Three-Dimensional Assessment of Dentofacial Deformity in Children with Clefts

By

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To my beloved sister, Susan F.G. Garrahy

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Glossary of Anthropometric Landmarks

Alar curvature - (ac) - the most lateral point in the curved baseline of each ala, indicating the facial insertion of the nasal wingbase.

Alare - (al) - most lateral point on each alar contour.

Alare ' - (al') - the marking level at the mid portion of the alae (al' - al') where the thickness of each ala is measured.

Cheilion - (ch) - the point located at each labial commissure.

Christa philtri - (cph) - the point on each elevated margin of the philtrum just above the vermillion line.

Columellar high point - (c) - the point on each columella crest, level with the top of the corresponding nostril.

Endocanthion - (en) - the point at the inner eye fissure commissure.

Exocanthion - (ex) - the point at the outer eye fissure commissure.

Gnathion - (gn) - the lowest median landmark on the lower border of the mandible.

Gonion - (go) - the most lateral point on the mandibular angle close to the bony gonion.

Labiale superiorus - (ls) -upper midline limit of the vermillion border.

Labiale inferiorus - (ls) - lower midline limit of the vermillion border.

Nasion - (n) - the deepest point of the midline concavity between nose and forehead.

Otobasion inferiorus - (obi) - the point of attachment of the earlobe to the cheek.

Pogonion - (pg) - the most anterior midpoint of the chin.

Pronasale - (prn) - the most protruded point on the nasal apex.

Stomion - (sto) - midline contact point between upper and lower lip, or closest upper and lower lip points where lips incompetent.

Stomion inferiorus - (stoi) - the crossing of the vertical facial midline and the upper border of the mucocutaneous border of the lower lip.

Stomion superiorus - (stos) - the crossing of the vertical facial midline and the lower border of the mucocutaneous border of the upper lip.

Subalare - (sbal) - the point at he lower limit of each alar base, where the alar base disappears into the skin of the upper lip.

Subnasale - (sn) - the midpoint of the angle at the columella base where the lower border of the nasal septum and upper lip surface meet.

Subnasale' - (sn') - the midpoint of the columella crest where the thickness of the columella is measured.

Tragion - (t) - the notch on the upper margin of the tragus.

List of Abbreviations

2D	Two dimensional
3D	Three dimensional
4D	Four dimensional
AcL	Left alar curvature
ACPA	American Cleft Palate Association
AcR	Right alar curvature
AlL	Left alare
AlR	Right alare
Al'Li	Left inner alare dash
Al'Lo	Left outer alare dash
Al'Ri	Right inner alare dash
Al'Ro	Right outer alare dash
ANOVA	Analysis of variance
AU	Action unit
BCLP	Bilateral cleft lip and palate
ChkL	Left cheekpoint

ChkR	Right cheekpoint
ChL	Left cheilion
ChR	Right cheilion
CLP	Cleft lip and palate
CphL	Left christa philtri
CphR	Right christa philtri
CPO	Cleft palate only
CL	Left columellar high point
CMM	Co-ordinate measuring machine
CMOS	Compound metal oxide semiconductor
CR	Right columellar high point
CSAG	Clinical Standards Advisory Group
CT	Computerised tomography
CV	Co-efficient of variance
DICOM	Digital Imaging and Communication in Medicine
DLT	Direct linear transformation
DMFS	Decayed, missing and filled teeth score
EnL	Left endocanthion

EnR	Right endocanthion
ExL	Left exocanthion
ExR	Right exocanthion
Gn	Gnathion
GSL	Gingival smile line
Lhp	Lip high point
Llp	Lip low point
Ls	Labrale superiorus
Li	Labrale inferiorus
MRI	Magnetic resonance imaging
MSRA	Maximal static response assay
N	Nasion
ObiL	Left otobasion inferiorus
ObiR	Right otobasion inferiorus
OFC	Orofacial clefting
PCI	Peripheral component interconnect
PD	Percent displacement
Pg	Pogonion

Prn	Pronasale
RMSE	Root mean square error
SbalL	Left subalare
SbalR	Right subalare
SCALP	Scottish Association for cleft lip and palate
Sl	Sublabialis
Sn	Subnasale
SNAP	Scottish Needs Assessment Programme
Sn'L	Left subnasale dash
Sn'R	Right subnasale dash
Sto	Stomion
Stoi	Stomion inferiorus
Stos	Stomion superiorus
T	Tragion
TGFA	Tissue growth factor alpha
.tif	Tagged image file
UCL	Unilateral cleft lip
UCLP	Unilateral cleft lip and palate

USB	Universal serial bus
UPS	Uninterruptable power supply
V, L, S	Vermillion, lip, scar
VRML	Virtual reality modelling language

Publications and Presentations

Published Abstract:

Assessment of the Stability of Recorded Paediatric Facial Expression

British Journal of Oral & Maxillofacial Surgery, 2002, vol. 40, no. 4, p. 371

Presentations to Scientific Societies:

1. Application of Computerised Stereophotogrammetry to Assessment of Paediatric Facial Morphology

Presented to the 9th International Congress on Cleft Palate and Related Craniofacial Anomalies

Göteborg, Sweden,

June 24-28, 2001

2. Early Assessment of Surgical Outcome in Unilateral Cleft Lip and Palate

Presented to the Craniofacial Society of Great Britain & Ireland

East Grinstead, England, 11 - 12 April, 2002

Summary

Background

- Changes in clinical management.
- Advances in non-invasive three-dimensional imaging.
- Developments in methods of shape analysis.

Aim

- To assess three-dimensional dentofacial deformity with a view to early appraisal of primary surgical outcome.

Results

- Significant differences in upper lip morphology were found between the cleft children and their unaffected peers.
- Nasal asymmetry that became more obvious in function was noted in cleft children.
- The maxillary dental arches of the children with repaired cleft palate were shallow, short and narrow.
- The dental arch deformity and the facial soft tissue deformity were unrelated.

Contributions to the field

- It has been shown that deviation from normal could be detected as young as 3 years of age using computerised stereophotogrammetry.

- Preliminary, objective, three-dimensional analysis of facial function has been completed in young children.

- The accuracy of three-dimensional CT scanning of dentate study models and the time cost of data collection were quantified.

- This study has produced a body of three-dimensional data that can test and support analytical advances.

The background to this study was formed by changes in clinical management of individuals with orofacial clefting, advances in non-invasive three-dimensional imaging and developments in methods of shape analysis.

