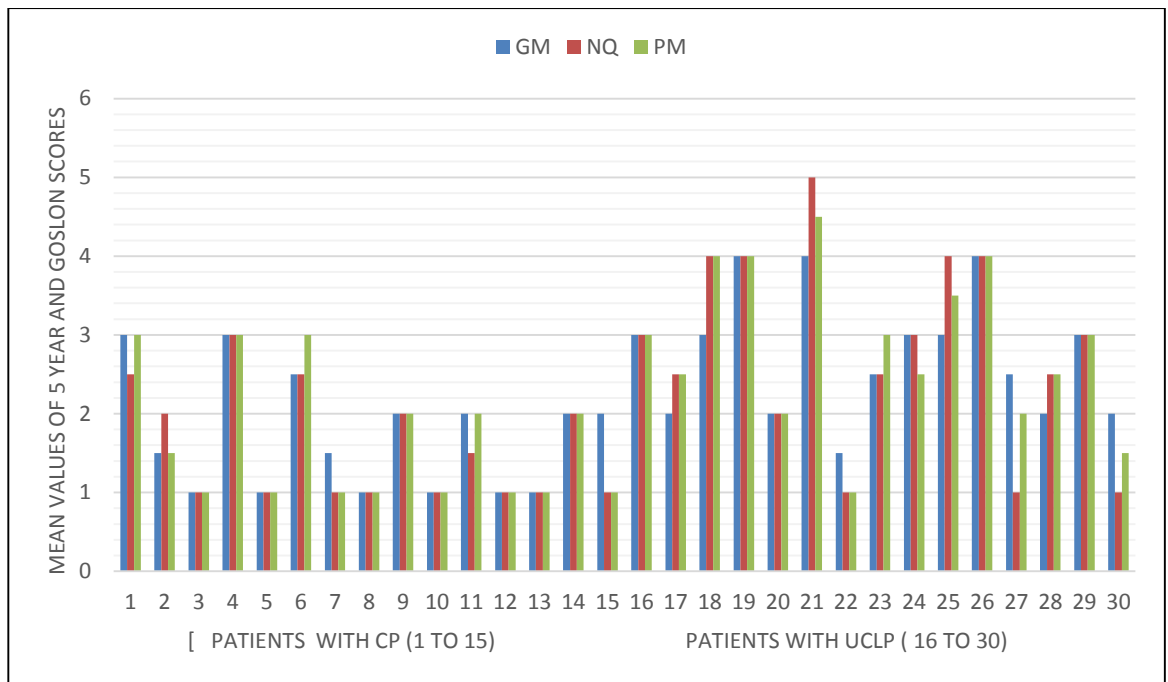


Figure 34: Distribution of mean 5 year old/GOSLON scores for plaster models of the patients at 5, 10 and 15 years²

² CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and UCLP (16 to 20: 5 year UCLP, 21 to 25:10 year UCLP, 26 to 30: 15year UCLP)

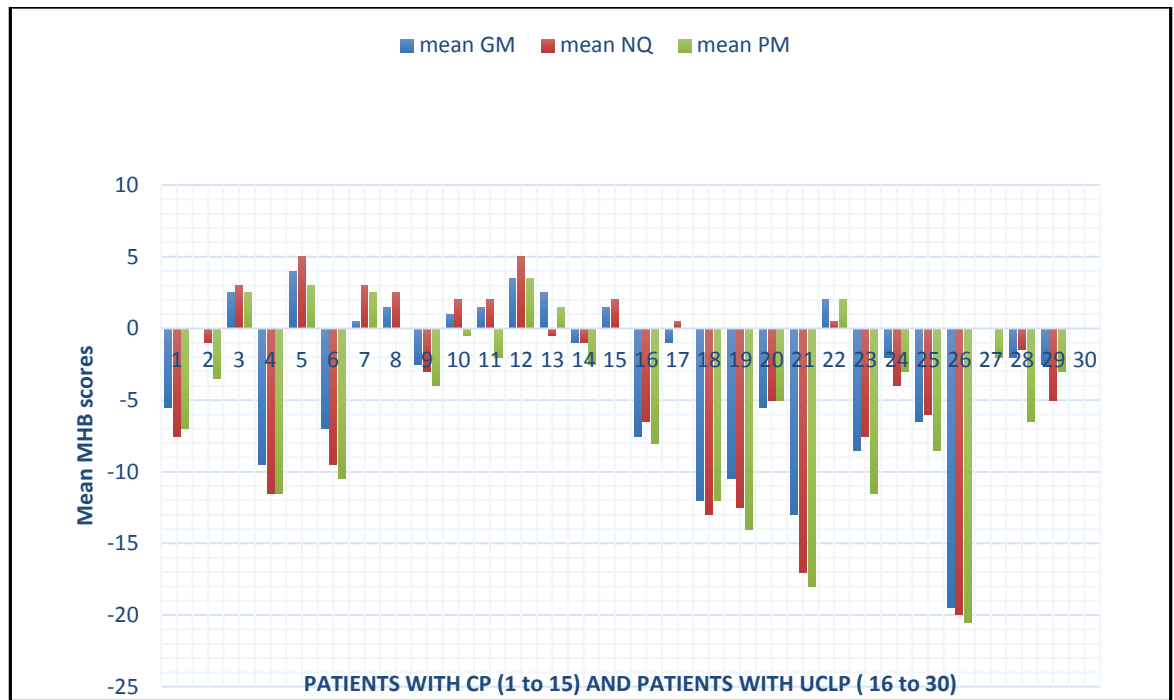


Figure 35: Distribution of mean MHB scores for plaster models for the patients at 5, 10 and 15 year³

The histograms in Figures 34, 35 and 44, 47 illustrate the relationship of mean 5 year old/GOSLON and MHB scores plotted on y axes with the age and group (CP and UCLP) on x axes demonstrated by digital and plaster models, respectively. The illustrations confirmed that the 5 year old/GOSLON and MHB scores (a measure of relative maxillary arch constriction) fluctuate between age 5 to age 15 in patients belonging to both groups. It was clear from these figures that relative maxillary arch constriction was greater in 5 year old patients and the constriction decreased in both groups at ages 10 and 15 in both CP and UCLP groups.

³ CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and UCLP (16 to 20: 5 year UCLP), (21 to 25:10 year UCLP), (26 to 30: 15year UCLP)

Model Type	Scores	Effect of age on scores p value##	Effect of group (UCLP & CP) on scores p value ##	Effect of age and group together p value ##
Plaster	5 year old/ GOSLON	0.242366	0.000935***	1.0000
Plaster	MHB	0.2813	0.0113*	0.7565
Digital	5 year old/ GOSLON	0.12132	0.00106**	0.96166
Digital	MHB	0.09810	0.00371**	0.69289

Table 11: Interaction of model type, age and cleft type (group) with scores (Measure of relative maxillary arch constriction) ⁴

The results in Table 11 show that the effect of age on 5 year-old/GOSLON and MHB scores for both plaster and digital models was not statistically significant (p-values ranging from 0.0981-0.281).

The pattern of illustrations displayed by boxplots and histograms together with p value>0.05 suggest the relative maxillary constriction does not deteriorate progressively with growth. Therefore the null hypothesis was not rejected.

5.2 Hypothesis 2: There is no difference in relative maxillary arch constriction in patients with surgically treated UCLP and CP.

⁴ ## * p < 0.05, ** p < 0.01, *** p < 0.001 denotes level of significance * Significant ** Highly significant *** Very highly significant

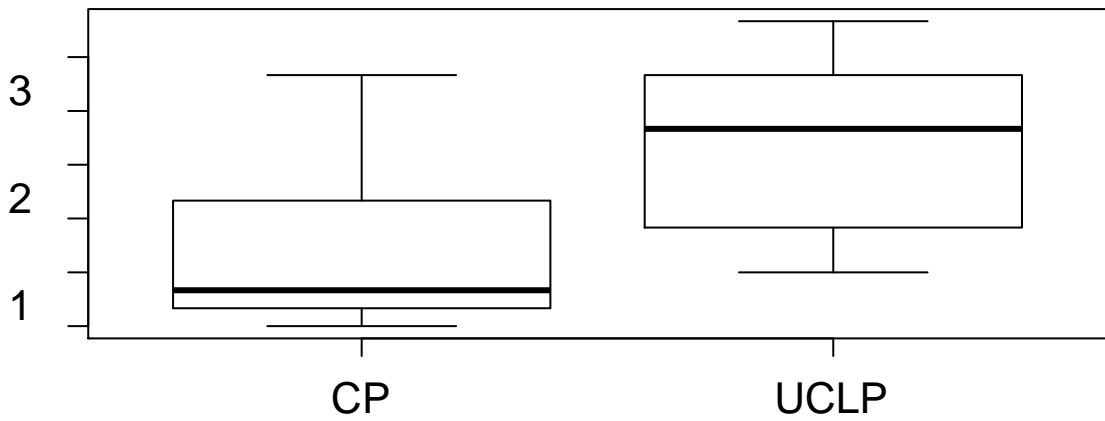


Figure 36: Relationship between combined mean digital 5 year old/GOSLON scores (y axis) and groups (CP or UCLP)

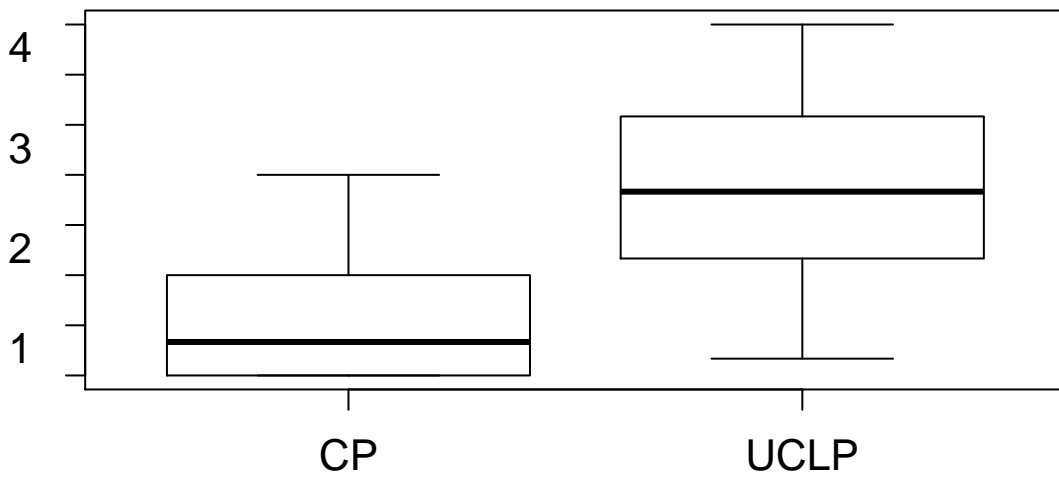


Figure 37: Relationship between combined mean plaster 5 year old/GOSLON scores (y axis) and group (CP or UCLP)

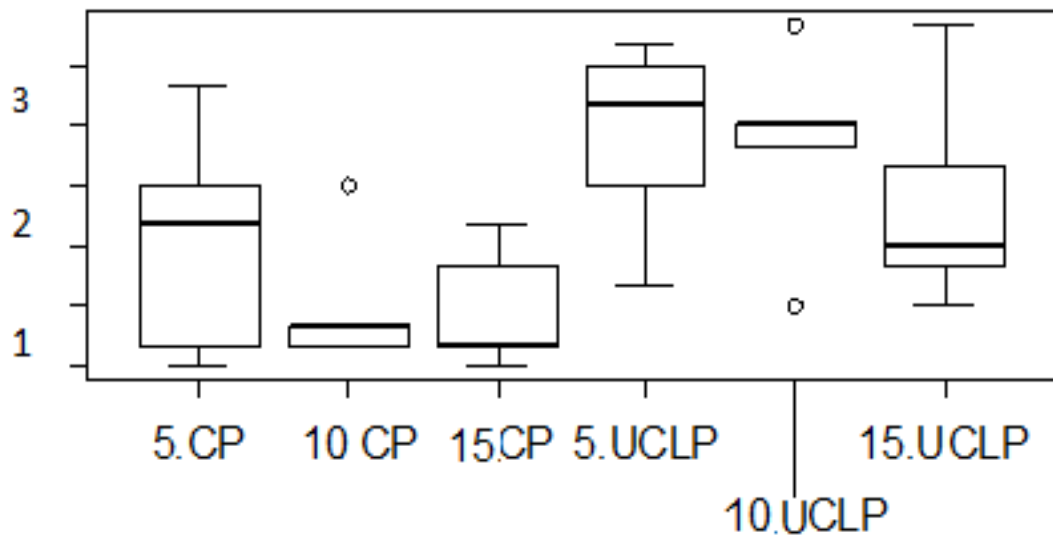


Figure 38: Relationship between combined mean digital 5 year old/GOSLON scores (y axis) with age and group⁵

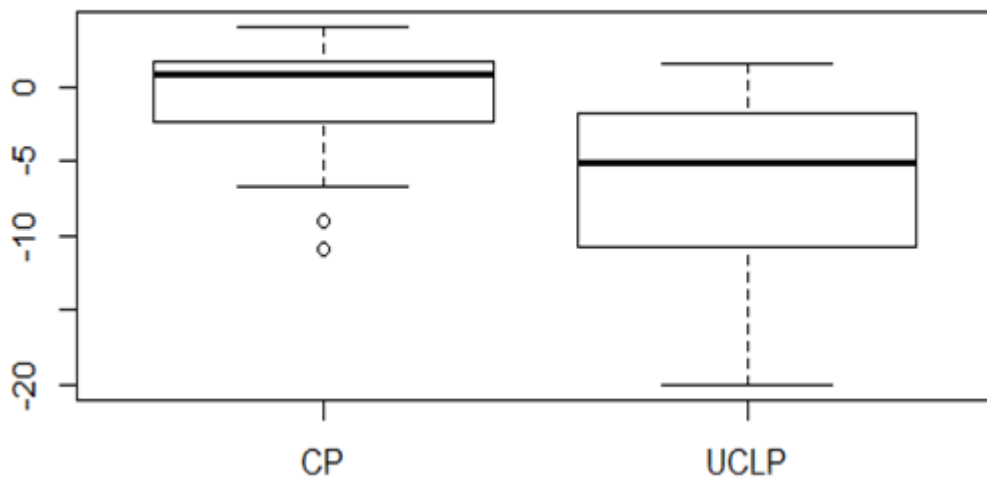


Figure 39: Relationship between combined mean plaster MHB scores (y axis) with type of group (UCLP or CP)⁶

⁵ The bold black line in the middle of the boxes is the median of each data set and represents relative maxillary arch constriction, the small circles are outliers.

⁶ The bold black line in the middle of the each box represents the median of the data set representing relative maxillary arch constriction.

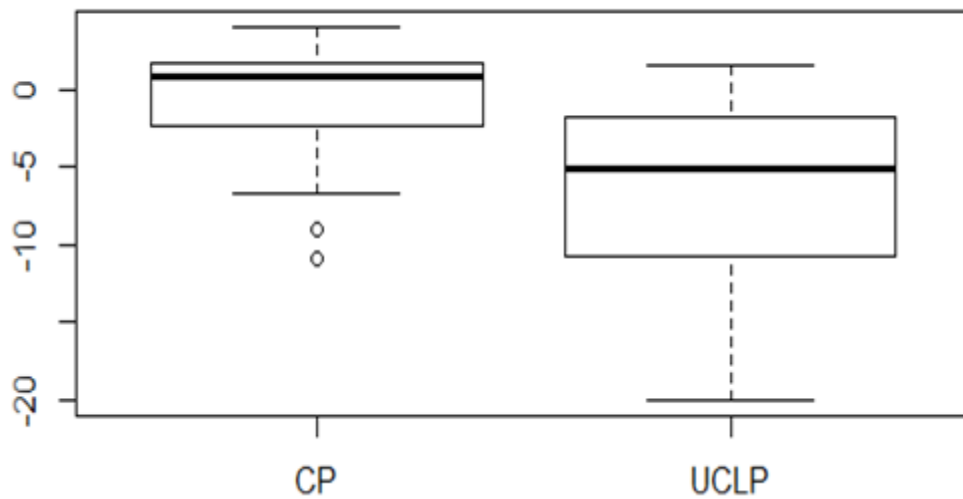


Figure 40: Relationship between combined mean digital MHB scores (y axis) with type of group (CP or UCLP)

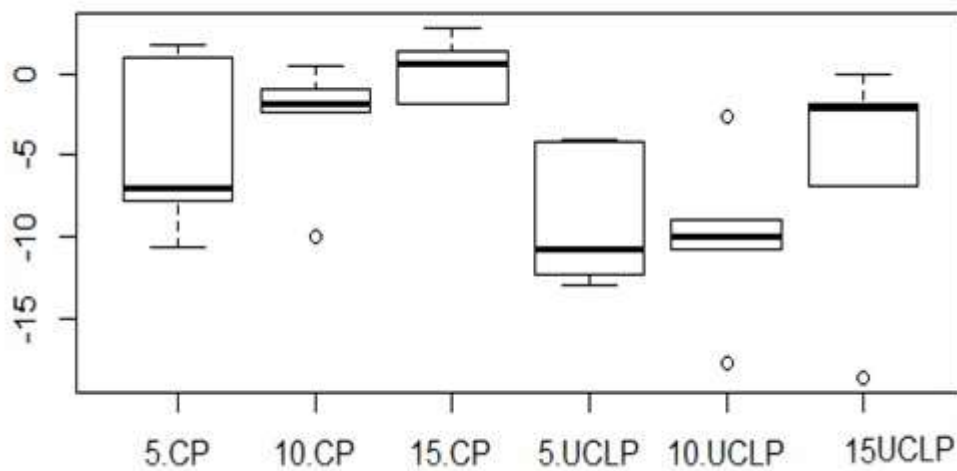


Figure 41: Relationship between combined mean digital MHB scores (y axis) with age and group⁷

⁷ The bold black line in the middle of the boxes is the median of each data set and represents relative maxillary arch constriction.

The boxplots in Figures 32, 36, 37 and 38 show the effect of group (UCLP/CP) on the combined mean 5 year old/GOSLON scores and Figures 39, 40 and 33, 41 show its effect on the combined mean MHB scores using digital and plaster models respectively. These boxplots in the figures mentioned above show that worsening of scores is greater in the UCLP group when compared to the CP group. Table 11 also shows the effect of group (UCLP and CP) on 5 year-old/GOSLON and MHB scores for both plaster and digital models was highly statistically significant (p-values ranging from 0.011-0.0009). The combined effect of age and group (UCLP and CP) together on 5 year old/GOSLON and MHB scores was not statistically significant (p-value ranging from 0.692-1.000). The histograms in Figures 34, 35 and 44, 47 also illustrate the pattern of relative maxillary constriction in both groups (CP and UCLP) as detailed in section 5.1. The constriction was greater at age 5 in both groups (CP and UCLP), the UCLP group displayed a slight decrease in relative maxillary arch constriction between age 5 and 10 followed by a substantial decrease in arch constriction between age 10 and 15. Likewise, within the CP group relative maxillary arch constriction reduced substantially between ages, 5 and 10 followed by a slight reduction in constriction at 15 year age group. This suggested that the relative maxillary arch constriction showed no particular trend for deterioration with growth in either the CP or UCLP groups. The second null hypothesis was rejected because relative maxillary arch constriction was greater in the UCLP group compared to the CP group as assessed by the 5 year old/GOSLON and MHB scores using both plaster and digital models.

5.3 Hypothesis 3: The reproducibility and reliability of the 5 year old/GOSLON and MHB indices are not different when assessed using plaster and digital models.

5.3.1 Intra-observer reproducibility:

<i>Examiner</i>	<i>Model type</i>	<i>Weighted Kappa +++</i>
<i>GM</i>	<i>Plaster</i>	<i>0.747</i>
<i>NQ</i>	<i>Plaster</i>	<i>0.845</i>
<i>PM</i>	<i>Plaster</i>	<i>0.809</i>
<i>GM</i>	<i>Digital</i>	<i>0.619</i>
<i>NQ</i>	<i>Digital</i>	<i>0.463</i>
<i>PM</i>	<i>Digital</i>	<i>0.795</i>

Table 12: Intra-observer reproducibility for 5 year old/GOSLON indices using plaster and digital models

The weighted Kappa values for the 5 year old/GOSLON indices using plaster and digital models for all the three examiners (GM, NQ and PM) ranged from 0.747 to 0.845 for plaster models and 0.463 to 0.795 for the digital models Table 12. This confirmed that the intra-observer reproducibility for this index was good to very good for plaster models and moderate to good for the digital counterparts.

<i>Examiner</i>	<i>Model type</i>	<i>Kendall's correlation coefficient</i>
<i>GM</i>	<i>Plaster</i>	<i>0.727</i>
<i>NQ</i>	<i>Plaster</i>	<i>0.767</i>
<i>PM</i>	<i>Plaster</i>	<i>0.858</i>
<i>GM</i>	<i>Digital</i>	<i>0.752</i>
<i>NQ</i>	<i>Digital</i>	<i>0.698</i>
<i>PM</i>	<i>Digital</i>	<i>0.667</i>

Table 13: Intra-observer reproducibility of MHB scores using plaster and digital models

The Kendall's correlation coefficient for the MHB index ranged from 0.727 to 0.858 for plaster models and 0.667 to 0.752 for digital models Table 13. This confirmed that intra-observer reproducibility was substantial when using plaster models and

moderate for the digital counterparts. It can be deduced that the level of reproducibility is different for the two model types.

5.3.2 Inter-observer reliability:

The reliability of the 5 year old/GOSLON indices using plaster and digital models was assessed between the three observers pairwise. The inter-rater reliability for 5 year old/GOSLON index was determined by the Weighted Kappa statistic. The weighted kappa values ranged from 0.727 to 0.853 for plaster models, whilst the kappa values for digital models ranged from 0.448 to 0.671 Table 14. This confirmed that 5 year old/GOSLON indices are moderately reliable on digital models, whereas the level of reliability on plaster models was good.

<i>EXAMINERS</i>	<i>MODEL TYPE</i>	<i>WEIGHTED KAPPA++</i>
<i>GM-NQ</i>	<i>Plaster</i>	<i>0.727</i>
<i>GM-PM</i>	<i>Plaster</i>	<i>0.778</i>
<i>PM-NQ</i>	<i>Plaster</i>	<i>0.853</i>
<i>GM-NQ</i>	<i>Digital</i>	<i>0.448</i>
<i>GM-PM</i>	<i>Digital</i>	<i>0.671</i>
<i>PM-NQ</i>	<i>Digital</i>	<i>0.558</i>

Table 14: Inter-observer reliability for 5 year old/GOSLON scores using plaster and digital models for patients with CP and UCLP

Figures 42 and 43 show the scatter plots for the 5 year old/GOSLON scores using digital and plaster models respectively. The plots show that the majority of data points from the three examiners are superimposed, confirming the corresponding level of the agreement between the examiners using these indices.

The trendline is the best fit line that mimics the trend of the data. The trendline is more reliable when R- squared value is at or close to 1. The R- squared values are given in Tables 16, 17. The trendline for individual examiners demonstrated the agreement

between the examiners, particularly as the trend lines for GM and PM were almost superimposed in Figures 42 and 43.

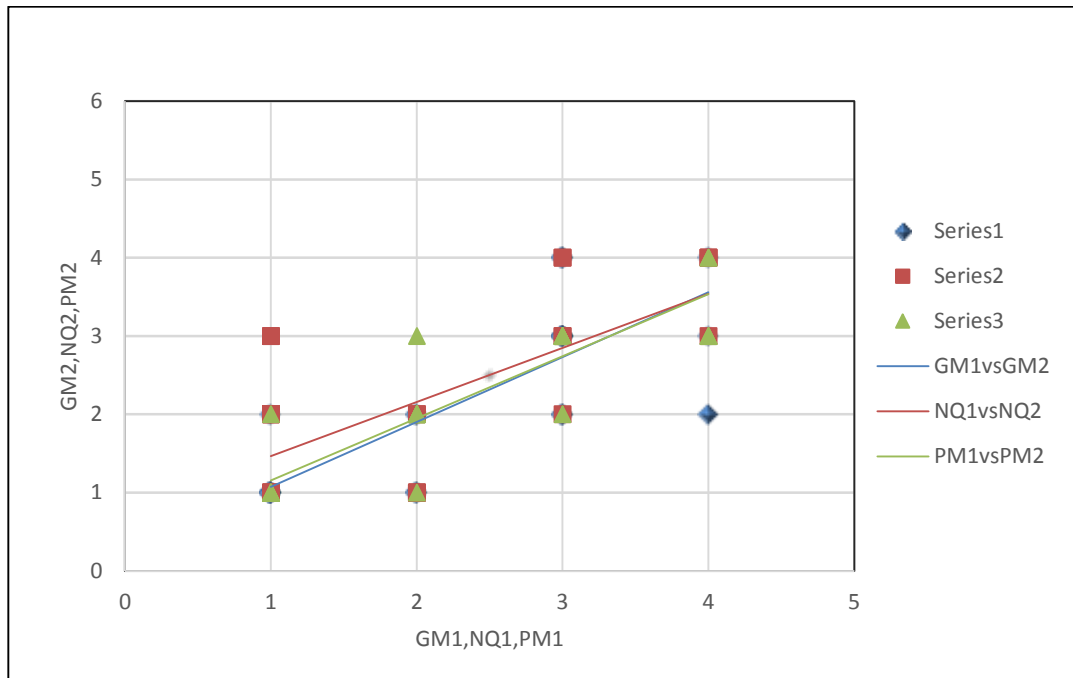


Figure 42: Scatter plot with three trendlines showing distribution of 5 year old/GOSLON scores by GM, NQ and PM for digital models of patients with CP and UCLP at two rounds of scoring

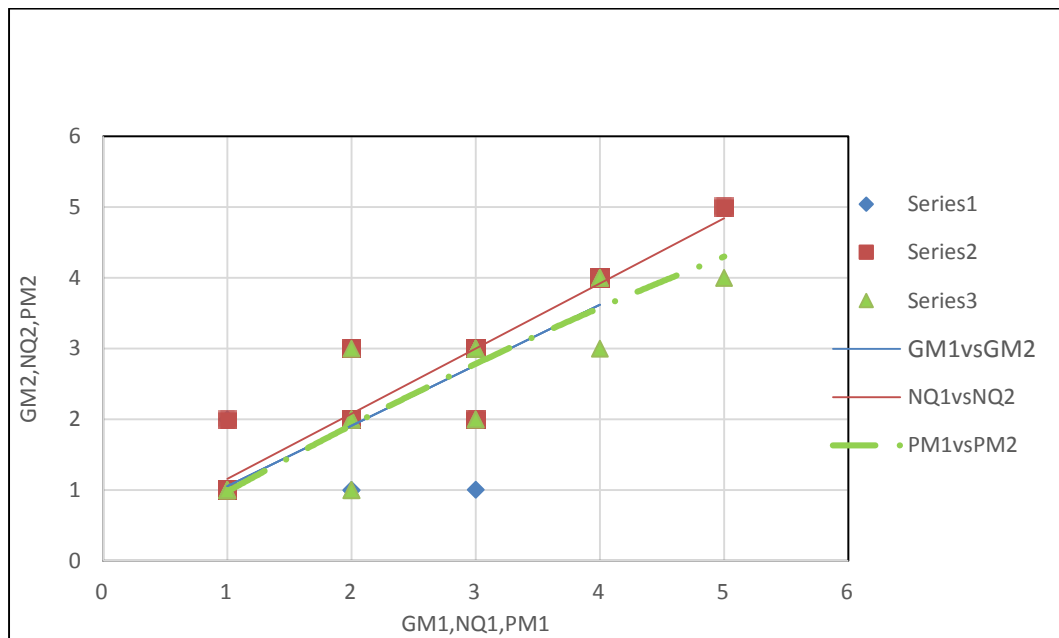


Figure 43: scatter plots with three trendlines showing distribution of 5 year old/GOSLON scores by GM, NQ and PM using plaster models for patients with CP and UCLP at two rounds of scoring

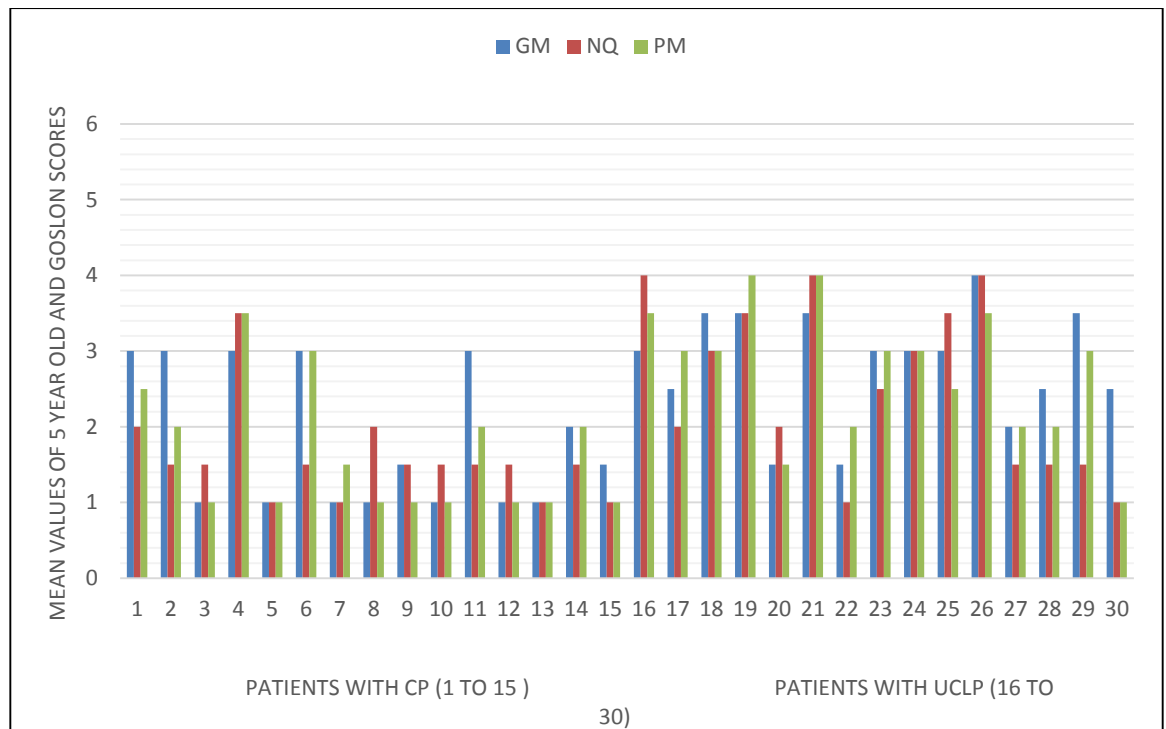


Figure 44: Distribution of mean 5 year old/GOSLON scores for digital models⁸

The histograms in Figures 34 and 44 show the distribution of the mean 5 year old/GOSLON scores (y axes) using plaster and digital models of 5, 10 and 15 year old patients with CP and UCLP.

5.3.3 Inter- observer reliability for MHB Score using plaster and digital models

For inter-observer reliability of the MHB index, Kendall's correlation coefficient was calculated to determine the level of agreement between observers Table 15. The values ranged from 0.847 to 0.864 using plaster models and from 0.665 to 0.714 using digital models. This indicated a substantial agreement among examiners when using the MHB index on plaster models compared to moderate agreement on digital models. The difference between the values was narrower when compared to 5 year old/GOSLON indices using plaster and digital models.

⁸ Patients with CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and patients with UCLP (16 to 20: 5 year UCLP), (21 to 25:10 year UCLP), (26 to 30: 15year UCLP) plotted side by side on x axis

EXAMINERS	MODEL TYPE	KENDALL COEFFICIENT
GM-NQ	Plaster	0.857
GM-PM	Plaster	0.864
PM-NQ	Plaster	0.847
GM-NQ	Digital	0.665
GM-PM	Digital	0.714
PM-NQ	Digital	0.686

Table 15: Inter-observer reliability of MHB scores using plaster and digital models for patients with CP and UCLP

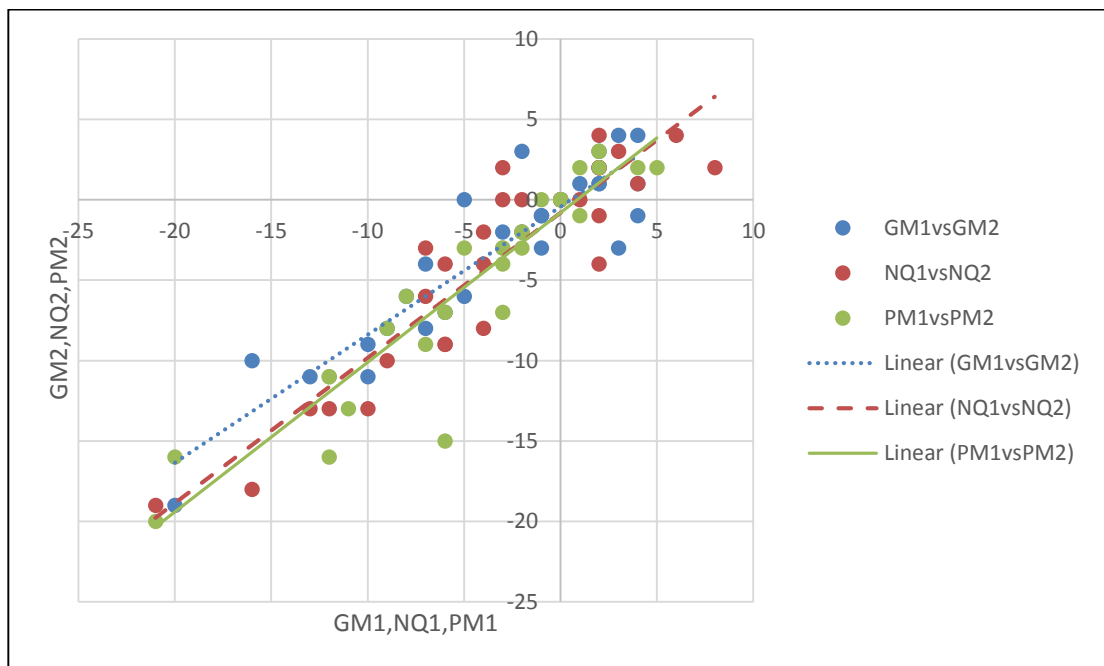


Figure 45: Scatter plot with three trendlines showing the distribution of MHB scores by GM, NQ and PM at two rounds of scoring using plaster models of Patients with CP and UCLP

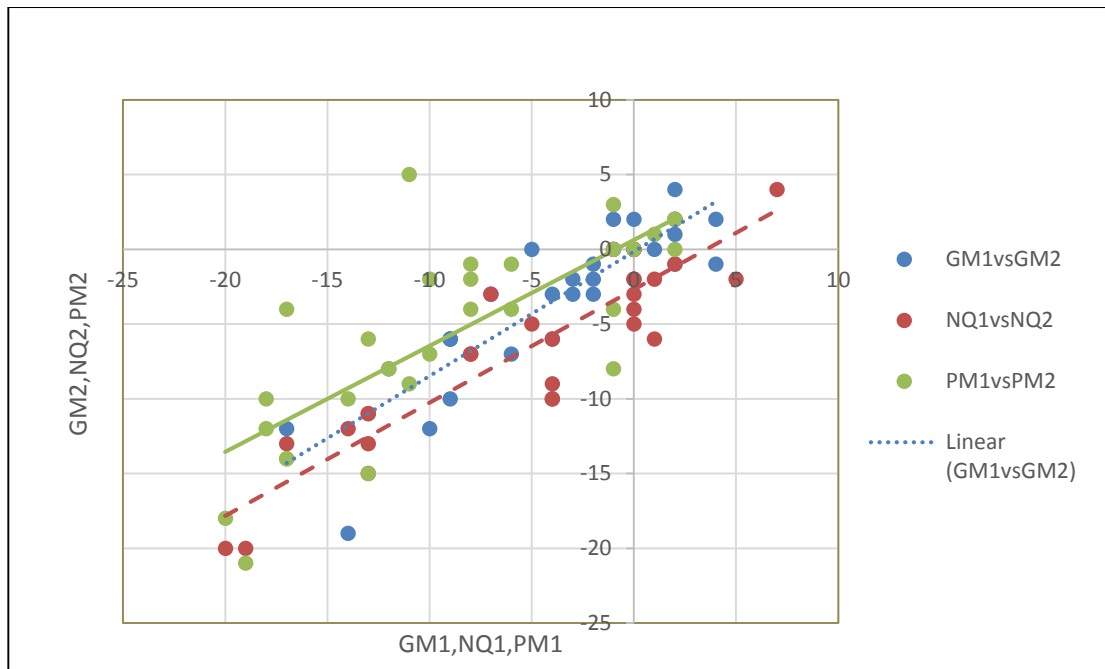


Figure 46: Scatter plot with three trendlines showing the distribution of MHB scores by GM, NQ and PM at two rounds of scoring using digital models of patients with CP and UCLP

The scatter plots in Figures 45 and 46 show the distribution of MHB scores taken at two rounds of scoring by all the three examiners plotted on the x and y axes. The data points in Figure 46 are scattered compared to linear distribution of data points in Figure 45 suggesting that the examiners demonstrated a good agreement for scoring plaster models than digital models when using the MHB index.