To address the problem of poor clinical outcome demonstrated in the United Kingdom in 1997, the provision of primary care was restricted to fewer larger centres. Recent advances in vision based three-dimensional imaging technology with subjective improvement in image quality offered a possible method to judge surgical outcome. The dimensional accuracy had not been quantified and comprehensive analysis of the data collected remained underdeveloped.

The aim of this study was to assess three-dimensional dentofacial deformity with a view to early appraisal of primary surgical outcome. The subjects were 3-year-old Scottish children with unilateral cleft lip, or cleft lip and palate, and a matched control sample. The null hypotheses were that there would be no differences between the samples for any of the features being investigated.

Images of the children's faces, at rest and performing a maximal smile, were recorded using computerised stereophotogrammetry. Dental impressions were recorded. Direct measurement and CT scanning of the dental study models was carried out to examine the dental arch shape and size.

Prior to application to the study samples, the system errors for computerised stereophotogrammetry and three-dimensional CT scanning were quantified. The reproducibility of paediatric expressions was determined. And found to be acceptable.

Linear analysis of the facial and dental arch data was carried out to allow comparison with the work of other researchers in the field. The data were also analysed using more advanced shape analysis techniques. Procrustes and Bookstein superimposition were employed to create average face and dental arch shapes for the subject types, retaining the geometric integrity of the

biological shapes. Symmetry was investigated, without reference to a midline plane.

Significant differences were found between the cleft children and their unaffected peers. In addition to visible scarring of the upper lip, there was irregularity of the lip outline, which was exacerbated on smiling. The repaired cleft nose was splayed and asymmetric and this asymmetry became more obvious in function. The nasal abnormality was not limited to the side of the face affected by the congenital cleft.

The maxillary dental arches of the children with repaired cleft palate were shallow, short and narrow anteriorly, possibly suggesting deficiency of nasomaxillary complex development. The dental arch deformity and the facial soft tissue deformity were unrelated to each other on correlation testing. Primary surgery had not restored normal appearance and function and may have had adverse effects on maxillary growth potential. The null hypotheses were rejected.

Geometric analysis of the data was very sensitive, detecting statistical significance in excess of clinical significance, especially in the dental arches. Presenting the results, which are not in metric units, of such analysis to a clinical readership remains a challenge.

This study has made several contributions to the field of facial assessment in children with clefts.

It has been shown that deviation from normal could be detected as young as 3 years of age using computerised stereophotogrammetry. This would permit early assessment of surgery and early identification of adverse features of management.

Preliminary, objective, three-dimensional analysis of facial function has been completed in young children. Examination of maximal expression is a rudimentary investigation of facial function but this had not been investigated in young children previously. The assumption of reproducibility of resting and smiling expressions was tested and found to be acceptable. The assumption of homology of anatomical landmarks in varying expressions that underpinned the analytical model remains to be tested.

Three-dimensional CT scanning of dentate study models was a new use of generally available equipment. The accuracy of this technique and the time cost of data collection were quantified.

The stereophotogrammetry equipment had to be modified to permit shorter capture times for use with children. This tested the limits of camera and flash synchronisation capability and forced further development of the interface with the personal computer. The next stage in the development of the equipment will simplify, and therefore shorten, the image collection process.

It will be necessary to develop more robust analytical methods to analyse longitudinal three-dimensional data. The variability introduced by multiple facial growth centres will further challenge the validity of anatomical landmarks as points of reference. This study has produced a body of three-dimensional data that can test and support analytical advances.

Chapter 1

Introduction

1.1. Orofacial Clefting

Facial appearance can shape a person's destiny as society attaches great value to attractive physical appearance and attributes positive personal characteristics to individuals who are attractive. The converse is also true. This awareness of facial appearance begins in early childhood, influencing family dynamics and social interaction with contemporaries. Conditions that affect the facial appearance, therefore, may have wide ranging consequences. Facial deformity and unattractiveness are not synonymous but regardless of the level of attractiveness, individuals with facial abnormality may suffer adverse social effects.

Cleft lip and / or palate is one of the commonest of the congenital malformations affecting the head and neck. Clefting may occur as an isolated event or may be one feature of a congenital syndrome. It has a wide spectrum of severity, ranging from bifid uvula or submucous palatal clefting to complete bilateral cleft lip and palate. Isolated cleft palate does not directly affect the facial soft tissues but the effect of the original cleft and subsequent palatal reconstruction may become manifest, indirectly, through skeletal abnormalities.

The child with a repaired cleft enters the wider society outside the family as early as 3 years of age, when he first attends nursery school. It is incumbent on the team managing the child's care that he resembles his peers as closely as possible. He should be able to communicate effectively and interact with his peers and teachers as unaffected children do. It has been shown that teachers interact with children with facial deformity differently, perceiving their behaviour as more disruptive than unaffected children. This is another reason to aim to restore appearance to as near normal as possible early.

If normal appearance is to be achieved, a clear description of what constitutes the range of normal is required.

1.2. Clinical Management

A single embryological misadventure in the first trimester of gestation can create a structural defect requiring multidisciplinary care and staged surgical reconstruction into adulthood. The orofacial cleft may lead to significant facial and dental deformity and impairment of verbal communication, both expressive and receptive. The aim of treatment is to avoid all of these adverse sequelae. Comparison of different cleft management centres demonstrates that the quality of care has a direct relationship with eventual outcome.

The challenge facing the surgeon performing the primary reconstruction is to restore anatomical continuity that will persist, without destroying growth potential. Some features of the repaired cleft, such as the palate, will evolve with growth. Successful obturation of the patency in the neonatal period may not be maintained. Other features, such as the reconstructed nose, may remain structurally unchanged to adulthood. Successful primary surgery is the key to good treatment outcome.

The dental arch relationship may be considered a signifier of the health of the nasomaxillary complex. Occlusal deformity can be modified by orthodontic and orthognathic treatment. Alveolar bone grafting and early orthodontic intervention can improve the occlusion in childhood and adolescence but orthognathic surgery and definitive reconstruction must be deferred until maturity.

1.3. Facial Expression

Orofacial clefting affects dynamic tissue. Assessment of surgical repair is not complete without an assessment of the repaired tissue in function. Investigation of this complex structure is a challenge to researchers. The variability of movement of the complex facial musculature and the effect of emotion on the face complicates objective assessment of structure. Qualitative and quantitative descriptions of the range of movement of the repaired defect would be useful as another form of clinical appraisal for the operating clinician.