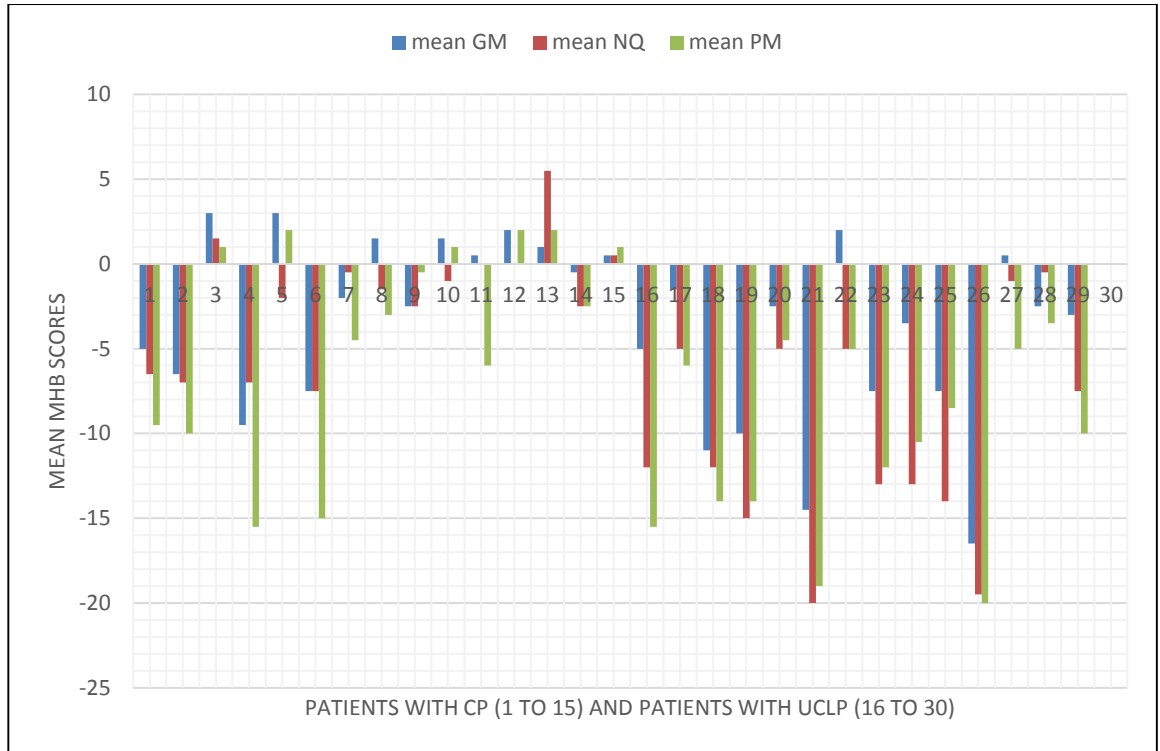


Figure 47: Distribution of mean MHB scores (y axis) for digital models⁹

The histograms in Figures 35 and 47 show the distribution of the MHB mean scores recorded on plaster and digital models of patients with CP and UCLP at age 5, 10 and 15 years.

It was concluded that the 5 year old/GOSLON and MHB indices are well correlated (reproducible/reliable) when assessed using plaster models and the correlation decreased when the indices were used on digital models. Furthermore, the MHB index is comparatively more reliable when assessed using digital models as compared to 5 year old/GOSLON index. The null hypothesis was rejected as there was a difference in the reproducibility/reliability of these indices when assessed on plaster and digital models.

⁹ Patients with CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and patients with UCLP (16 to 20: 5 year UCLP), (21 to 25:10 year UCLP), (26 to 30: 15year UCLP) plotted side by side on x axis

5.3.4 Reliability of trendlines

The reliability of trendlines in scatter plots in Figures 42, 43 and 45, 46 is determined by the R squared values which are given in the Tables 16, 17. The trendline determines the relationship of the data scored at two rounds of scoring.

Examiner	R Squared value (plaster)	R Squared value (digital)
GM	0.683	0.601
NQ	0.864	0.469
PM	0.843	0.810

Table 16: R squared values of trendlines using 5 year old/GOSLON indices for plaster and digital models

Examiner	R Squared value (plaster)	R Squared value (digital)
GM	0.836	0.776
NQ	0.843	0.825
PM	0.868	0.610

Table 17: R squared values of trendlines using MHB index for plaster and digital models

5.4 Hypothesis 4: There is no difference in the linear measurements using plaster and digital models.

Linear dimensions	Model type	Mean -value of dimensions	p-value
Inter-canine width	Plaster	27.58 mm	0.529
Inter-canine width	Digital	28.24 mm	
Inter-molar width	Plaster	38.38mm	0.848
Inter-molar width	Digital	38.51mm	

Table 18: Mean value and p-value of linear dimensions measured on plaster and digital models of patients with CP and UCLP

The differences between the linear distance measurements made on the upper plaster and digital models were not statistically significant ($P > 0.05$) with the mean differences being less than 1mm Table 18.

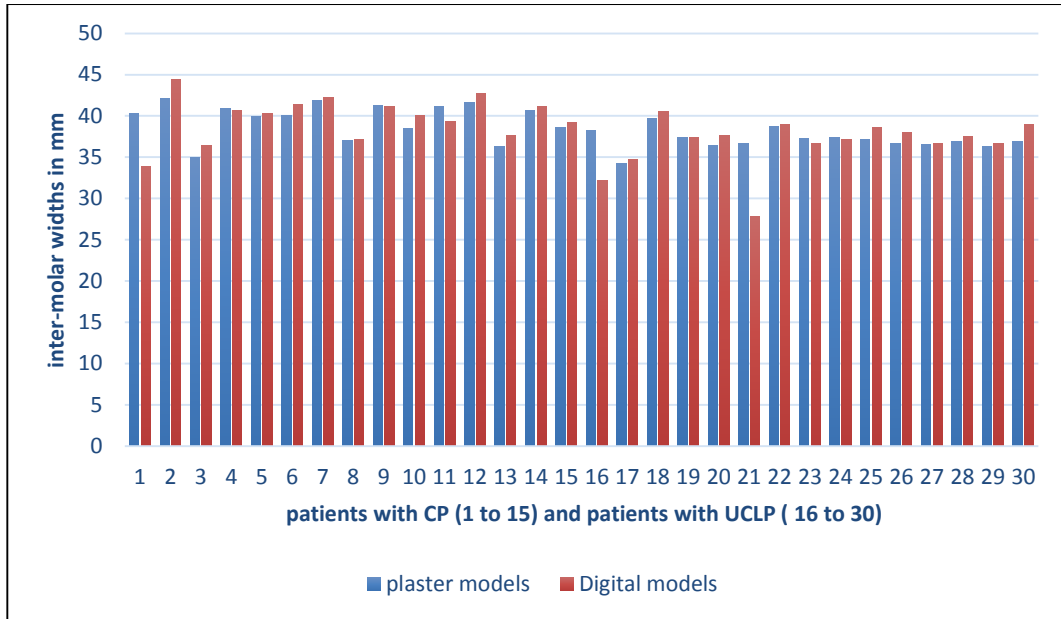


Figure 48: Inter-molar dimensions (plotted on the y axis) for plaster and digital models¹⁰

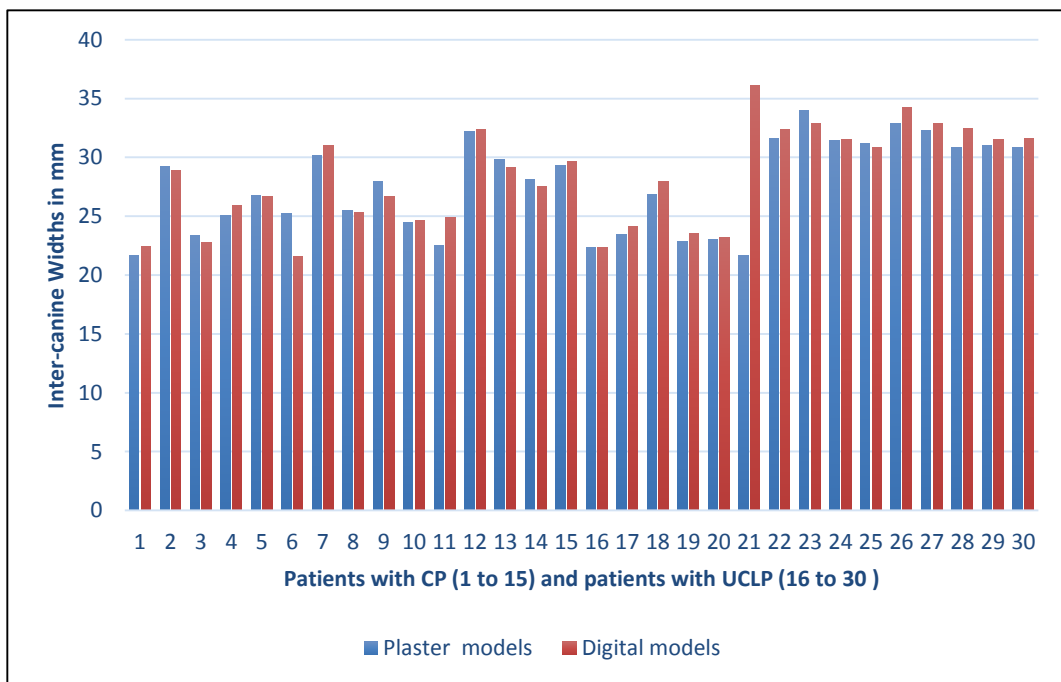


Figure 49: Inter-canine widths measured (plotted on the y axis) for plaster and digital models¹¹

¹⁰Patients with CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and patients with UCLP (16 to 20: 5 year UCLP), (21 to 25:10 year UCLP), (26 to 30: 15year UCLP) plotted side by side on x axis

The aim of measuring the inter-molar and inter-canine distances was to evaluate the accuracy of the measurements made on digital models and compared it to the gold standard i.e. plaster models. The inter-molar and inter-canine measurements were consistent on both plaster and digital models except for few patients as depicted by the height of the bars of histograms in Figures 48 and 49. Therefore this null hypothesis was not rejected.

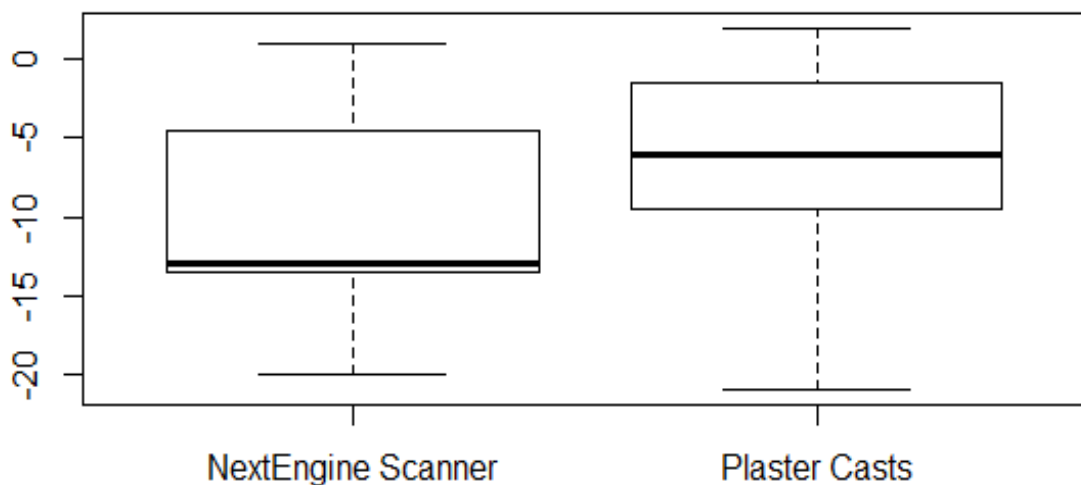


Figure 50: Distribution of the MHB scores (y axis) for the digital models obtained from the NextEngine scanner and corresponding plaster models (Cochrane, 2014)¹²

The boxplots in the Figure 50 compare the MHB scores from digital models produced from the NextEngine scanner and plaster models. The median of the two boxplots show a marked difference, suggesting the two datasets are substantially different which was statistically significant ($p < 0.05$). The Scatter plots show the distribution of data points and the Q-Q plots illustrated the distribution of data points on or around the ideal model. These figures are shown in the appendices H-O.

¹¹ Patients with CP (1 to 5: 5year CP), (6 to 10: 10 year CP), (11 to 15: 15 year CP) and patients with UCLP (16 to 20: 5 year UCLP), (21 to 25:10 year UCLP), (26 to 30: 15year UCLP) plotted side by side on x axis.

¹² The bold black line in the middle of the boxplots represent the median for the dataset

CHAPTER 6: Discussion

The findings of the present work suggested that relative maxillary arch constriction did not deteriorate progressively with growth, although this was greater in patients with UCLP than patients with isolated CP. Furthermore, the indices (5 year old/GOSLON and MHB) were more reproducible and reliable using plaster models than digital models. However, inter-observer reliability was superior for the MHB index when compared to the 5 year old/GOSLON indices when using plaster models. The MHB index performed well on digital models compared to the 5 year old/GOSLON index.

6.1 Relative maxillary arch constriction and growth

The first null hypothesis was not rejected because there was no statistically significant difference between the 5 year old/GOSLON and MHB scores (a measure of relative maxillary arch constriction) and growth ($p > 0.05$) Table 11. This explains that growth and relative maxillary arch constriction are independent of each other.

A study was conducted by (Enlow and Bang, 1965) to evaluate the post-natal growth of maxilla, they observed that as the maxilla grow in size, complex variety of remodeling movements take place simultaneously. The purpose of remodeling is to maintain the constant over-all shape of the maxilla and its component parts. The maxillary dental arch increases in size following the principle of the V, which involves apposition of bone on palatal side and resorption on entire labial and buccal side. The maxillary tuberosity lies posterior to the dental arch contributes to a major portion of the growth of maxilla. The palatine processes also follow a similar V principle for their growth, where apposition takes place on the oral side and resorption on nasal side. The growth of the maxilla is also influenced by growth taking place at the sutures including the palatine bones, the resulting growth by this activity is ceased by about the seventh year of life (Weinmann and Sicher, 1955). These research studies give an insight about the complex nature of maxillary growth, so any disturbance (iatrogenic) in the sutural growth activity could result in maxillary retrusion. The growth of maxilla parallels that of the mandibular growth but latter is continuously moved in a forward direction. The details of growth in maxilla in normal subjects and patients with CLP is given in section 1.1 and 1.2. It was noted in the present study that the surgically treated 5 year old patients had a relatively constricted maxilla in relation to the

mandible in both groups (UCLP and CP) with the level of constriction showing a decreasing trend in all patients at age 10. The decrease in relative maxillary arch constriction continued to age 15 as confirmed by the lower ranks of GOSLON scores and the near zero or positive scores for the MHB index data when compared to the corresponding scores at age 5 and age 10. Figures 32 and 33 illustrate that relative maxillary arch constriction as represented by the median of each boxplot demonstrated a similar trend for scores in both groups for age 5 in Figures 34 and 35. It can be observed from the present study that relative maxillary arch constriction is improving at the age 15 in both groups, keeping in mind that the orthodontic treatment is highly likely to have increased the width of the arch during (10 to 15 years) although growth of maxilla and mandible does not stop at this age. A larger sample of adult patients with treated CLP (with a known treatment history) could be taken in future to study the effect of growth on relative maxillary constriction more precisely.

The maxillary arch constriction in relation to the mandibular arch is the most common long-term complication of cleft surgery, with the aetiology being manifold. Many researchers believe relative maxillary arch constriction to be iatrogenic (mainly caused by scarring resulting from primary surgery) while others attribute this to be an intrinsic growth deficiency in patients with CLP. Mello et al. (2013) found that infants born with UCLP had increased maxillary dimensions compared to infants without clefts yet another study (Nyström and Ranta, 1989) reported patients aged 3 year old with surgically treated cleft palate had crowded maxillary arches when compared to children without clefts suggesting that the primary surgery may be the cause of the crowding. Mars and Houston (1990) further investigated this finding in children (aged 13 years and over) with a surgically treated UCLP and compared them to untreated patients with UCLP. They concluded that the maxillary growth deficiency was attributed to the surgical intervention (mostly palatal repair) as untreated patients had normal maxillary growth. Xu et al. (2015) investigated surgical protocols involving one stage or two stage palatal repair and its effect on maxillary growth in patients with UCLP. They concluded that the position and the sagittal length of maxilla is adversely affected by both procedures, furthermore the early closure of the palate has a

deleterious effect on maxillary growth. So the arguments favouring the involvement of surgical procedures as a cause of maxillary arch constriction outweigh those which say that intrinsic growth deficiency is the cause. Some argue that delaying the closure of hard palate allows the maxilla to grow normally but affects the development of speech in such patients. Lai et al. (2015) claims that agenesis of maxillary lateral incisors is a strong indicator of severe maxillary hypoplasia and intrinsic growth deficiency in patients with CL/P, and the absence of these teeth is also a predictor of a need for a Le Fort I advancement surgery in patients aged 14 years and over.

A Swedish study which was conducted on patients aged 19.2 - 29.5 years with treated CLP, occlusal stability was evaluated among other aspects of CLP and compared to patients without clefts. It was found that patients with treated UCLP aged between 19 and 25 years showed substantial deterioration in the occlusal scores and maxillary arch dimensions compared to non-cleft subjects, the changes were independent of the type of retention used (Marcusson, 2001). This finding should be further investigated in future by a retrospective/prospective longitudinal study to include the adult patients, as the maxillary arch constriction seems to continue until adulthood.

The present study was a retrospective longitudinal study which not only identified differences between two cleft types regarding relative maxillary arch constriction, but also highlighted the growth pattern of the maxilla in these cleft types over a period of ten years suggesting that there is no specific trend of maxillary growth deterioration with time. Patients with crossbites (which are also an indicator of maxillary constriction) generally have poor masticatory muscle control during chewing (Li et al., 1998). When the maxilla is constricted to the extent that there is a unilateral crossbite, the mandibular deviation may result, which if left untreated could lead to temporomandibular dysfunction syndrome (TMPDS). The crossbite in patients with CLP can be associated with speech problems such as defective articulation (Laitinen, 1999). There is no clear-cut evidence regarding the relationship between maxillary constriction and the aetiology of the upper airway problems such as obstructive sleep apnoea (OSA) but some morphological differences exist between patients with OSA and control subjects thereby associating the role of maxillary constriction as an etiological factor for OSA (Johal and Conaghan, 2004).

6.2 Relative maxillary arch constriction and cleft sub-phenotypes

The effect of cleft sub-phenotype on 5 year old/GOSLON and MHB scores (a measure of relative maxillary arch constriction) was highly statistically significant (p values ranging from 0.01 to 0.0009) Table 11. The relative maxillary arch constriction was illustrated as shown in Figures 36, 37, 38, 39, 40, 41 in patients with CP and UCLP using both plaster and digital models, the p value <0.05 and the illustrations shown in Figures (boxplots and histograms) confirmed the relative maxillary arch constriction was greater in patients with surgically treated UCLP when compared to patients with surgically treated CP. The difference between 5 year old/GOSLON and MHB scores according to these two CLP sub-phenotypes could have various causes. The tissue defect seem to be greater in patients with UCLP compared with patients with CP. Many studies have been conducted to investigate the aetiology of the reduced maxillary arch dimensions in patients with treated UCLP than patients with treated isolated CP. The embryological development of lip and palate is of paramount importance in understanding the etiology of cleft lip and palate, the details of which is given in section 2.3. According to Moss (1972) the growth and development of the skeletal component is determined by the interplay between the genetic and environmental factors with a significant contribution from the soft tissue components such as the lips, tongue and cheek musculature on the morphogenesis of the nasomaxillary complex. This research work was further revisited and it was concluded that these hypothetical concepts are still debatable (Moss, 1997). Some researchers hypothesise that there is abnormal muscular activity in the upper lip during function when analysed electromyographically in patients with a repaired cleft lip (Genaro et al., 1994).

The disturbance in lip musculature in the UCLP group could affect the normal growth of the maxillary complex, thereby producing greater relative maxillary arch constriction in this group when compared to patients with CP. Although the reduction of maxillary width is associated with a disturbance in the palatal sutural system in patients with cleft palate (Smahel and Brejcha, 1983), the combination of scarring caused by surgical repair of both the cleft lip and cleft palate results in significant growth restraint and maxillary hypoplasia in UCLP (Mars et al., 1987). Nyström and Ranta (1989) found that

3 year old patients with treated UCLP had a smaller maxilla and a normally sized mandible and patients with treated CP had a smaller maxilla and mandible (by a similar magnitude) when both were compared with children without clefts. These results support the findings from the present study that the relative maxillary arch constriction was greater in patients with UCLP compared to patients with CP.

In consequence, surgical intervention restricts normal facial growth to a greater extent in patients with UCLP than in patients with isolated CP. It is interesting to note that despite the cleft size and morphology of the maxilla in infants with UCLP being different to that in infants with CP, the primary cleft repair and subsequent treatment protocols for both cleft sub-phenotypes are similar resulting in different treatment outcomes (Reiser et al., 2013). The findings of the present study will contribute to raising awareness of gaps within the literature pertaining to relative maxillary arch constriction in patients with treated UCLP and CP.

6.3 Reproducibility and reliability of indices using plaster and digital models

Three observers (GM, NQ, and PM) scored the plaster and digital models using the 5 year old/GOSLON and MHB indices on two separate occasions. The following two statistical terms were selected to test the hypothesis, i.e. to assess whether the indices used are reproducible and reliable when the similar conditions are repeated at the second round of the scoring process. The term “Reproducibility” assessed intra-observer agreement between the two rounds of scoring taken at two separate occasions and “Reliability” assessed inter-observer agreement. The second round of scoring was undertaken after a three week interval to minimise recall bias. The Weighted Kappa (K) statistic was used to estimate the reproducibility/reliability for the 5-year old/GOSLON indices using both plaster and digital models as both indices are categorical in nature. The Kendall’s correlation coefficient was used to determine the levels of agreement between the observers using the MHB index on both plaster and digital models as the MHB is a continuous ordinal index. The interpretation of the Weighed Kappa statistic is given in section 4.15. The interpretation of Kendall’s correlation coefficient varies with values ranging from -1 to +1, where the value of -1 means negative correlation and +1 means a strong positive association between the data sets. It has been suggested that values greater than 0.70 suggest strong

association (Smeeton, 2005) whereas others believe that values closer to +1 represents a strong correlation.

The intra-observer reproducibility for the three examiners was higher for plaster models when compared to digital models for all the indices as shown by weighted Kappa and Kendall's coefficient values Tables 12 and 13. However, the MHB index displayed a higher level of reproducibility on digital models compared to the 5 year old / GOSLON indices.

The level of inter-observer reliability was higher using plaster models when compared to the digital models for all the indices as shown by Kappa and Kendall's values Tables 14 and 15. The data for MHB scores obtained using digital models were more accurate when compared to the 5 year old/GOSLON scores determined using the same models. Moreover, the inclusion of examiners with either extensive experience or no previous experience of scoring models with clefts offered an opportunity to test the effect of inexperience on the reproducibility and reliability of the indices. Interestingly, the first time user achieved very good agreement for the 5 year old/GOSLON indices when using plaster models and good agreement for the MHB index using digital models confirming that the MHB index is more 'user friendly' for digital models.

Patel (2011) investigated the use of the MHB index in determining the surgical outcome for patients with UCLP and concluded that the MHB index is more user-friendly than the Eurocran yardstick. The present study provides some clues about the 'learning effect' with these indices as demonstrated by improved reproducibility and reliability with increasing level of operator experience. The learning effect has been explained in the study involving validation of the MHB index for clinical use (Dobbyn, 2009). Calibration is necessary before using 5 year old/GOSLON indices. In this study all three examiners were calibrated in use of the 5 year old/GOSLON indices and no calibration was required for using MHB index. The intra-observer reproducibility was higher for the 5 year old/GOSLON indices than the MHB index using plaster models. However, when the same examiners scored the digital counterparts, the intra-observer reproducibility decreased from a higher to good level for the 5 year old/GOSLON indices and from good to moderate level for the MHB index.

The inter-observer reliability was higher for the MHB index when compared to the 5 year old/GOSLON indices when both were applied to plaster models. Similarly, the reliability was good for the MHB index when compared to the 5 year old/GOSLON indices when used on digital models, thus rejecting the third hypothesis.

In the present study reproducibility and reliability improved with examiner experience. This is evident from the scores for the inexperienced examiner (NQ), who scored the digital models more accurately when using the MHB index than the 5 year/GOSLON indices. This suggests that the MHB index is more user friendly for digital models. The assessment of reproducibility and reliability of the indices on digital models was a continuation of the previous work related to validation of the use of digital models against the gold standard i.e. plaster models (Brief et al., 2006, Gracco et al., 2007, Asquith and McIntyre, 2012, Chawla et al., 2012, Nicholls et al., 2013).

6.3.1 Scoring indices

The scoring indices (5 year old/GOSLON and MHB) have been validated on plaster models extensively (Mars et al., 1987, Atack et al., 1997a, Mossey et al., 2003, Dobbyn et al., 2011, Patel, 2011, Tohill and Mossey, 2007) and there are only few studies that having validated the use of these scoring systems on digital models (Asquith and McIntyre, 2012, Chawla et al., 2012, Nicholls et al., 2013). The aim of the present study was to further explore the use of digital models in the assessment of relative maxillary arch constriction using the above indices.

The indices were selected in line with the research work that has validated the use of these indices (Mars et al., 1987, Atack et al., 1997a, Mossey et al., 2003). The MHB index provides an objective and reliable method of the assessment of relative maxillary arch constriction (Gray and Mossey, 2005). The GOSLON index has also been endorsed for use in all ages including 5 year olds (Mars et al., 2006). Altalibi et al. (2013) performed a systematic review of the studies involving the indices used for the measurement of surgical outcomes in patients with CLP and found that the current evidence showed the MHB index performed very well compared to the other indices according to WHO criteria. It was further stated that the GOSLON index was the most commonly used index in previous research work. It was concluded that the MHB index

could be considered as a standard measure for surgical outcome for all cleft types, at any age.

The reliability of the trendlines shown in Figures 42, 43 and 45, 46 is determined by the R squared values depicted in Tables 16 and 17. The higher value suggests a normality of the distribution of data. The R squared value is higher for MHB scores followed by the 5 year old/GOSLON scores for the plaster models. Likewise, the R squared value for the trendlines for MHB scores was higher than 5 year old /GOSLON scores for the digital models.

6.3.2 Plaster versus digital models

The inter-canine distance (I/C) measurement is considered to be an essential variable for evaluation of treatment protocols in patients with CLP. It has been shown that linear distance measurements (I/C) obtained from digital models in a single observer study involving a group of infants with CLP differed to those from a non-cleft group (Mello et al., 2013). Taking on board this finding, a short single intra-observer (NQ) study involving linear distance measurements (I/C and I/M) was carried out using the maxillary plaster and digital models of the same patients with UCLP and CP. The results of this study in Table 18 indicated that there were no statistically significant differences between the measurements made on either plaster or digital models of patients with UCLP and isolated CP ($p>0.05$). Furthermore, the datasets for plaster and digital models had similar mean values. These findings are in line with a study conducted by Abizadeh et al. (2012) on plaster models and their digital counterparts belonging to patients with different types of malocclusions, where linear measurements and occlusal relationships recorded on both plaster and digital models were compared. The study concluded that digital models could be an adjunct to clinically assess the occlusion but as yet cannot overtake the current methods available for scientific purposes. The systematic review performed by Luu et al. (2012) assessed the reliability of linear measurements by comparing the plaster models and virtual models, finding virtual models are clinically acceptable indicated by higher intra-observer reliability and linear measurement validity. To avoid any systematic and random errors, it has been suggested that the landmark (point) identification should be done before any measurements are recorded on both plaster and digital models

(Asquith et al., 2007). The same mode of landmark identification was utilised in the present study to minimise the chance of errors. The marks were placed on the mesio-palatal cusp tips of first maxillary molars on plaster and digital models by a pen and MeshLab software respectively as illustrated in Figure 28, 29 and 30.

Table 19 shows the previous studies carried out regarding the role of digital models in measuring surgical outcome in patients with CLP.

Author	Method	Tests	Results
Brief et al., 2006	Micromasure 70 three-dimensional laser scanner (Micromasure, Bischoffen, Germany) (n=40) Forty plaster models of newborns up to 8 months of age 9 landmarks 4 observers	Intra-observer Error Study Inter-observer Error Study	Intra-observer The landmark placement error ranged from 0.31 to 1.33 mm Inter-observer error for landmark placement ranged from 0.61 to 1.99 mm
Oosterkamp et al., 2006	LDI-scanner Viscam RP version 2.1 software plaster cast models of BCLP patients were scanned n=10 2 observers	variance components intraclass correlations (ICC)	ICCs (0.81 to 0.96) Acceptable to good
Asquith and McIntyre 2012	R250 Orthodontic Study Model Scanner n=30 sets of study models of 5-year-old patients with UCLP were scanned 2 observers	Intra-observer and inter-observer reproducibility Friedman test	Intra-observer and inter-observer reproducibility were good (0.62 to 0.83 and 0.64 to 0.78, respectively)

Chawla 2012	5 year index tested on four reference model formats including digital models n=45 sets of plaster models of patients with UCLP examined by seven examiners at two rounds of scoring, 3 weeks apart.	Intra-observer reliability was determined by using Weighted Kappa statistic	Reliability of 5 year old index using all the formats was good to very good. The 3D digital model format of 5 year index was a reliable alternative to plaster models of 5 year old's index												
Mello et al., 2013	3Shape's R700™ Scanner 3D Software OrthoAnalyzer™ n=94 children aged from 3 to 9 months 1 observer	Measurement of intercanine distance	<table border="1"> <thead> <tr> <th>Control (n=19)</th> <th>Mean inter-canine (mm)</th> <th>SD</th> </tr> </thead> <tbody> <tr> <td>UCLP (n=50)</td> <td>27.52</td> <td>2.07</td> </tr> <tr> <td>BCLP (n=25)</td> <td>36.50</td> <td>3.66</td> </tr> <tr> <td></td> <td>34.83</td> <td>3.69</td> </tr> </tbody> </table>	Control (n=19)	Mean inter-canine (mm)	SD	UCLP (n=50)	27.52	2.07	BCLP (n=25)	36.50	3.66		34.83	3.69
Control (n=19)	Mean inter-canine (mm)	SD													
UCLP (n=50)	27.52	2.07													
BCLP (n=25)	36.50	3.66													
	34.83	3.69													
Nicholls et al. 2013	3M Unitek Lava™ system N=30 consecutive UCLP patients 2 observers	Linear Weighted Kappa statistic and Kendall's Coefficient of Concordance statistic	Intra-rater repeatability of digital study models (0.89 and 0.97). Intra-rater repeatability of study model casts scores (0.86 and 0.97). Inter-rater digital study model scores (0.80 and 0.87) inter-rater study model casts scores (0.80 and 0.90). KCC statistic (0.99) and Correlation Coefficient (0.86) very high score, good agreement												

Table 19: Previous studies carried out regarding the role of digital models in measuring surgical outcome in patients with CLP

6.4 Strengths and limitations of the methodology

The advantage of this pilot study was to address any flaws in the research methodology before embarking on a larger scale study. The examiners participating in this study have the opportunity to provide feedback on the viability of undertaking a large scale project. The future researcher can assess the limitations of this study and modify and address them accordingly. A large scale project will have financial implications which will have to be addressed before embarking on the project.

For the present study the 3D laser scanner was used to produce digital models to measure the treatment outcome in patients with UCLP and isolated CP. Recently, various 3D scanners have been introduced as a tool for the analysis of the dimensions of digital models of the maxillary arch either individually or in occlusion for patients with CLP. The NextEngine desktop laser scanner was chosen for the present study as it has not been used on dental plaster models of patients with CLP so far but the literature review identified some studies, where this scanner has been used directly on patients' face (Ciocca et al., 2010). This scanner has not been specifically made for dental use however, it was chosen for its economic viability and cost effectiveness. There are scanners available which have a high degree of resolution as compared to the scanner used for this project, but they are not universally available in less developed areas of the world. The digital models in occlusion produced using the NextEngine scanner had only one intermaxillary relationship compared to conventional plaster models, where the occlusal relationship could change (varies from examiner to examiner). There is definite scope for improvement in the process of digitisation in the future, so that same standard is achieved as the gold standard of the conventional plaster models. The accuracy of the digitally recorded intermaxillary relationship was assumed to be similar to that of the plaster models as they were secured into a correct occlusion by the transparent adhesive tape to prevent them sliding from the scanner turntable during the scanning process. Dunbar et al. (2014) attempted to scan the models with the NextEngine scanner, however they abandoned the use of this scanner as they concluded that an articulating arm was required to enable full, high quality imaging of the study models. However, this problem was overcome in the current

study by temporarily occluding the U/L plaster models together using transparent adhesive tape.

6.5 Advantages of the NextEngine scanner

The scanner used in this study was commercially available. It is a portable desktop scanner, and as such, plaster models or dental impressions do not need to be transported away for scanning. It is relatively cheap compared to other scanners available in the market. Furthermore, the ScanStudio software is very easy to use for scanning individual plaster models. It is non-destructive and reasonably fast and unlocks the full potential of the 3D scanner. This scanner does not need any dark room or special background for functioning.