Other aspects of function in orofacial clefting have been examined, such as speech and hearing, but the function of facial expression has received less attention.

1.4. Imaging Techniques

From the intricate pen-and-ink drawings found in historical surgical texts to evolving radiation-free imaging modalities, graphic presentation of the subject with orofacial clefting has added to clinical knowledge. It is the advance in statistical analysis of the images produced that offers the greatest opportunity to render such subjective impressions objective.

"Virtual" subjects with orofacial clefting have been created to aid teaching in surgical techniques. Such images are relatively crude as little is known about the soft tissue of the young subject beyond the findings in the area of the cleft at the time of surgery.

1.5. Assessment of Treatment Outcome

Standards of cleft care in Britain have, unfortunately, been found to fall below those set by other centres. The scattered model of treatment delivery has been criticised and the quality of record keeping over extended clinical management has also been found to be deficient.

A recent decision was taken, at national level, to concentrate cleft care in fewer centres, with a full range of clinical expertise available, treating higher numbers of subjects. With larger surgical centres and fewer surgical protocols, there will an opportunity to examine a larger body of less varied data. This will, in turn, provide a base of evidence for subsequent clinical management decisions.

The aim of clinical intervention should always be to restore to normality, not to judge the outcome of cleft management against that of other cleft subjects. For every child born with orofacial clefting there are almost 1000 children born without, this larger population should set the standard for cleft outcome.

Chapter 2

Literature Review

2.1. Overview of Cleft Management

2.1.1. Incidence

In Scotland each year, for the purposes of health service planning based on need assessment, approximately 100 children are considered to be born with orofacial clefting (OFC), (SNAP Oral Health Group 1997). The Scottish population has a higher incidence of congenital malformations, including OFC, than the rest of the United Kingdom (UK) (Stone & Dolk 1994). It has a unique ratio of cleft lip or cleft lip and palate to isolated cleft palate of almost 1:1; elsewhere a ratio of 2:1 is more common. A review of cleft births, registered in the West of Scotland, from 1980 to 1985, reported a total birth prevalence of 1.53 per 1000 births (FitzPatrick et al. 1994). There was a higher rate of associated congenital abnormalities than in other areas of the UK and an unusually high rate of isolated cleft palate. A review of cleft births in the same area over a longer period, from 1974 to 1985, reported a similarly high rate of 1.56 per 1000 and confirmed the high proportion of cleft palate only (CPO), 52% of the total, (Womersley & Stone 1987).

Northern Ireland, which has a similar ethnic background and close geographic proximity to Scotland, had a similar proportion of isolated cleft palate at 53%, but a lower birth prevalence of 1.28 clefts per 1000 births (Gregg et al. 1994). A birth prevalence of 1.4 per 1000 births has been reported in Southeast Scotland and the Highlands in the period 1971 to 1990, with 45% of the cases being isolated cleft palate (Bellis & Wohlgemuth 1999). The latter two studies were of clefts as a proportion of live births, which would explain the apparent discrepancy between the reported prevalences in adjacent geographic regions.

Outwith the United Kingdom, prevalence of OFC varies from the high rates observed in Japan and certain Scandinavian countries to the low rates observed in African ethnic groups, regardless of location. It must be recognised that some of the reported variations in the prevalence of OFC may be related to registration methodology rather than true variation in birth rates. Countries such as Norway, with high rates of OFC and centralised cleft management services, are acknowledged to have the most reliable records of cleft registration.

2.1.3. Classification

In the early part of the 20th century, Veau categorised clefts of the lip and / or palate as follows: (1) clefts of the soft palate alone; (2) clefts of the hard and soft palate; (3) complete unilateral clefts of the lip and palate and (4) complete bilateral clefts of the lip and palate. This classification did not include clefts of the lip alone or incomplete clefts. Cleft lip and cleft lip and palate involve the primary palate and result from failure of fusion of one or both of the maxillary prominences with the fused nasal prominence. Isolated cleft palate is a deformity of the secondary palate resulting from failure of fusion of the palatal shelves (Ferguson 1988).

In 1958, a classification that recognised the incisive foramen as the boundary dividing clefts of the primary palate from the secondary palate was introduced (Kernahan & Stark 1958). The stripped Y modification of this classification was added in 1971, which assigned identifying numbers to areas of the lip and palate and allowed more detailed description of the degree of clefting (Kernahan 1971). In the United States of America (USA), a palindromic system (LAHSHAL) has been adopted by the American Cleft Palate - Craniofacial Association to describe the areas affected by the cleft. It uses upper and lower case letters to describe complete and incomplete clefting. For example, LAHS would describe a complete unilateral cleft lip and palate (UCLP) whereas LA would describe a unilateral cleft lip (UCL) affecting the alveolus.

2.1.4. Aetiology

Orofacial clefting has a multifactorial aetiology and occurs as a result of an interplay of genomic and epigenetic factors of varying significance.

2.1.4.1. Genetic

Segregation analysis of the inheritance of the non-syndromic cleft lip and palate (CLP) phenotype in pedigrees has indicated a model of autosomal major gene inheritance, with or without multi-factorial contributions. Linkage and association analyses have identified multiple clefting loci, 19q13 and 14q24, 17q12, 4p13 and 4q25-31 (Shaw et al. 1993); (Maestri et al. 1997); (Lidral et al. 1998). Different results from different research groups could be attributable to the genetic difference of these populations.

2.1.4.2. Adverse Environmental Factors

Lifestyle choices of the mothers of cleft infants have been implicated in the aetiology of OFC. Maternal smoking and pre-natal alcohol exposure have been identified as adverse factors (Khoury et al. 1989); (Shaw & Lammer 1999). North American studies had indicated that maternal first trimester smoking and transforming growth factor alpha (TGFA) locus mutations were associated with non-syndromic OFC, and that a synergistic effect occurred. However, a Danish case control study confirmed the association with smoking but found no association between the TGFA genotype and clefts (Christensen et al. 1999). Scandinavian CLP frequency is among the highest in the world and, of note, there was a 25% prevalence of the TGFA genotype reported in both the case and control groups in Denmark while 14% prevalence was reported for other white control groups.