Polo and Felicísimo (2012) shed some light on the accuracy of NextEngine scanner in a study titled "Analysis of uncertainty and repeatability of a low-cost 3D Laser Scanner". They concluded that, reliability of scans was better with the scanner in macro- mode than in wide-mode and the scanner did not perform as per the specifications given by the manufacturer. Some changes were suggested by the authors, such as adding a reference scale externally to the scanner and providing a self-calibrated object for permitting calibration operation by scanner. For the present study the macro-mode was selected for the scanning procedure. The NextEngine scanner has been used in maxillo-facial rehabilitation of patients with facial cancers and certain craniofacial syndromes specifically Treacher Collins Syndrome (Ciocca et al., 2010, Ciocca et al., 2009). They used Laser scanning, Reverse Engineering, CAD, CAM and Rapid Prototyping Technology for assisting in the fabrication of the maxillofacial prosthesis. The complete workflow involved direct scanning of the affected part of face by the NextEngine Laser scanner, digitising the patient's facial model making use of virtual models from 'Ear and nose digital library'. The final physical moulds for processing silicone, the substructure for retaining the prostheses were fabricated by making use of Rapid Prototyping Technology. The studies confirmed the direct use of this scanner on the patient's face, thus validating the direct use of this Laser scanner on the face. The morphology of the palate has been widely evaluated in patients with CLP to correct the speech impairment because palate has a central role in speech formation. At present palatal modelling techniques are still in their infancy. Yunusova et al. (2012)

utilised a novel thin plate spline tracing technique to reconstruct the palatal surface and to validate it against the scanned maxillary models. The plaster models of subjects were scanned using the NextEngine Laser scanner. They concluded that there was a slight error of fit between the traced and scanned models. It was found out in the present study that the scanned maxillary models were very close to their plaster counterparts with regards to the linear measurements.

The NextEngine ScanStudio software links with the online support centre called NextWiki Support Centre. The online expert advice is offered through this service for smooth working of the scanner with the software. The MeshLab software package integrated well with the NextEngine ScanStudio software. This was only possible because ScanStudio allows STL file conversion. These files not only helped with the calculation of linear measurements on the digital models used in this study, but can also work well within the Orthodontic laboratory, chair side use and milling process and also the exchange of clinical information for research.

Furthermore, the ability of the ScanStudio (NextEngine) software to convert NextEngine files into STL files and Rapidworks could be of use in Rapid prototyping for 3D printing. By utilising the technology of 3D printing, physical models can be fabricated from inexpensive materials which could be discarded later, thus eliminating the problems associated with the storage of plaster models for medicolegal reasons.

The 3D Laser scanning process encompasses all aspects of managing patients with craniofacial anomalies starting from the documentation, analysis and finally evaluating treatment outcomes (Da Silveira et al., 2003).

6.6 Disadvantages of the NextEngine scanner

If the surface of the plaster model has a high level of reflectivity, the accuracy of the digital model is affected adversely. To overcome this problem, the manufacturer recommends use of talc, hairspray or white paint. These tools can help in capturing data but they could damage the surface of plaster models, which are an important part of patient records.

The accuracy of the NextEngine scanner is 127micron (NextEngine, 2014) compared to 20 micron accuracy with the R700 Orthodontic study model scanner from (Great-

Lakes-Orthodontics, 3 Shape 700). A pilot comparative study involving the NextEngine scanner and the R700 Orthodontic study model scanner was undertaken, it was found that digital models produced using the 3Shape R700 Orthodontic scanner were more accurate than the digital models from the NextEngine scanner (Cochrane, 2014). The poster for this study is attached in appendix G. The digital models produced by the NextEngine scanner were compared to their plaster counterparts (gold standard) in the same study; the MHB scores were less consistent on digital models when compared to MHB scores on the corresponding plaster models as shown in Figure 50. The difference in scores could be due to the inherent difficulty of scoring MHB on the digital models, suggesting that the occluded digital models produced using the NextEngine scanner are not suitable for measuring surgical outcome as assessed by the MHB index. The 3-shape R700 Laser scanner was compared to the gold standard (SLP 250 Laser probe by Laser Design, Detroit, Michigan) in another comparative study, where the 3Shape R700 scanner was found to be sufficiently accurate to undertake any 3 dimensional scanning procedure (Hayashi et al., 2013). Both are easy to use and do not need a further training of staff to use either type of scanner. In a study where the 3M Unitek Lava TM scanner was used for the digitisation of plaster models, the reliability of the digital models was assessed using GOSLON index, with the resulting scores displaying a high degree of reproducibility and repeatability (Nicholls et al., 2013). In the present study, despite using a scanner with lower resolution, the results were almost identical to a study undertaken by Asquith and McIntyre (2012), where R250 Orthodontic Study Model Scanner (3Shape A/S, Copenhagen, Denmark) was used. In the present study, the examiners displayed a substantial level of reproducibility using the MHB index on digital models compared to the 5 year old/GOSLON indices Tables 12 and 13. This finding has the potential to be further investigated in the future so as to ensure the MHB index is more valid on digital models. In the present study, the examiners displayed a substantial level of inter-observer reliability when using the MHB index on plaster and digital models when compared to the 5 year old/GOSLON indices Tables 14 and 15. The lower levels of reproducibility associated with scores using digital models could be attributed to many reasons; the lower accuracy of the NextEngine scanner compared to other higher-accuracy scanners, only one intermaxillary relationship being possible with digital

models in occlusion and lastly the issue of assessing a 2D image of a 3D model. The use of adhesive tape might have interfered with the functioning of the laser beam. Most observers have found that when using digital models, they need to rotate the digital image several times to assess the occlusal relationship and to identify any relevant landmarks. By using digital models, planes and angles are more difficult to assess than using plaster models making the digital scores potentially less reliable than scores produced using plaster models (Wiranto et al., 2013). Digital 'holes' were present in the maxillary casts shown in Figure 26, suggesting that the laser did not scan the undercuts to their full depth.

6.7 Ideal study design / alternate strategy to answer the research questions

In an ideal situation, the study could have been done prospectively, but it would have taken considerably longer time to complete. This study does not shed light on the effect of primary surgery alone on the maxillary arch. Including the plaster models of patients with UCLP and CP at birth could have addressed this question. What happens between the ages of 15 years and adulthood could have been addressed by including models of patients at age 20 years of age because growth does not necessarily stop at 15 years of age. These models were not included in the study as very few models of these age groups were available in the CLEFTSiS archive.

The more ideal design would begin with a power calculation and evaluation of the effect size. A retrospective longitudinal study involving plaster study models from birth to 20 years with access to the patients treatment records should be undertaken. The use of MHB index for calculation of relative maxillary arch constriction for evaluation of more subtle changes in occlusion would be ideal.

Photographs, radiographs and cephalometric records could have been included in the study, however these have been studied extensively in other studies. A high resolution scanner could have been used, unfortunately due to the budgetary limitations this was not possible.

3D printing to compare the printed physical models with the original plaster models could also have been performed. Unfortunately this was not possible due to time

constraints also increase in sample size was precluded by long scan time and budgetary limitations however, this could be a proposal for a future study.

6.8 Contribution of the study to clinical practice and audit

The study highlights the differences in the relative maxillary arch constriction within different types of clefts. This finding if proved to be relevant on a larger sample of subjects with CLP would enable surgical protocols to be tailored according to type of cleft.

The study highlights the comparable performance of the 5 year old/GOSLON and MHB indices on digital models as opposed to plaster models. More research work could be built on existing knowledge so that the indices could be used more quickly and precisely on digital models. It was observed in the present study that relative maxillary arch constriction did not deteriorate progressively with growth for the whole sample. It was further suggested that there is no need to wait till the end of the facial growth to determine the outcome of surgery, the cleft care outcomes should aim to determine the success or failure at or around the age of 10 years. Furthermore, as there was more relative maxillary arch constriction for the UCLP group, perhaps Orthodontists should aim to treat this adequately during preparation for alveolar bone grafting. The present study also stated that the occluded digital models produced by the NextEngine scanner were inferior to occluded plaster models in determining the treatment outcome and as such are not suitable for archiving clinical records.

6.9 Inter-centre audit

The treatment outcome measurements could be shared with other international experts for clinical research and audit. This could allow larger centres to act as a baseline for smaller centres to compare their work. After a CSAG review (1998) and the subsequent re-organisation of the cleft services in UK, tricentre and quadcentre audits were setup which involved three or four of the nine designated UK cleft centres. The five point scoring system in the form of a questionnaire was selected for auditing the severity and treatment outcome of patients with repaired UCLP. This method was considered to be very subjective as most of the methods used for audit are predominantly subjective (Kim et al., 2011). The need for a more objective three-

dimensional tool which is internationally agreed was suggested previously by (Al-Omari et al., 2005). The present study focused on the performance of the MHB index (which is more objective) against the 5 year old/GOSLON indices (which are more subjective) on both plaster and digital models to fill the knowledge gap in this field of research. The success of an inter-centre audit is massively reliant on adequate record keeping. The archived plaster models of patients with CLP are considered as a permanent record and the digitisation of archived plaster models into an electronic record reduces the risk of the records being damaged or deteriorating over time. The electronic record keeping for patients with CLP should be in line with the 'gold standard'. The process of digitisation could revolutionise CLP management by this novel method of record keeping as the accumulated digital model archive would enable future research to be carried out anywhere in the world by file sharing.

The long term multidisciplinary approach to management of patients with CLP is well documented, providing such an approach by humanitarian organisations that provide surgical treatments across borders is a huge challenge. The repair of the palate must be followed by speech therapy otherwise lifelong implications of speech deficiencies are profound (Damiano et al., 2007). The use of modern telecommunication modalities has made it easier to render some services through telemedicine, so far speech therapy has been introduced successfully, parents and patients have a positive experience with this method of therapy (Whitehead et al., 2012).

In terms of a direct benefit to patients, the digitised models being a part of the electronic record could lead to increase in the ease of CLP management. The digitisation of plaster models would allow an increase in speed of consultation with cleft specialists (Wiranto et al., 2013). The post-operative needs for patients with CLP in developing countries are similar to those in the developed nations. Due to reduced accessibility or poor quality of cleft care in developing countries, non-government organisations such as Smile train and Transforming faces worldwide are concentrating on the provision of preliminary surgical procedures in developing countries like India (Mossey and Little, 2009). The cleft care provided by these humanitarian organisations should be expanded by providing pre-operative evaluation and post-operative care. This can be undertaken by making use of the innovative technology using wireless 3G

connections which are available worldwide. The present study showed that the digital models scanned by NextEngine were less reliable than the plaster models and as such, a scanner with a higher resolution should be employed for scanning digital models. (Furr et al., 2011). This could become possible with the increasing availability and decreased cost of high resolution scanners. Dunbar et al. (2014) carried out a pilot study to assess the use of teleorthodontics in comparison to conventional face to face consultation. The cone beam CT was used to scan plaster models to be used in teleorthodontics. They concluded that inter-observer reproducibility was superior using conventional methods for treatment planning and found that the patients were more satisfied with the conventional type of consultation.

6.10 Drawbacks of this study

The main shortcoming of the present study was the selection of the sample of plaster study models belonging to patients with surgically treated CP and UCLP. This was based on the availability of the complete set of 5, 10 and 15 year old patients with UCLP and CP. Within the CLEFTSiS MCN archive, there was a shortage of plaster models of 5 year olds, because some children at the age of 5 are reluctant to have impressions taken.

The smaller sample size was another drawback of the present study, which might have resulted in a fluctuation of the weighted kappa and Kendall's coefficient values. This study should be ideally repeated on a larger sample to produce a clearer evaluation of reproducibility and reliability. The subjects at age 10 and 15 may have undergone orthodontic treatment, that would have mitigated the effects of primary surgical intervention alone. Restorative procedures and alveolar bone grafting could also mask the effect of primary and any revision surgery. A longitudinal study on subjects who have undergone only primary surgical procedures could be investigated, the surgical outcome in such cases would not have been diluted by any subsequent orthodontic or surgical interventions.

The NextEngine scanner was not accurate enough to render a precise digital model. This was sufficiently good for scanning individual models rather than models in occlusion. The scanning process for occluded models was more complicated, involved

superimposing two scan families which were scanned at two different tilts in an endeavour to scan all the areas, avoiding any blind areas in the final scan, making this process more time consuming. Furthermore, the low resolution scan was made worse by the technical problems associated with keeping the set of plaster models in occlusion. The adhesive tape would have an impact on the functioning of Laser beam which might have lead to an error but this was not investigated in the present study. It is recommended to either check for any interference or avoid placing the tape in an area to be measured in future research projects.

6.11 Sources of bias and its effects on the results

The small sample size is a major cause of bias, as is the variation in the characteristics of the clefts at birth.

6.11.1 Selection bias: Only those cases were selected which had a complete set of plaster models in the CLEFTSiS archive. This method of sample selection could have an adverse effect on the resultant data that could distort the results.

The sample is not an exact representation of the UCLP and CP groups. It was a retrospective study so patients' previous records were essential for the study. However, selection bias would not impact on the results to a large extent because of the longitudinal nature of the study design.

6.11.2 Recall bias: The examiners can remember the findings from the previous knowledge in this field or the findings from the first round of scoring that could affect the reliability of the results.

6.12 Future research

Further research is required to study these cleft types along with other types including bilateral cleft lip and palate (BCLP), lip and soft palate clefts to determine any differences in the pattern of maxillary growth.

This study sheds some light on the performance of the MHB index on digital models however, further research is needed to expand current observations to validate the use of the MHB index on digital models. It will be only possible if the quality of digital models is identical to that of plaster models. This could in turn facilitate the validation

of the use of the MHB index using digital models in future. Using a scanner with greater accuracy and a larger sample size would be a priority.

Further research should also investigate the use of RapidWorks software for reverse engineering. The reliability of the physical models generated from reverse engineering can be compared to conventional plaster models which could in turn evaluate the accuracy of other scanners e.g. R 700 (3shape), CBCT and their associated software. Research regarding scanning and converting mesh model data into editable scanned models for CAD programmes such as SolidWorks, Creo and Inventor is needed. This technology could revolutionise the cleft care management.

If guidelines are in place regarding the required specifications of digital scanners for dental use, treatment outcome assessments could be agreed at a national level. This could in turn help the planning of more accurate treatment protocols for patients with CLP through research and audit.

Direct intra-oral scanning also offers an opportunity for the future for recording the occlusal relationships on 'direct' digital models, or alternatively applying the indices clinically rather than on plaster or digital models. Many of the 5 year olds with CLP do not turn up simply because of lack of cooperation for impression taking, so direct use of the MHB index on the patient at the chairside could be considered (Dobbyn et al., 2013, Dobbyn, 2009). A project is underway in Dundee involving the use of MHB and GOSLON indices on digital models produced from intra-oral scans.

It has been emphasised in an annual report by CLEFTSiS that up to 50% of 5 year records can be difficult to obtain for various reasons (CLEFTSiS, 2014). The problems encountered in a cohort of 5 year old patients were also highlighted in the CSAG report where 7% of the cohort failed to attend for impression taking. This report was based on four research studies collaborated with CSAG, encompassing reports and recommendation related to complete cleft management in the UK and comparing this with European standard (Sandy et al., 1998, Williams et al., 2001, Bearn et al., 2001).

All the digitally stored confidential information should be protected using encryption technology (coding of the information) to prevent it from being accessible to unauthorised persons and only accessible to authorised persons using the computers

and computer storage medium (e.g. USB drive, CD, DVD) containing confidential information. The encrypted files decrease the risk of confidential information from being intercepted in case of loss or theft of data (Leicester, 2014). As with any digitally stored information the increased security risk and the risk of information deletion must be considered. All information should be backed up and all systems should be secure.

As the results of this study showed that the relative maxillary arch constriction was greater at age 5 in both groups (UCLP and CP) there is a need for further research in this age group to determine the cause of this and potential methods of prevention through altering surgical protocols.

The cleft management centres should be able to show positive treatment outcomes. The quality improvement and audit of these services should be the main focus. Audits should be repeated periodically to ascertain whether any identified concerns are being addressed. The audit should also focus on whether the patients are registered with a General Dental Practitioner and keeping up with their oral hygiene practices because patients with CLP are at a risk of developing dental caries.

A future study should involve a larger cohort to fully understand the pattern of maxillary growth in patients with treated UCLP and other cleft phenotypes.

6.13 Digital recording

The present study evaluated the use of the MHB index on digital models and was found to be more versatile and sensitive as compared to 5 year old/GOSLON indices. There is a greater possibility of the MHB index being used in conjunction with the electronic patient record in future. This could allow meticulous documentation and the use of data for research and audit purposes. More research is needed to develop software to automatically generate MHB scores on digital models. The digital recording of scores should be employed in Orthodontics in general. Future work should also investigate the possibility of automatic scoring of outcomes for non-clefts (e.g. IOTN / PAR / Orthognathic surgery outcomes).

CHAPTER 7: CONCLUSIONS

The following conclusions were drawn for the null hypotheses tested in this study.

Null hypothesis 1. Relative maxillary arch constriction in patients with surgically repaired UCLP and CP does not deteriorate progressively with growth.

The null hypothesis was not rejected;

There was no progressive relative maxillary arch constriction found between 5, 10 and 15 years for children with CP or UCLP.

Null hypothesis 2. There is no difference in relative maxillary arch constriction in patients with surgically repaired UCLP and CP.

The null hypothesis was rejected;

Relative maxillary arch constriction was found to be greater in patients with UCLP compared to patients with isolated CP.

Null hypothesis 3. The reproducibility and reliability of the 5 year old/GOSLON and MHB indices are not different when assessed on plaster and digital models.

The null hypothesis was rejected;

Reproducibility and reliability of the 5 year old/GOSLON and MHB indices were generally higher for plaster models (good to very good) in comparison to digital models (moderated to good). Inter-observer agreement was superior for the MHB index in comparison to the 5 year old/GOSLON indices when using plaster models. Intra-observer and inter-observer agreement was more consistent for the MHB index in comparison to the 5 year old/GOSLON indices when using digital models.

Null hypothesis 4. There is no difference in linear measurements using plaster and digital models.

The null hypothesis was not rejected;

There was no statistically significant difference found between linear measurements for plaster and digital models.

CHAPTER 8: REFERENCES

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Appendix:

Laptop specifications:

Intel R core TM i5-3337U 1.8GHz with turbo boost up to 2.7 GHz

Intel HD Graphics 4000 up to 1792 MB Dynamic video memory

15.6 " HD LED LCD

4GB DDR3 Memory

500 GB HDD

DVD Super Multi DL drive, 4-cell Li -ion battery

CPU: Dual Core processor

Memory: 4GB RAM

GPU: 512MB

Operating System: Microsoft Windows 8 (part of the Windows NT family operating system).

USB 2.0 Powered Hub

DIGITAL CALLIPER

Resolution: 0.01 mm

Power: one 1.5 V button cell

Measuring speed: < 1.5m/s

It has the following advantages:

1. Zero setting at any position, easy to take relative measurements.
2. Metric/inch system interchange at any position.
3. With data output interface, data can be input to a special printer or a computer via a special cable for data processing and printing.
4. Special function: With data holding, fast display, fast tracing of maximum and minimum value during measurements, conversion between relative and absolute measurement and tolerance zone setting.

Plaster Study Model Scoring Sheet

Examiner:

Date of scoring:

Subject number:

Goslon Index: Score 1 2 3 4 5

Huddart & Bodenham Score:

Right Buccal Segment

6	E	D	C
-3	-3	-3	-3
-2	-2	-2	-2
-1	-1	-1	-1
0	0	0	0
+1	+1	+1	+1
+2	+2	+2	+2
+3	+3	+3	+3

Sum

Labial Segment

1	1
-3	-3
-2	-2
-1	-1
0	0
+1	+1

Sum

Left Buccal Segment

C	D	E	6
-3	-3	-3	-3
-2	-2	-2	-2
-1	-1	-1	-1
0	0	0	0
+1	+1	+1	+1
+2	+2	+2	+2
+3	+3	+3	+3

Sum

TOTAL SUM

D

Information Governance
Maryfield House South
Mains Loan
Dundee
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www.nhstayside.scot.nhs.uk



Nafeesa Qureshi
17 Donald Gardens
Dundee
DD2 2RZ

Date 29 January 2014
Your Ref
Our Ref Caldicott/CSAppNQ1010
Enquiries To Sender
Extension 71438
Direct 01382 740074
Email peter.mckenzie@nhs.net

Dear Nafeesa,

Caldicott Approval – Outcome of cleft lip and palate repair measured by maxillary arch constriction

Proposal Sponsor: Dr Grant McIntyre, Consultant Orthodontist, NHS Tayside
Data Users: Nafeesa Qureshi, MSc Student, University of Dundee
Professor Peter Mossey
Dr Grant McIntyre, Consultant Orthodontist, NHS Tayside

Caldicott approval is given for you to use patient identifiable data to access dental study models of selected patients registered in CLEFTSIS as described in your application and supporting information.

Thank you for your co-operation in providing us with the information requested by us in this process.
Please contact me should any queries arise from the application of this approval.

Peter McKenzie
Information Governance Manager

Copy to:
Professor Peter Mossey
Dr Grant McIntyre, Consultant Orthodontist, NHS Tayside

Headquarters
Ninewells Hospital and Medical School, Dundee DD1 9SY

Chairman, Mr Sandy Watson OBE DL
Chief Executive, Mr Gerry Marr

NHS Tayside Caldicott Approval Form



1. Proposal Title :

Outcome of cleft lip and palate repair measured by maxillary arch constriction.

2. Sponsor Details and Declaration

Name:	Grant McIntyre	Address:	Orthodontic Department
Position:	Consultant Orthodontist		2 Park Place
Organisation:	NHS Tayside		Dundee
Email:	grant.mcintyre@nhs.net		DD1 4HR
Telephone:	01382 635964		

Sponsor's Declaration ¹:

I declare that the named Data User is engaged in a reputable research/audit project and that the data requested can be entrusted to this person in the knowledge that they will discharge their obligations in regard to the confidentiality of the data.

Sponsor's Dated Signature

Grant

17 JANUARY 2014

3. Data User Details and Declaration

Name:	Nafeesa Qureshi	Address:	17 DONALD GARDENS
Position:	MSc STUDENT		DUNDEE
Organisation:	UNIVERSITY OF DUNDEE		DD2 2RZ
Email:	nyqureshi@dundee.ac.uk		
Telephone:	01382619405		07574871900

Data User's Declaration :

I declare that I understand and undertake to abide by the rules of confidentiality and security in the use of patient identifiable information received from NHS Tayside.

Data User's Dated Signature

Nafeesa

17/1/2014

4. Caldicott Approval

Approval is given for the release and use of patient identifiable information as specified in this application

[Signature]
Medical Director, NHS Tayside

Director of Public Health, NHS Tayside

¹ to be signed by an NHS Tayside Consultant if the applicant is not of that status or is not medically qualified.



Does relative maxillary arch constriction worsen with growth for patients with unilateral cleft lip and palate (UCLP)?

GURESHI N, MOSSEY PA, MCINTYRE GT* University of Dundee



INTRODUCTION:

1. Digital models are equivalent to plaster models and are frequently used in cleft care for the assessment of surgical outcomes.¹
2. Quantifying relative maxillary arch constriction using the modified Huddart/Bodenham scoring system is one method of assessing dental arch relationships.^{2,3}
3. Relative Maxillary prominence is known to deteriorate towards the end of skeletal growth but no study has evaluated longitudinal maxillary arch constriction in patients with unilateral cleft lip and palate (UCLP).⁴

OBJECTIVE: To evaluate relative maxillary arch constriction for patients with unilateral cleft lip and palate (UCLP) during growth.

SUBJECTS AND METHODS: Plaster study models of five patients with UCLP taken at ages 5, 10 and 15 were randomly selected from the CLEFTSIS (Cleft Service in Scotland) archive. The study models were scanned in occlusion using a Next Engine 3D Scanner HD laser scanner (www.nextEngine.com). The occluded models were scanned using two scan families and superimposed with a resulting accuracy of 0.127mm. The models were then scored using the modified Huddart/Bodenham (MHB) scoring system to evaluate relative maxillary arch constriction.^{2,3} The data were tested using Kendall's *W* coefficient of concordance to determine the level of agreement of relative maxillary arch constriction at each of the time points.



Figure 1: Plaster models



Figure 2: Next Engine laser scanner



Figure 3: Digital models



Figure 4: Modified Huddart/Bodenham scoring system

Modified Huddart/Bodenham scoring system used to score plaster and digital models

SUBJECT NO.	MHB SCORE 5 YRS	MHB SCORE 10 YRS	MHB SCORE 15 YRS
57	-15	-20	-19
297	-3	-4	-5
238	-10	-7	-9
130	-10	-11	-9
546	-6	-8	-8

Table 1: Modified Huddart/Bodenham scores

RESULTS: The mean MHB score at age 5 was -9.8, at age 10 was -9.8 and had worsened slightly by age 15 to -10.2. The Kendall's *W* coefficient value of 0.9321. As this was close to 1 there was good agreement across the data.

DISCUSSION:

1. The data were consistent across all three time points indicating that there was no substantial worsening of maxillary arch constriction during growth for patients with UCLP.
2. The increase in relative maxillary arch constriction from age 10 to age 15 may have been due to a change in the skeletal growth pattern, although this would have been mitigated by Orthodontic treatment to a degree.
3. This is a pilot study so further work is required to investigate maxillary growth in isolated cleft palate and other cleft sub-phenotypes to determine if our findings are specific to UCLP or are typical of all cleft sub-phenotypes.

CONCLUSION: There is no substantial worsening of maxillary arch constriction during growth for patients with UCLP.

References

1. Fleming PS, Kaban V, Jhal A. Orthodontic measurements on digital study models compared with plaster models: a systematic review. *Orthod Craniofac Res* 2011; 14:1-18
2. Kogut JA, McIntyre GT. Dental arch relationships on three dimensional digital study models and conventional plaster study models in patients with unilateral cleft lip and palate. *Cleft Palate Craniofac J* 2011; 24: 500-4
3. Gray D, Mossey PA. Evaluation of a modified Huddart/Bodenham scoring system for assessment of maxillary arch constriction in unilateral cleft lip and palate subjects. *Eu J Orthod* 2005; 27:507-11



Comparison of two digital model scanners for the evaluation of maxillary arch constriction in cleft lip and palate

By Heather Cochrane

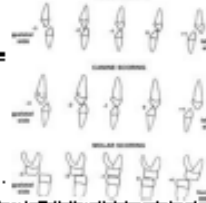
1. Introduction



Cleft lip clefts are the most prevalent craniofacial deformity occurring in 1 in 700 live births, although they range in severity as shown in Figure 1.



Clefts can cause problems with breathing, eating, speech, the dentition and aesthetics¹¹, seen in figure 2. In developed countries clefts are surgically repaired which results in maxillary arch constriction¹².



Scoring systems have been developed to grade the severity of this to aid in diagnosis and treatment planning. The Modified Huddry/Bodnar/M (MHB) Index is a continuous scale making it less subjective than others¹⁴. The scoring criteria is shown in figure 3.

2. Aims and Objectives

Traditionally MHB scoring has been done on plaster study models. However 3D scanning means it can be done using digital models, allowing for cost savings as no storage space is required, no risk of damage and immediate information exchange for consultation or referral¹⁵. There are a number of 3D scanners on the market however at present there are no guidelines in place stating the minimum necessary specifications for a 3D scanner intended for dental use. In this study two different 3D laser surface scanners were compared to determine the impact of difference in accuracy on the scanners ability to produce a digital model of sufficient standard to allow for correct diagnosis and treatment planning.

3. Methodology

20 subjects were selected from an existing archive, each have unilateral cleft lip and palate and having both maxillary canines and first permanent molars present.

The study casts were scanned on two 3D laser surface scanners of different accuracies, both in occlusion and each jaw in isolation. The scanners can be seen in figure 4.

NextEngine scanner
Accuracy 127 microns¹⁶



3Shape OrthoScan study model scanner
Accuracy 20 microns¹⁷



Figure 4: NextEngine scanner, higher degree of accuracy and 3Shape scanner

On the upper jaws the inter-canine width, inter-molar width and upper right and upper left first permanent molar to tooth widths were measured using digital calipers for the plaster casts and MeshLab software for the digital models as shown in figure 5. These measurements were then repeated a week later. Measurements on the digital models were compared to the plaster models (gold standard) using the analysis of variance (ANOVA) statistical test.

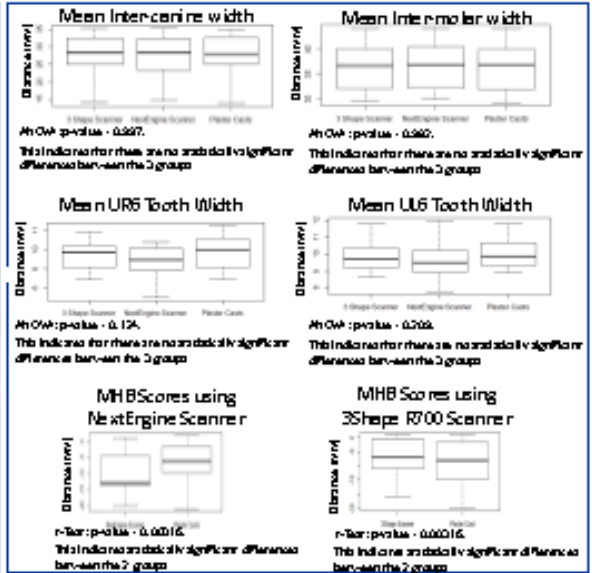


Figure 5: Digital models with measurements taken from the digital models



The digital models and the respective plaster study casts, shown in figure 6, were then scored using the MHB Index. Scoring on the digital models was compared to the plaster models using the t-test statistical test.

4. Results



5. Conclusions

Linear measurements. There were no statistically significant differences found between the two scanner types and the plaster study models but there were clinically significant differences in the digital models obtained from the less accurate NextEngine scanner only.

MHB scores. Statistically significant differences were found between both scanner types and the respective plaster study models. The linear measurements indicate that the digital models from the 3Shape scanner were accurate so this is likely to be due problems when scoring. Digital models must be rotated multiple times complicating the scoring process^{10d}. Previous studies have found digital models to be a suitable alternative to plaster casts^{11,12}, however the clinicians in these studies may have been more experienced in working with digital models.

Clinical implication. A 3D laser surface scanner accurate to 127 microns is not sufficient for dental use whereas one accurate to 20 microns is. However a higher level of experience in working with digital models may be required to allow accurate MHB scoring.

6. References

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17. Cleft lip and palate. In: *Textbook of Paediatric Dentistry*. 4th ed. London: Elsevier; 2012: 100-110.

7. Acknowledgements

I would like to thank Professor Mawaz and Dr. Malvina for their help and support throughout this project. I would also like to thank the local Quinichan for support in this project and the MHB scores for the digital models from the NextEngine scanner. Finally I would like to thank Dr. Chalmers for the use of the digital casts from the 3Shape R700 OrthoScan study model scanner and the MHB scores for these models.

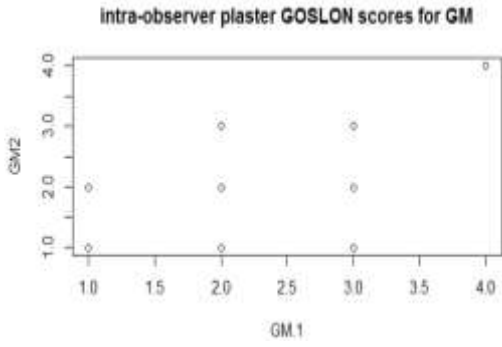


Figure 11: Scatter plots for the distribution of 5 year old and GOSLON scores by GM at two rounds of scoring GM1 and GM2 on plaster models.

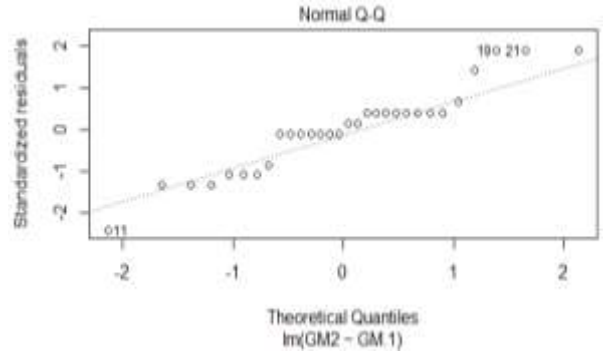


Figure 14: Q-Q plots for the distribution of 5 year old and GOSLON scores by GM on or around the linear line using digital models

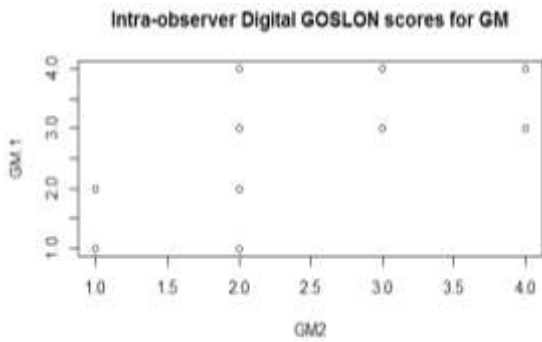


Figure 12: Scatter plots for the distribution of 5 year old and GOSLON scores by GM at two rounds of scoring GM1 and GM2 on digital models

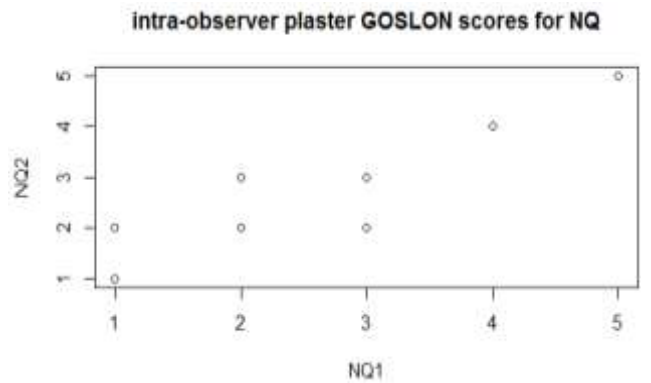


Figure 15: Scatter plot for the distribution of 5 year old and GOSLON scores by NQ at two rounds of scoring NQ1 and NQ2 on plaster models

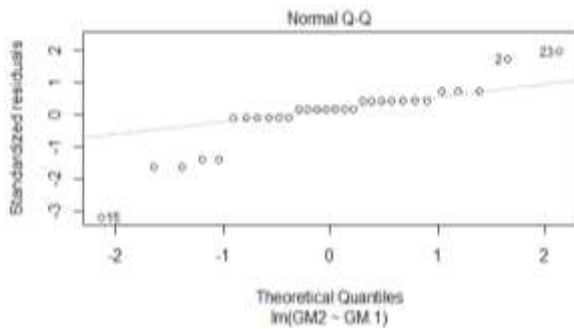


Figure 13 : Q-Q plots for the distribution of 5 year old and GOSLON scores by GM on or around the linear line using plaster models

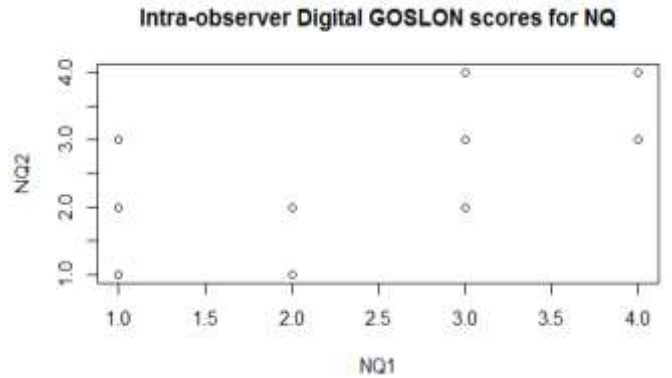


Figure 16: Scatter plot for the distribution of 5 year old and GOSLON scores by NQ at two rounds of scoring NQ1 and NQ2 on digital models

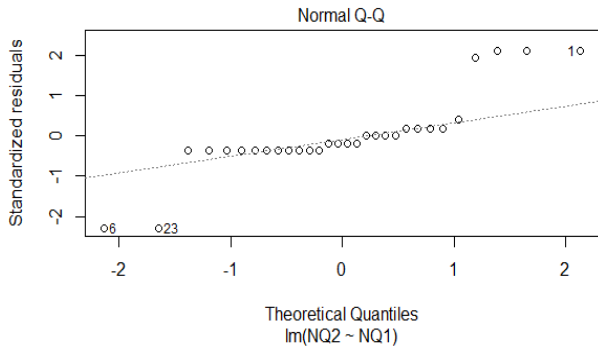


Figure 17: Q-Q plot for the distribution of 5 year old and GOSLON scores on or around the linear line by NQ for plaster models.