Risk of clefting, 10 times that of normal, was detected in a group of substance abusing mothers in Australia but no particular drug was implicated. Maternal nutrition was suggested as another factor in cleft causation in this group (Thomas 1995). Prenatal exposure to amphetamines has been definitely associated with clefts (Plessinger 1998). Environmental pollution has also been identified as an aetiological agent, with high environmental lead levels and open chemical combustion co-inciding with local increases in birth prevalence of OFC, in Italy and the Netherlands respectively (ten Tusscher et al. 2000); (Vinceti et al. 2001). The higher incidence of children with clefts born to mothers with epilepsy is attributed to the teratogenic effect of anticonvulsant drugs, such as phenytoin, rather than familial aggregation of epilepsy and clefting disorders. The teratogenic effect of phenytoin is related to its folate antagonist properties, which leads to the next segment of aetiological factors (Dansky et al. 1992).

2.1.4.3. Nutrition

The role of folate supplementation in the prevention of neural tube defects, which is another failure of embryological fusion similar to clefting, has lead to the investigation of folate deficiency as a factor in OFC (Loffredo et al. 2001). A link between hyperhomocystinaemia, a metabolic marker for folate deficiency, and oral clefting has been suggested (Wong et al. 1999). A recent Hungarian prospective cohort study concluded that high dose folic acid supplementation should commence before weeks 7-8 gestation and continue throughout pregnancy (Czeizel et al. 1999). Maternal malnutrition has also been suggested as an aetiological factor, based on a study in rural communities where the seasonal changes in rates of OFC births has been associated with growing seasons and fluctuating food supplies (Cooper et al. 2000). The link between pre-existing maternal diabetes and OFC is documented but not yet explained (Aberg et al. 2001).

Deprived maternal socio-economic circumstances are associated with higher rates of non-chromosomal congenital anomalies. While clefting is a congenital anomaly, a recent small epidemiological study in the UK found that clefting was not actually tied to maternal deprivation category (Vrijheid et al. 2000). As relatively deprived socio-economic circumstances, poor nutrition, maternal smoking and substance abuse are positively correlated, identifying the most significant aetiological agent is very difficult (Pugh et al. 1991); (Bonassi et al. 1994); (Royo-Bordonada et al. 1997); (Siahpush et al. 2002); (Wardle et al. 2001); (Radhika et al. 2002).

2.1.5. Applied Anatomy

Investigation of the effects of OFC on the osseous, nasal and perioral anatomy serves several purposes. Simple description of the neonatal cleft provides a basis for clinical classification. Dissection and other forms of exploration provide a basis for clinical management, especially primary reconstruction, and suggest reasons for the subsequent evolution of the congenital defect (Breitsprecher et al. 1999); (Breitsprecher et al. 2002). OFC is associated with other anomalies but rarely leads to perinatal mortality, so few studies of the neonatal deformity are based on cadaveric dissection. The studies rely instead on imaging techniques such as cephalometry (Ishiguro et al. 1976). At the time of primary repair, clinical observation of the distorted anatomy is possible and impression taking for cast studies can be performed on the anaesthetised child (Mulliken et al. 1993); (Wada et al. 1990). Wide dissection of normal tissue in the lip and palate has an adverse effect on subsequent growth and is best avoided at the time of surgery. Perioperative anatomical study must be restricted, therefore, to the immediate area of surgical dissection (Ishikawa et al. 1999).

The UCLP defect allows direct comparison of normal and abnormal anatomy. There is consensus on the rotated premaxilla and retropositioned, collapsed lateral maxillary segment. The deviation of the nasal septum to the non cleft side anteriorly and to the affected side posteriorly, is agreed upon as is the shortness of the columella, outwardly rotated cleft side alar bases and deficient vestibular lining on the cleft side. The condition of the lateral cartilage is considered attenuated by some, histologically normal and only macroscopically deformed by others (Burt & Byrd 2000); (Breitsprecher et al 1999). The gross anatomy of the unilateral cleft lip is obvious on visual inspection, with a short philtrum, absent philtral column and partially absent Cupid's bow.

The effect of clefting on the facial muscle slings, their symmetry and power vectors has not been studied in children. This means the basis for functional surgical repairs has had to be extrapolated from dynamic studies in cleft adults, post repair, and at best, small microsurgical dissection studies allied to imaging such as MRI or 3D CT scanning (Breitsprecher et al 2002).

2.1.6. Surgical Repair

The extensive literature in this area has not been exhaustively reviewed as the surgical intervention in this study was standardised and the outcome of cheiloplasty and palatoplasty, in general, was being assessed rather than the outcome of particular surgical variations. The aim of the review of the literature in this area was to map the development of concepts of surgical reconstruction of lip and palate and to point to some of the areas of controversy in reconstruction, rather than discuss surgical techniques in detail.

The first cleft lip repair is credited to an unidentified Chinese physician in the 4th century AD, supposedly performed on a subject who went on to become the Governor General of six Chinese provinces (Burt & Byrd 2000); (Kirschner & LaRossa 2000). The earliest lip repairs, which were simple incisions of the cleft edges and straight line closures, evolved through the mid 19th century when lateral flap advancement was described. In the mid to late 20th century rotation-advancement and functional repair principles were introduced. The developments in cleft lip repairs have been directed towards anatomical reconstruction and away from simple closure of a patency.

The Millard rotation advancement technique, combining lateral advancement into the upper portion of the lip and downward rotation of the medial segment is very popular. Millard repair was the procedure performed in the cleft subjects reported in this thesis The Delaire "functional" repair, differential reconstruction of the orbicularis oris muscle in UCLP repair, also has its advocates (Markus et al. 1993); (Markus & Delaire 1993).

Correction of the associated cleft alar deformity is now considered by many to be part of the lip repair procedure with some claiming beneficial effects for active pre-surgical nasoalveolar moulding (Cutting et al. 1998). Opinion is united on the longevity of good nasal appearance and the indications for subsequent revision surgery (McComb & Coghlan 1996); (Russell et al. 2001). The beneficial effect of secondary rhinoplasty has been shown to be minimal (Timoney et al. 2001).

The rate of development of palatal repair has lagged behind that of lip repair. Obturation of the palatal cleft mirrored straight-line lip closure as a historical treatment. The 19th century von Langenbeck closure of the palate with mucoperiosteal flaps was superseded by palate lengthening procedures such as the Veau-Wardill - Kilner V-Y pushback which remains in use today. Palatal closure with undermining of the mucoperiosteum, rather than releasing incisions, has been recommended to minimise the exposure of denuded bone but tension free repair often demands relaxing incisions (Furlow 1986). Opinion on the effect of bone denudation is divided. Palatal scarring with "push back" procedures may, or may not, affect vertical palatal growth (Ishikawa et al 1999). A switch from "push back" palatoplasty to a procedure requiring less palatal denudation in a recent Danish study, showed that gain in arch width growth occurred only in cases of velar clefting (Friede et al. 2000).

The timing of palatal closure is still debated. It has been recognised that the best speech results are obtained when the palate is repaired before meaningful connected speech develops. As babbling in early infancy is accompanied by phonologic development, some surgeons advise palatoplasty as early as 6 months (Copeland 1990). Closure before 12 to 18 months seems to have the same effect on speech as early closure (Kirschner et al. 2000). Velar closure in infancy, with hard palate closure delayed until midface growth is almost complete, is generally agreed to be an unsatisfactory procedure because of the poor speech outcomes (Van Demark et al. 1989).

2.1.7. Clinical Management

From surgery on adults with unrepaired OFC in less affluent countries to considerations of prenatal diagnosis and repair in utero in Western Europe, a major determinant of clinical management of the multi-faceted problems of OFC is the local health service model (Mars et al. 1990); (Canady et al. 1994). The benefits of a team of health care professionals providing multidisciplinary care to individuals with OFC from birth, or earlier in cases of prenatal diagnosis, is not disputed. It is recommended that specialist nurses, orthodontists, paediatric dentists, speech and language therapists, clinical geneticists, psychologists, ENT (ear, nose and throat) surgeons, plastic and oral and maxillofacial surgeons are part of the management team (Shaw et al. 1996). The team should be supported by administrative staff to maintain databases and records archives and to ensure continuity of care from birth to adulthood.

Early surgical repair of the lip, and later repair of the palate, to confer a near normal appearance and assist normal speech development is also agreed upon. In cases of palatal clefting, routine alveolar bone grafting is recommended in the mixed dentition. Early treatment is considered successful if the need for later revision such as maxillary osteotomy and secondary cheiloplasty and rhinoplasty is low. Comparisons of surgical techniques and timing, investigation of the efficacy of presurgical moulding and studies of primary bone grafting are studies of details of treatment, within the broad management structures described above (Friede & Enemark 2001); (Markus et al 1993); (Mishima et al. 1996); (Maull et al. 1999).

The investigation with the greatest impact on clinical management is the national, or international, study of models of treatment delivery. Two examples of these surveys, rather than scientific research studies, are the Clinical Standards Advisory Group (CSAG) report in the U.K. in 1995 and the earlier six European centre trial in 1992 (Shaw et al. 1992); (Sandy et al. 1998). The latter study of cleft outcome was based in north western Europe and examined the end result of treatment of unilateral cleft lip and cleft lip and palate in six centres where clinical management was state funded and, in some cases, centralised to one national centre. When the standards of facial appearance, skeletal development and dental occlusion were published, these became benchmarks for outcome of extended clinical management against which other clinicians could judge their own work. The outcome of some centres compared well with the published standards and there was no perceived need to significantly alter clinical practice (Leonard et al. 1998).

The CSAG study was conducted in the United Kingdom and was a result of the 1992 trial. The latter gave cause for concern about the quality of specialist cleft care in the United Kingdom when compared with European data. This subsequent nation-wide study of speech, facial development and appearance, oral health, bone graft quality and patient satisfaction at the ages of 5 and 12 years had a significant effect on the provision of cleft services for a population of sixty million people. The recommendations of the CSAG Committee were directed at clinicians, Royal Colleges and Faculties, provider units, the Department of Health and the Office of National Statistics (Sandy et al 1998). It was recommended that cleft management services should be concentrated in 8-15 centres in the U.K., not the 57 operational at that time. A full range of clinical skills was to be available at the major service centres. It was recommended that a common database should be agreed on, which would provide data for comparative audit studies. A specialised training pathway for cleft surgeons was to be agreed upon. The completeness of the recording of cleft births in the U.K. was criticised and this was to be improved.

In countries where the health service is not state-funded or state-managed, research methodology can be affected by what one author referred to as "clinic shopping" (Hurwitz et al. 1999). Rather than attending centralised clinics, patients may attend several units for piecemeal clinical management, which precludes long-term follow-up and provides only small sample sizes for research. This forces a heavy reliance on archived data (Prasad et al. 2000).

2.1.8. Outcome Assessment

The assessment of treatment outcome in a condition such as OFC which involves serial surgical intervention, staged orthodontic treatment and altered growth potential, whether inherent or iatrogenic, is controversial. Methods of outcome assessment can be divided into assessments of form or function or amalgamations of the two. Assessments of speech and hearing are tests of function that can be related to the form of the repaired cleft palate and velopharynx giving an indirect assessment of the latter structures. This thesis deals with the deformity of OFC that may be visible to the public, or on visual clinical examination of the anterior oral cavity, so speech and hearing will not be discussed. Considerations of the psychological morbidity associated with facial anomalies, which is a confounding factor in all aspects of this condition, are also omitted in this literature review.

Clinical examination of an individual with repaired OFC by an experienced clinician can provide a comprehensive appraisal of the outcome of treatment. It is, however, highly subjective, can be unreliable and the recorded descriptive data is not amenable to statistical analysis even if minutely detailed clinical notes are recorded (Al-Omari 2001). The V,L,S (vermillion, lip, scar) classification of secondary lip deformity based on the appearance of the vermillion, lip and scar is a systematic approach to appraisal of clinical photographs (Assuncao 1992). This, however, can be criticised as an assessment of treatment outcome for the same reasons that clinical examination of the live subject can be criticised. The challenge has been to provide the clinician with methods of outcome assessment that are objective, sensitive, specific and reliable so that studies across time and distance can be valid.

The length of time that a course of treatment may span means that sufficient data for meaningful statistical analysis of outcome may take longer than any individual clinician's working lifetime to collect (Shaw et al 1996). Early applicability of assessment methods shortens this wait for data and also allows assessment of the early stages of treatment, the primary cleft repair, before subsequent treatment interventions are made and differential growth rates confound the clinical picture (Atack et al, 1997).