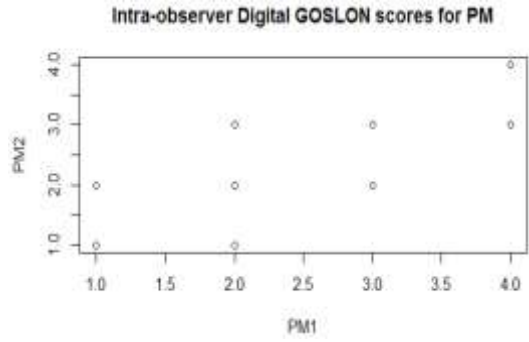


Figure 20 : Scatter plot for the distribution of 5 year old and GOSLON scores by PM at two rounds of scoring PM1 and PM2 on digital models.

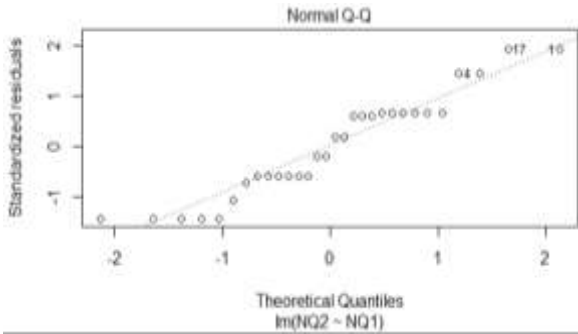


Figure 18: Q-Q plot for the distribution of 5 year old and GOSLON scores on or around the linear line by NQ for digital models

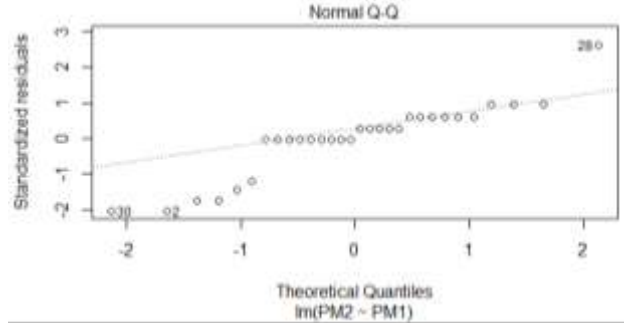


Figure 21 : Q-Q plot for the distribution of 5 year old and GOSLON scores on or around the linear line by PM for plaster models.

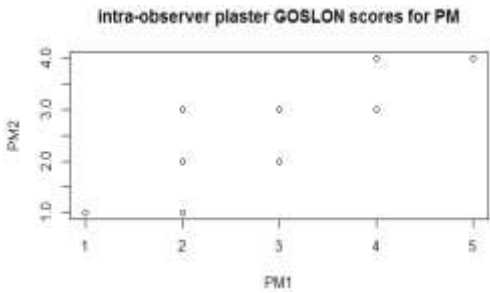


Figure 19 : Scatter plot for the distribution of 5 year old and GOSLON scores by PM at two rounds of scoring PM1 and PM2 on plaster models.

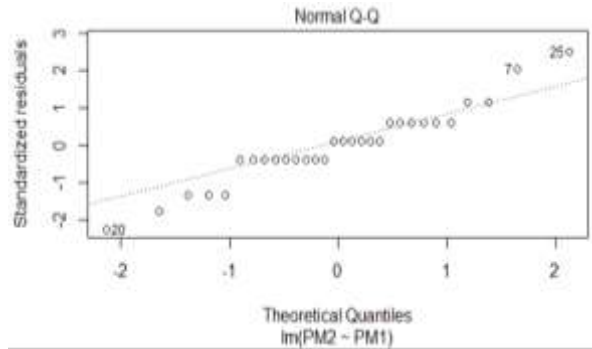


Figure 22: Q-Q plot for the distribution of 5 year old and GOSLON scores on or around the linear line by PM for digital models.

Intra-observer Repeatability for MHB scores

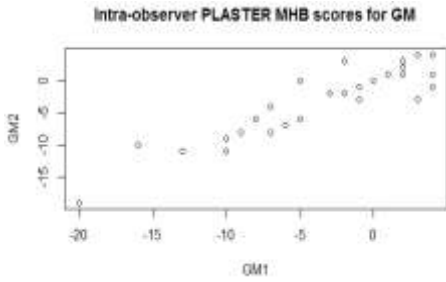


Figure 23 : Scatter plot for the distribution of MHB scores by GM at two rounds of scoring GM1 and GM2 on plaster models.

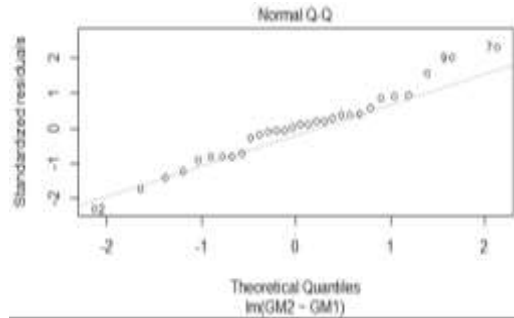


Figure 26: Q-Q plot for the distribution of MHB scores on or around the linear line (by GM) using digital models.

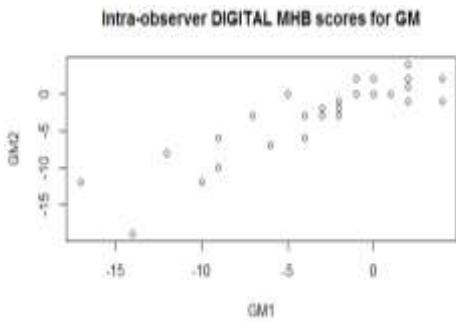


Figure 24: Scatter plot for the distribution of MHB scores by GM at two rounds of scoring GM1 and GM2 on digital models.

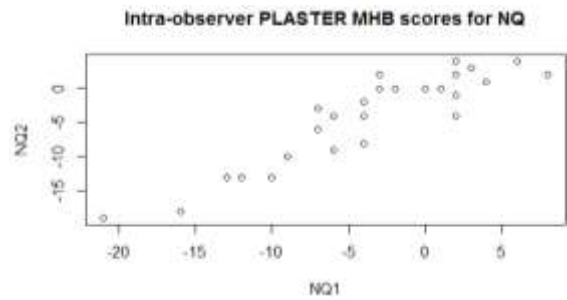


Figure 27 : Scatter plot for the distribution of MHB scores by NQ at two rounds of scoring NQ1 and NQ2 on plaster models.

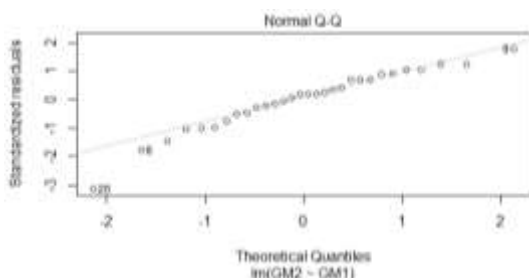


Figure 25: Q-Q plot for the distribution of MHB scores on or around the linear line (by GM) using plaster models.

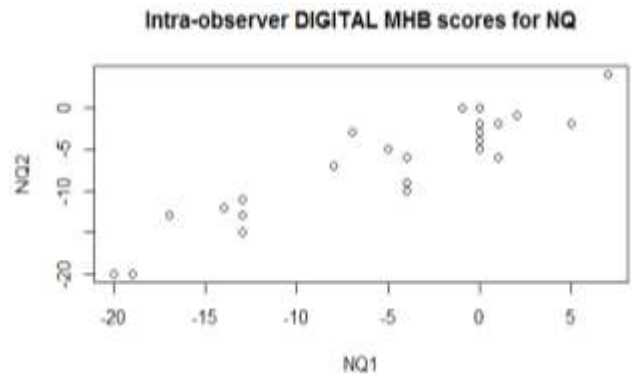


Figure 28: Scatter plot for the distribution of MHB scores by NQ at two rounds of scoring NQ1 and NQ2 on digital models.

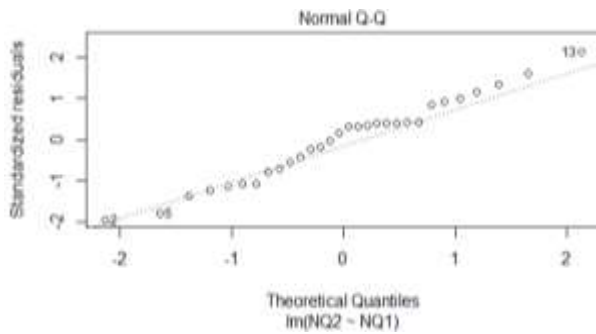


Figure 29 : Q-Q plot for the distribution of MHB scores on or around the linear line (by NQ) using plaster models.

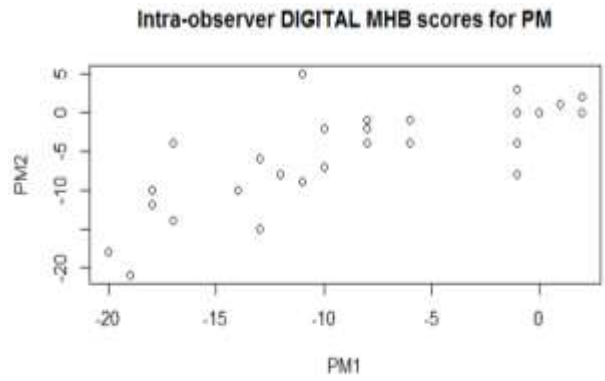


Figure 32 : Scatter plot for the distribution of MHB scores by PM at two rounds of scoring PM1 and PM2 on digital models.

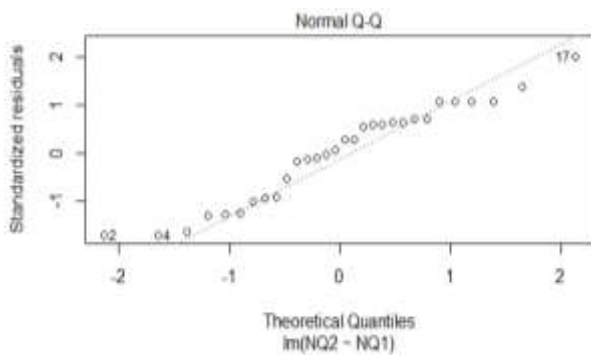


Figure 30: Q-Q plot for the distribution of MHB scores on or around the linear line (by NQ) using digital models.

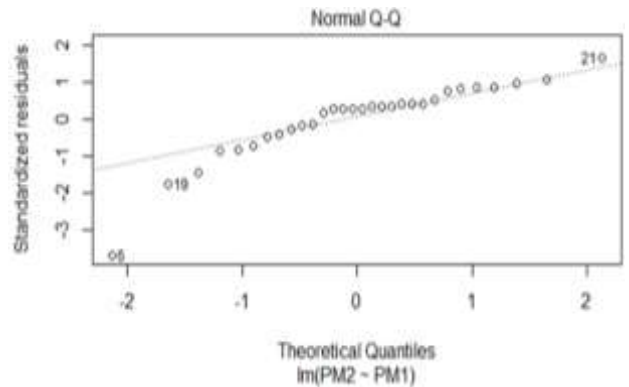


Figure 33: Q-Q plots for the distribution of MHB scores on or around the linear line (by PM) using plaster models.

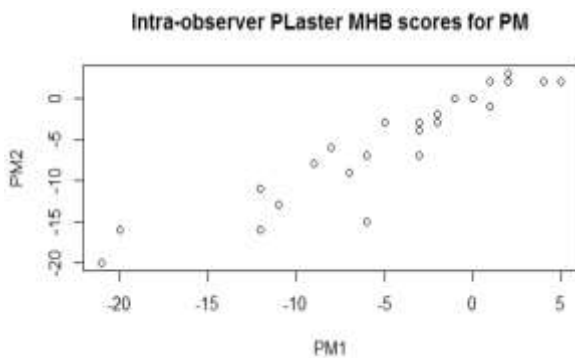


Figure 31: Scatter plot for the distribution of MHB scores by PM at two rounds of scoring PM1 and PM2 on plaster models.

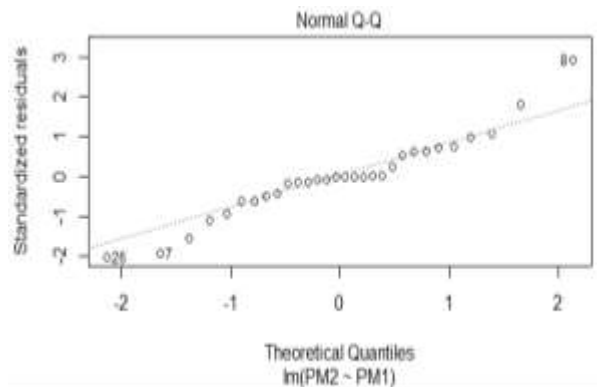


Figure 34: Q-Q plots for the distribution of MHB scores on or around the linear line (by PM) using digital models.

Inter-observer reliability
For the 5 year and GOSLON index:

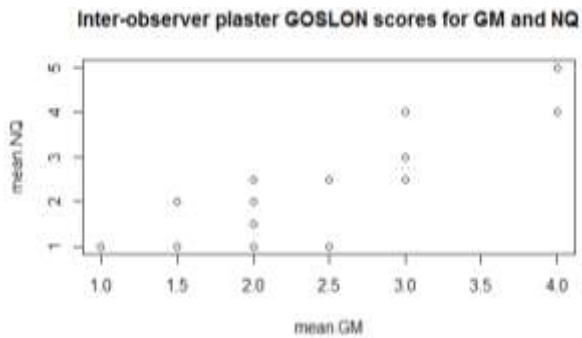


Figure 35: Scatter plot for the distribution of mean 5 year-old and GOSLON mean scores for GM and NQ using plaster models

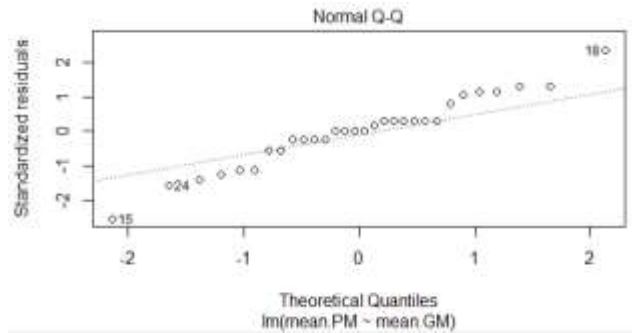


Figure 38 (bottom): Q-Q plot for the distribution of 5 year-old and GOSLON mean scores by GM and PM using plaster models.

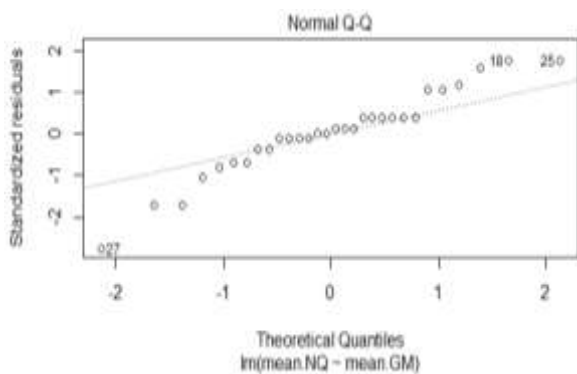


Figure 36 : Q-Q plot for the distribution of 5 year-old and GOSLON mean scores for NQ and GM using plaster models.

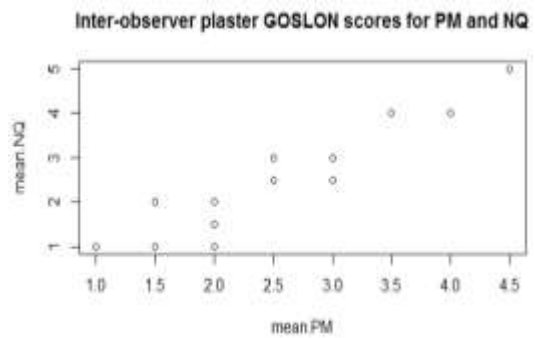


Figure 39: Scatter plot for the distribution of 5 year-old and GOSLON mean for PM and NQ using plaster models.

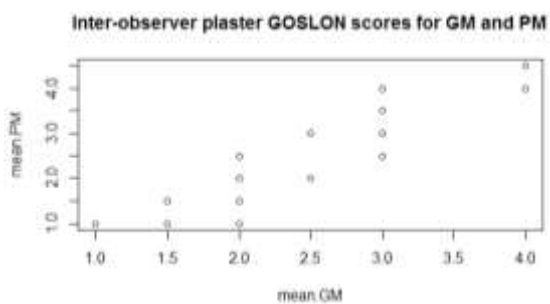


Figure 37 : Scatter plot for the distribution of 5 year-old and GOSLON mean scores for GM and PM using plaster models

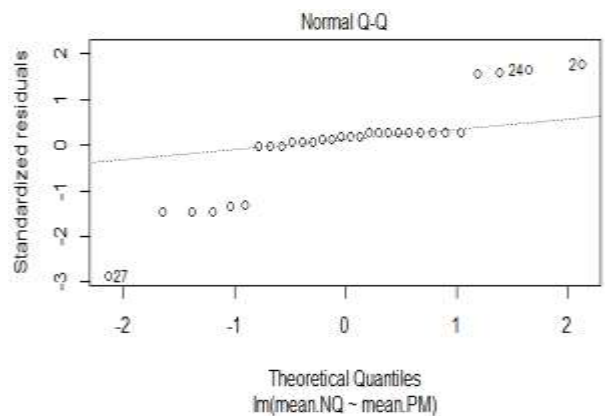


Figure 40: Q-Q plot for the distribution of 5 year-old and GOSLON mean scores for PM and NQ using plaster models.