Objective assessment methods can be classified into two broad categories, objectivity in the data collection process or objective analysis of standardised clinical data. Recording the number of absent teeth in the cleft arch would be an example of objective data collection and measurement of radiographic, or other images, would be an example of analysis of standardised clinical data. The ideal method would incorporate objectivity at both the data collection and analysis stages.

Examination of the extensive literature pertaining to analysis of standardised data in OFC identifies certain noteworthy trends. Despite the contribution of cephalometry to the knowledge of growth and development of the OFC, non-invasive imaging techniques with attendant risk reduction are considered preferable. Researchers in imaging techniques such as laser surface scanning, opto-electronic digitisation or structured-light scanning techniques have all stated that reduction of radiation dosage is an advantage of the techniques under investigation (Aung et al. 1995); (Ferrario et al. 1996); (Kawai et al. 1990).

Objective analysis has advanced from the skeletal to the soft tissue morphology (Asher-McDade et al. 1992); (Mackay et al. 1994); (McCance et al. 1997a). Subjective panel assessment of clinical photographs and grading of surgical outcome based on perceived facial impairment is being superseded by accurate measurement of facial surface images (Slade et al. 1995); (Tobiasen & Hiebert 1994); (Hurwitz et al 1999). The statistical analysis of data collected has become more complex and robust latterly (Hennessy & Moss 2001); (Grayson & Cutting 2001); (Yamada et al. 1999). It is clearly the ready availability of powerful computational ability that has permitted the above trends to develop (Urquhart 1997).

Paediatric application of recently available assessment methods has been limited to older children and to children with facial pathology rather than normal population samples. This occurs despite the early surgical treatment of individuals with OFC and the documented early maturation of the facial complex (Farkas et al. 1992). The collection of clinically justified radiographic data and impression recording perioperatively, in the first years of life, has provided records of the initial cleft defect and immediate result following cheiloplasty (Han et al. 1995); (Wada et al. 1984). The evolution of the soft tissue repair between initial surgery and later childhood is largely undocumented as many methods of data collection require much co-operation and are not feasible in young children without sedation or anaesthesia (Hermann et al. 1999).

In the United Kingdom, age five is the first generally agreed post-operative assessment point. The minimum data set consists of speech assessment, dental study models and clinical photographs, frontal, profile and worm's eye views. The trends identified above in research into certain aspects of OFC in adults have yet to become manifest in research in the paediatric cleft population. Even though facial appearance affects social interaction with family and peers, paediatric facial appearance has not been amenable, previously, to objective examination (Kasuya et al. 2000); (Coy et al. 2002).

2.2. Morphometrics and Anthropometry

2.2.1. Shape Analysis

2.2.1.1. Landmarks

A landmark is a point of correspondence on each object that matches between and within populations (Dryden & Mardia 1998). The specific point of correspondence on a biological form or image of a form is located according to some rule. Examples of such rules are the scheme of generally accepted facial soft tissue landmarks defined by Farkas et al (1994) or the more variable dental arch landmarks defined by a variety of authors (Moorrees et al. 1969); (Sillman 1964); (Moyers et al. 1976); (Farkas 1994).

Landmarks can be anatomical, mathematical, or pseudo-landmarks. Anatomical landmarks correspond between subjects in a biologically meaningful way, such as the tip of a tooth. Mathematical landmarks are located according to some geometrical property of the object, such as maximal curvature. Pseudo-landmarks are constructed points on an organism, either around the outline or between anatomical or mathematical landmarks.

Landmarks have also been categorised into three types labelled type I, type II and type III. Type I landmarks occur at tissue junctions, type II landmarks are defined by local properties such as curvature and type III are constructed landmarks. These three types do not correspond directly to the previously defined anatomical, mathematical and pseudo-landmarks. For example, anatomical landmarks may be of type I or type II (Dryden & Mardia 1998).

2.2.1.2. Traditional Morphometry

Morphometry derives from the Greek: "morph," meaning "shape," and "metron," meaning "measurement." Anthropometry is a specialised area of morphometry relating to the human form. Facial anthropometry is, therefore, the measurement of the shape of the human face. "Traditional" is the prefix added to describe the application of statistical methods to arbitrary collections of size or shape variables such as distances and angles. Even though the distances or measurements are defined to record biologically meaningful aspects of the organism, the geometrical relationships between these measurements are not taken into account. Measurement of symmetry based on comparison of paired distances is an example of this form of morphometry. It has been shown that there is no predictable relationship between significant findings in measurements and in landmark configurations (Shaner et al. 2000). Analysis of paired distances may detect differences in the size of regions of the face, but will not demonstrate any abnormalities in the relationship of those regions to each other.

2.2.1.3. Geometric Morphometry

As shape can be defined as the geometric properties of a configuration of points that are invariant to changes in translation, rotation and scale, this variety of morphometry deals with the relationship between landmarks in three dimensions. Analysis of the symmetry of a whole facial configuration of landmarks would be an example of geometric morphometry. One challenge of clinical application of this form of analysis is the presentation of results to the non-morphometrician. Some authors have presented colour coded schematic representations of clinical findings and others have presented line drawings of facial landmark configurations to give an impression of depth (Ferrario et al. 1998a); (McCance et al. 1997b). Scaling of facial shape to the same size for all subjects means that units of measurement familiar to the clinician are replaced by units which are divisions of the created average sized object (Grayson & Cutting 2001).

To become generally applicable in clinical assessment, the "currency" of shape analysis will need to be made comprehensible to the clinician. This may happen as certain methods of superimposition and data analysis, such as Procrustes analysis, become more widely used and applied to data acquired using a variety of imaging methods (Hennessy & Moss 2001); (Trotman et al. 2000). Procrustes analysis is a method of superimposition that rotates, translates and scales configurations of 3D landmarks to a position of maximal agreement while retaining shape. A criticism of geometric morphometrics is the arbitrariness of the choice of alignment procedure but it has been demonstrated that the choice of registration method is unimportant if the variation in shape is small (Lele & Richtsmeier 1991); (O'Higgins & Jones 1998).

2.2.1.4. Symmetry

This is one particular form of shape analysis that is readily understood and easily illustrated. Symmetry of form is a basic biological feature and physical symmetry is considered a desirable and attractive feature in humans, with asymmetry inversely related to perception of attractiveness (Grammer & Thornhill 1994). A seminal text defined three types of symmetry occurring in nature (Van Valen 1962). Anti-symmetry is the development of unilateral dominance on either side of the body such as handedness. Directional-symmetry is the normal development of one-sided structures such as the heart or liver. Fluctuating symmetry is the differing rates of development of sides of the body, such as the right and left face. The existence of fluctuating asymmetry in the face may explain the variability of facial asymmetry reported in the literature (Shaner et al 2000). Measurement of asymmetry has often been applied to the cleft face with its unilateral malformation.

Perfect symmetry is absent in nature and it has been shown that up to 3mm discrepancy between paired facial measurements in adults is not perceived as imbalance (Farkas & Cheung 1981). This range of normal asymmetry is a good example of the theory of epigenetic or extrinsic buffering that affects human growth development. It has been shown that dental asymmetry was generally higher in subjects with mental retardation which was associated with pre-natal rather than post-natal or birth-related insult (Barden 1980). The conclusion was reached that dental asymmetry reflected the relative success that developmental homeostasis had in countering developmental disturbance. The earlier the insult, the greater the effect and the less successful was biological buffering.

Since early 20th century subjective work demonstrated facial asymmetry by comparing constructed faces of mirrored half faces from left and right sides, a common assumption has been that the midline of the face is synonymous with the plane of symmetry of the face. When assessing the effect of gender and age on facial asymmetry in 314 subjects from 12 to 56 years, the authors assumed that a symmetry plane constructed through the bisection of the binocular width and the nasal bridge was a valid reference point ((Ferrario et al. 2001). What was termed the left centre of gravity, the centre point of the left-sided facial landmarks, was reflected across the constructed plane and the distance between right and left centres of gravity was calculated and stated to be a measure of asymmetry. It has been claimed that this plane, perpendicular to, and bisecting, the landmarks exocanthion, can be used accurately in analysis of asymmetry (Ras et al. 1995).

2.2.2. Direct Anthropometry

The accepted gold standard of facial measurement is direct measurement of distances and angles between anthropometric landmarks using calipers and angle meters. Co-operation of the subject is imperative and the method of data acquisition is heavily operator dependent. Even the author whose results are most frequently quoted has admitted that there are many opportunities for introduction of error associated with direct anthropometry (Farkas 1996). The same points must be found repeatedly for various measures, and distortion of the soft tissue by calipers can alter distance measurements. Also, it cannot be applied to young subjects.

An extension of this direct measurement technique is the digitisation of landmarks on the human face using a non-contact digitising device such as that introduced by (Ferrario et al. 1998b). The latter method acquires 3D co-ordinates of the facial landmarks suitable for analysis beyond the remit of traditional morphometrics. The co-ordinate is recorded without skin contact avoiding the possibility of skin indentation and each landmark needs only single digitisation. Handheld digitisers have also been applied to the study of dental study models (Derijcke et al. 1994); (Mishima et al 1996); (Heidbuchel & Kuijpers-Jagtman 1997); (Prasad et al 2000); (Stellzig et al. 1999).

Direct anthropometry has been the source of many published population norms and forms the basis for validation of other measurement techniques (Aung et al 1995); (Bulstrode et al. 1986); (Farkas et al. 1980). One of the limitations of direct anthropometry, restricted descriptive statistical analysis, is also a reason for it's universal acceptance as a source of reference values. The simple mathematical models for analysis are universally available and the outcome of analysis is directly related to the data collected.

2.2.3. Indirect Anthropometry

Indirect anthropometry is the measurement of the human form on recorded images of the subject. It can be based on two, three or four-dimensional images with consequent complexity of analytical models and perhaps additional sources of error. It can also be based on measurements of facial casts or images of casts. The main advantages of indirect over direct anthropometry are faster data collection and the opportunity provided to create an archive of original data that can be re-examined. The main disadvantage of indirect anthropometry is the possible distortion of the dimensions of the subject. The degree of distortion depends on the type of imaging process employed.

2.2.3.1. Cast Studies

The analysis of dental casts will be dealt with later in the section on orthodontic assessment. Cast studies can be one or two steps removed from direct anthropometry, involving either direct measurement of casts of biological subjects or measurement of images of biological casts. A brief account of indirect anthropometry based on facial casts is included here.

The problem of distortion of shape and size with cast production was alluded to in a study designed to validate videoimaging of nasal morphology in UCLP (Russell et al. 2001). If the discrepancy between direct measurements on the face and on the cast differed by more than 10%, the impressions of the nose were recorded and casts prepared again. The high threshold for error in this study was acceptable, as what was required was a lifelike stimulus for video-based measurement. In an investigation of maxillary dental arch growth, combined dental arch and midface impressions were taken in anaesthetised children (Wada et al 1984). The dimensional accuracy of the cast preparation was not assessed but the authors might have defended the methodology by claiming that even if distortion had occurred, the midface to arch relationship would not have altered significantly. It was the midface to arch relationship that was under investigation.

2.2.3.2. Two-Dimensional Imaging

The techniques most frequently applied to facial and dental imaging are cephalometry (posteroanterior and lateral), photography, flat-bed laser scanning and video recording. The techniques are readily standardised but the dimensional accuracy can be poor and assessment of the three dimensional structure of the face using two dimensions is limited (Gross et al. 1996).

Cephalometry is standardised by placing the subject in a cephalostat to restrain the head position and positioning the radiation source at a fixed distance from the midpoint of the subject's head. Cephalometric analysis was one of the key assessments of cleft treatment outcome of the influential six centre European study (Molsted et al. 1992). It was used to describe the development of the skeleton and to assess the soft tissue profile. Assessment of the facial surface, using lateral cephalometry, is limited to the profile of the face and information about the soft tissues is incidental to data acquired about the facial skeleton.

The authors admitted that cephalometry was not as sensitive to variations in outcome between treatment centres as dental study model analysis. The radiation dosage of this method renders it unsuitable for ethical application to the normal population. For this reason, most archives of so-called normal cephalometric data are derived from routine radiography of patients receiving orthodontic treatment for malocclusion of dental or skeletal origin. The validity of population norms based on this study sample must be in doubt (Kyrkanides et al. 2000).

Photography as a clinical record and a basis for photogrammetry remains in wide use. The American Cleft Palate Association (ACPA) has recently adopted a photographic based outcome assessment as the standard technique for inclusion in the ACPA central registry, despite the acknowledged impossibility of rigorous standardisation (Hurwitz et al 1999); (Vegter & Hage 2000). A study which aimed to establish the maximum number of reliable measurements in the various areas of the face and head compared photogrammetry to direct measurement (Farkas et al 1980). Only 32.2% of the 104 measurements derived from plain photography were found to be accurate The least accurate measurements involved landmarks around the ear. A systematic bias was identified in measurements of photographed images when compared with direct caliper measurement (Shaner et al. 1998).