Digital 5 year old and GOSLON indices

Inter-observer Digital GOSLON scores for GM and NQ

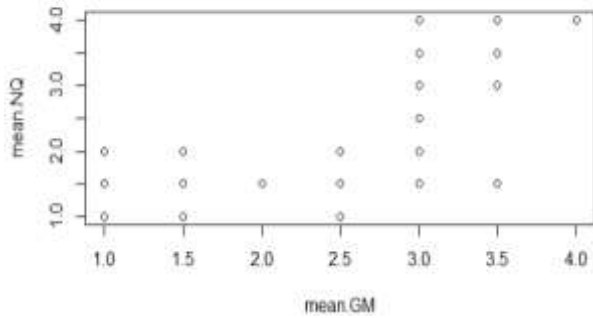


Figure 41: Scatter plot for the distribution of 5 year-old and GOSLON mean scores for GM and NQ using digital models.

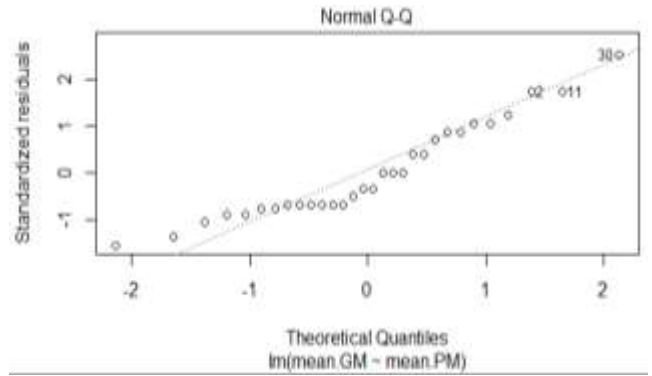


Figure 44: Q-Q plot for the distribution of 5 year-old and GOSLON mean scores for PM and GM using digital models

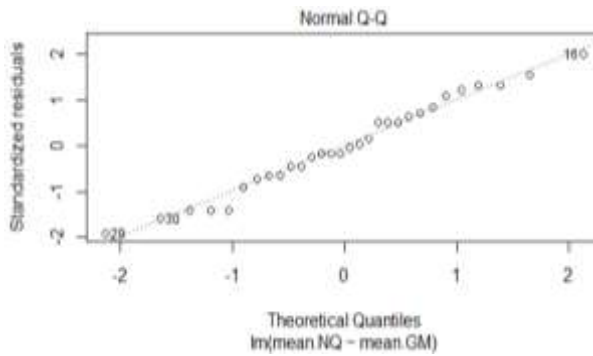


Figure 42: Q-Q plot for the distribution of 5 year-old and GOSLON mean scores for GM and NQ using digital models.

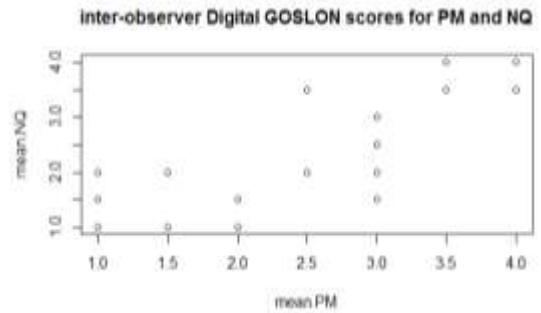


Figure 45: Scatter plot for the distribution of 5 year-old and GOSLON mean scores for PM and NQ using digital models

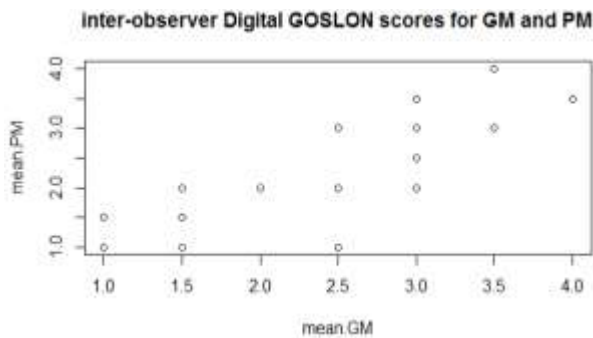


Figure 43: Scatter plot for the distribution of 5 year-old and GOSLON mean scores for PM and GM using digital models

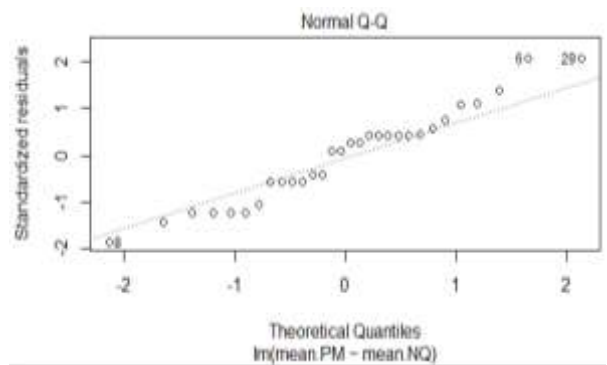


Figure 46: Q-Q plot for the distribution of 5 year-old and GOSLON mean scores for PM and NQ using digital models

Inter-observer reliability of MHB Score using plaster and digital models.

Inter-observer PLASTER MHB scores for GM and NQ

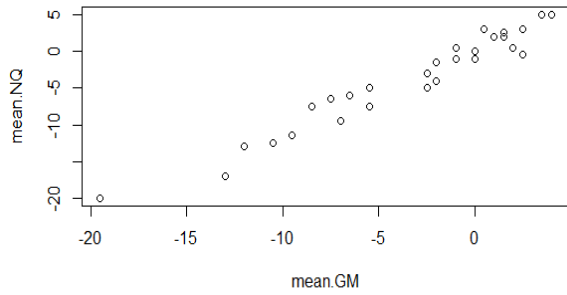


Figure 47: Scatter plot for the distribution of MHB mean scores for GM and NQ using plaster models.

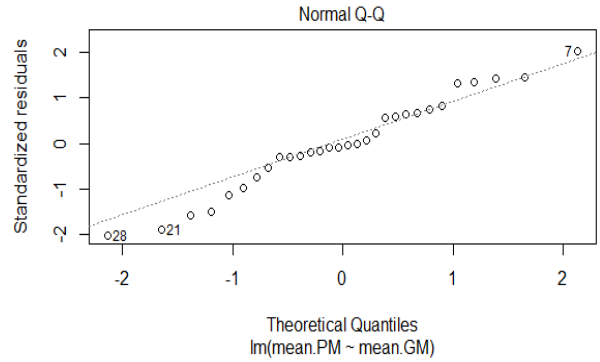


Figure 50: Q-Q plot for the distribution of MHB mean scores for PM and GM using plaster models.

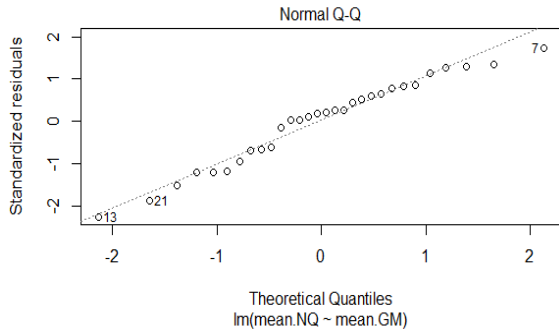


Figure 48: Q-Q plot for the distribution of MHB mean scores for GM and NQ using plaster models.

Inter-observer PLASTER MHB scores for NQ and PM

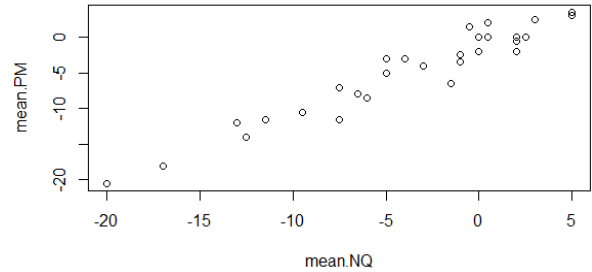


Figure 51: Scatter plot for the distribution of MHB mean scores for PM and NQ using plaster models.

Inter-observer PLASTER MHB scores for GM and PM

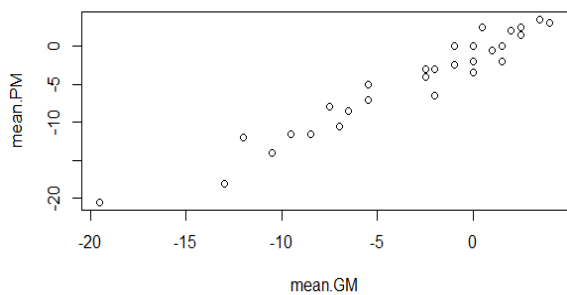


Figure 49: Scatter plot for the distribution of MHB mean scores for PM and GM using plaster models.

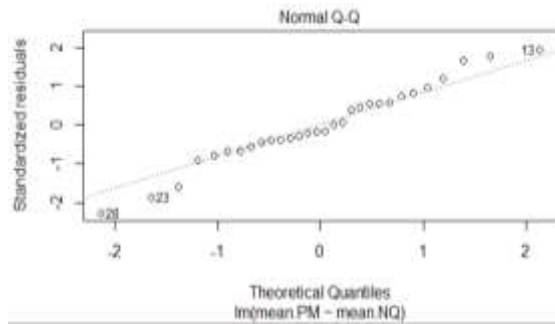


Figure 52: Q-Q plot for the distribution of MHB mean scores for PM and NQ using plaster models.

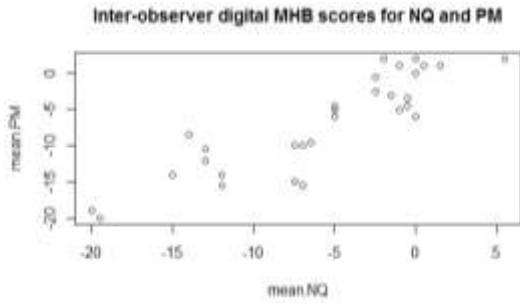


Figure 53: Scatter plot for the distribution of MHB mean scores for PM and NQ using digital models

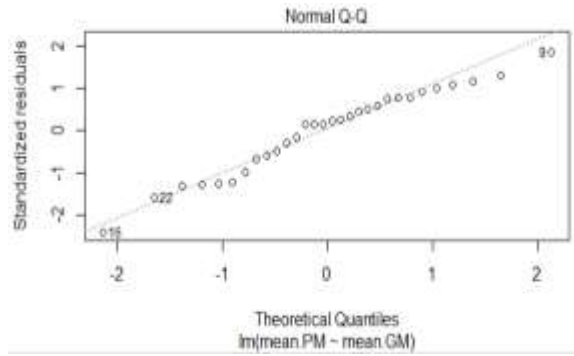


Figure 56: Q-Q plot for the distribution of MHB mean scores for PM and GM using digital models.

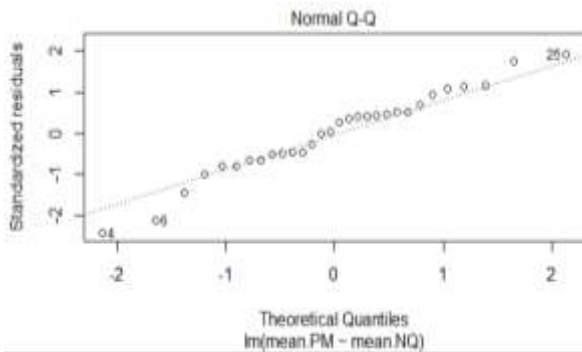


Figure 54 : Q-Q plot for the distribution of MHB mean scores for PM and NQ using digital models.

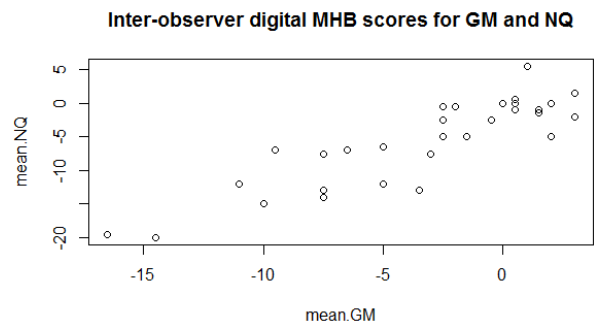


Figure 57: Scatter plot for the distribution of MHB mean scores for GM and NQ using digital models

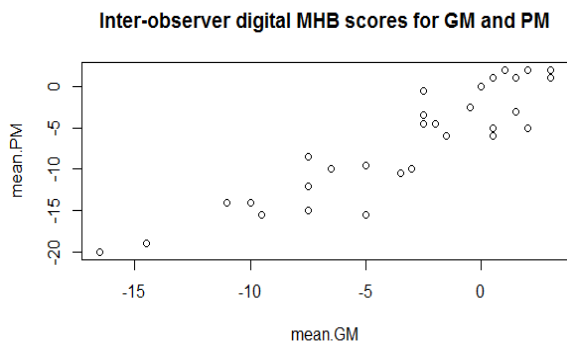


Figure 55 : Scatter plot for the distribution of MHB mean scores for PM and GM using digital models

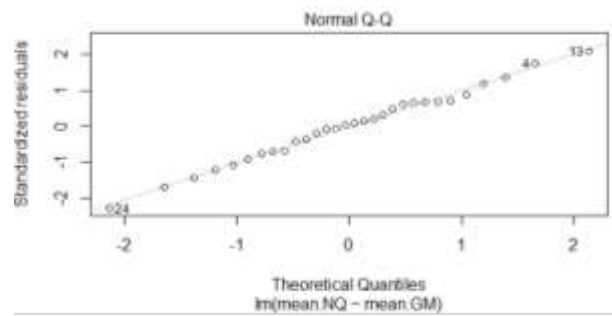


Figure 58 : Q-Q plot for the distribution of MHB mean scores for GM and NQ using digital models.

Data Request from Grant McIntyre
Data Extracted from Excelicare and NCAS – 18 September 2014

Cleft No: 67

No record on Excelicare or Clefthisis

Cleft No: 257

Record	Date	Available
5 yr photos	11/9/97	Y
5 yr study models	10/9/97	Y
5 yr ortho audit	-	N
Date of ABG	16/10/06	Y
Pre AGB Radiograph	13/10/04	Y
Post Op Radiology	-	N
Expansion Information	-	N
10 yr photos	-	N
10 yr study models	09/12/02	Y
10 yr x-ray	04/12/02	Y
10 yr ortho audit	GOLSON = 1 Comment: audit form not completed. Incomplete cleft of alveolus.	
15 yr photos	25/09/2007	Y
15 yr study models	25/09/2007	Y
15 yr x-ray		Y
15 yr ortho audit	-	-
20 yr photos	-	-
20 yr study models	-	-
20 yr x-ray	-	-

Cleft No: 289

Record	Date	Available
5 yr photos	-	N
5 yr study models	30/10/97	Y
5 yr ortho audit	30/10/97	5 Year Old Index Score = 4 No other data available
Date of ABG	09/08/01	Y
Pre AGB Radiograph	24/07/01	Y
Post Op Radiology	07/02/02	Y
Expansion Information	-	N
10 yr photos	08/08/02	Y
10 yr study models	21/11/02	Y
10 yr x-ray	21/11/02	Y
10 yr ortho audit	21/11/02	Golson = 2 No other data on form
15 yr photos	-	N
15 yr study models	14/01/08	Y
15 yr x-ray	14/01/08	Y
15 yr ortho audit	-	N
20 yr photos	6/11/12	Recorded on NCAS as taken
20 yr study models	6/11/12	Recorded on NCAS as taken
20 yr x-ray	6/11/12	Recorded on NCAS as taken

Cleft No: 436

Record	Date	Available
5 yr photos	-	N
5 yr study models	05/12/96	Y
5 yr ortho audit	05/12/96	5 Year Old Index Score = 4 No other data available
Date of ABG		
Date of ABG	28/03/02	Y
Pre AGB Radiograph	27/03/02	Y
Post Op Radiology	24/07/02	Y
Expansion Information:	Expansion started: 14/11/01 Expansion finished: 07/02/02	Outcome Assessment Score=2 50-75% bone fill
10 yr photos		
10 yr photos	22/05/03	Noted as taken. Not on excelicare
10 yr study models	05/11/02	Y
10 yr x-ray	22/05/03	Noted as taken. Not on excelicare
10 yr ortho audit	22/05/03	Golson = Not Available Centre Lines: Upper left by 2 mm Lower_?_ by 2 mm HB Total Sum = -5
15 yr photos		
15 yr photos	08/06/07	Y
15 yr study models	27/10/06	Y
15 yr x-ray	27/01/06	Y
15 yr ortho audit	-	N
20 yr photos		
20 yr photos	04/04/12	Recorded on NCAS as taken
20 yr study models	04/04/12	Recorded on NCAS as taken
20 yr x-ray	04/04/12	Recorded on NCAS as taken

Cleft No: 556

Record	Date	Available
5 yr photos	09/12/99	Y
5 yr study models	03/02/00	Y
5 yr ortho audit	03/02/00	5 Year Old Index Score = 2 No other data available
Date of ABG	11/05/06	Y
Pre AGB Radiograph	21/03/06	Y
Post Op Radiology	01/08/06	Y
Expansion Information:		No Expansion Information >75% bone fill
10 yr photos	03/03/05	Y
10 yr study models	05/04/05	Y
10 yr x-ray	13/07/04	Y
10 yr ortho audit	22/01/08	Golson = 3 No other data on form
15 yr photos	07/10/10	Y
15 yr study models	09/11/10	Y
15 yr x-ray	07/10/10	Y
15 yr ortho audit		N
20 yr photos	-	N
20 yr study models	-	N
20 yr x-ray	-	N