Photocopying of dental casts has been used to provide a 2D representation of the palatal casts of children at 14 weeks for comparison of cleft and control arch form (Huddart et al. 1978). This author was actively seeking to remove the perception of depth, which was held to complicate cast analysis. Photography has been applied to dental study models of non-cleft children also (Richey et al. 1995).

Flat-bed laser scanning is widely and cheaply available and has been used in analysis of dental study models and pre- and post-operative palatal study models. Without quantifying the distorting effect of distance from the scanning surface, scanned images of palatal casts prior to surgery were used to measure the size of the palatal cleft to investigate the correlation between severity of cleft and dental arch relationship at five years of age (Johnson et al. 2000a). Palatal blood flow, as a synonym for mucosal scarring, has been compared with arch form, using analysis of flat-bed laser scanned images of the occlusal surface (Ishikawa et al 1999). The shorter distance between the scanner and the occlusal surface would have given a more accurate representation of dental arch. There was, however, no attempt made to quantify the accuracy of the measurements.

Video recording is variously considered to be a 2, 3 and 4D imaging modality. Its most useful application in the field of OFC assessment is in speech analysis. A dynamic record of the face with an audio recording of the voice can be acquired simultaneously. It has been used for predicting soft tissue outcome of growth modification in skeletal malocclusions but it was acknowledged that the greatest value of video imaging was as a communication medium rather than a diagnostic tool for growing patients (Hoss et al. 1997). It has also been used to record the nasolabial area of children with UCLP which was then assessed by a panel of plastic surgeons, grading 9 features of the appearance and function of the upper lip and 10 features of the nose (Morrant & Shaw 1996). A wide range of panel reliability was demonstrated which was at odds with the authors' conclusion that this system would be useful for quality assurance and intercentre comparisons.

2.2.3.3. Three-Dimensional Imaging

Some three-dimensional imaging techniques are modifications or advancements of the methods mentioned in the last section such as radiography and photography. The common feature of three-dimensional imaging is an added data processing step to produce the final 3D image.

2.2.3.3.1. Three - Projection Cephalometry

This is a modification of the well established skeletal imaging method which allows comparison of 3D data with the extensive body of published norms

(Bhatia & Leighton 1993). Images are collected with the subject placed at three different angles to the radiation source and then the three projections are amalgamated to allow collection of 3D co-ordinates of landmarks of interest. A study of 30 infants with UCL found the precision of the technique was 0.8mm and 1.6 degrees for linear distances and angles respectively. The infants had to be sedated and data collected in this study was limited to the facial skeleton (Hermann et al. 2001).

2.2.3.3.2. CT Scanning

This method of imaging has recently been applied to dental study models to examine the neonatal palatal form and to compare the cleft maxillary arch form with control data (Darvann et al. 2001). The uniform density of the study model renders it suitable for radiographic scanning and the radiation dosage to the inanimate object is not of clinical consequence. Large batches of study models have been scanned which reduces the cost of data acquisition in time. Spiral or 3D CT scanning is also useful in the planning of craniofacial reconstruction when computer-aided machining of tissue replacements, such as segments of the cranial vault, provides a valuable aid to reconstructive surgery (Mole et al. 1995).

2.2.3.3.4. MRI Scanning

This is another imaging modality that can be modified to provide 3D surface data (Linney et al. 1993). Surface rendering of the varied digital output formats of CT scanning, MRI scanning and ultrasound has been developed which increases the diagnostic value of the original data. Surface data from CT scanning has also been combined with a photorealistic overlay to generate a virtual face as a basis for simulation of surgical outcome (Xia et al. 2000). This study was based on the flawed assumption that the soft tissue would respond directly to known movements of the underlying structure. This has been known to be an erroneous assumption for at least 40 years, when longitudinal cephalometric data was examined to map skeletal and soft tissue growth (Subtelny & Rochester 1959). However, just as the clinician must decide on the investigative method that is likely to yield the most information, the researcher in the field of shape analysis must choose the most appropriate method of data collection. The main advantage of CT scanning and MRI scanning is the ability to visualise tissue that would otherwise be inaccessible without invasive procedures, such as surgery. The additional yield of surface data is a bonus. If the focus of one's research question is surface shape, the research tool chosen should be primarily surface imaging.

2.2.3.3.5. Laser Scanning

A laser beam is fanned into a vertical line using a cylindrical lens, projected onto the patient's face and then viewed obliquely by a camera. This technique has been applied to dental casts and human faces and is probably most suitable for imaging of detailed inanimate objects. This surface scanning technique produces a 3D image which is texture free, limiting the number of anthropometric landmarks that can be identified (Hennessy & Moss 2001). Some landmarks on the face are identified at points of colour change rather than shape change and this requires a colour image of the face. A limitation of the technique is an inability to sample underneath overhangs but accuracy is good with errors as low as 0.05mm when applied to dental study models (Kuroda et al. 1996). It took 45 minutes to generate a 3D graphic of the model but a solid wax model can also be generated from the measurement data and a new tooth alignment could be computed for surgical planning. When applied to facial morphology, accuracy deteriorates. A comparison of direct facial measurement and measurement of laser scanned images on screen found only 33% of the latter measurements were within 1.5 mm of the former (Aung et al 1995). Facial scanning takes up to 10 seconds, which could encompass two breathing cycles with attendant unconscious facial movement. Even in a co-operative subject, intrinsic and extrinsic facial movement interfere with data collection. This technique has been applied to a small number of children as young as 7 years (Duffy et al. 2000).

Analysis and superimposition of multiple laser scanned images remain landmark based but the surface data has also been decomposed into regions of fundamental surface types e.g. saddle surface, ridges or grooves as a means of describing changes with surgery or growth (Coombes et al. 1991). Radial distances from the centre of rotation have also been compared in pre- and post-operative cases to assess soft tissue changes relative to skeletal changes (McCance et al. 1997b). The subject is rotated through the laser beam during imaging so the centre of rotation is a fixed location, provided the subject is seated in the same position and assumes the same pose at successive data collection sessions. It has been applied usefully in the characterisation of racial variation in nose shape with a view to maintaining appropriate ethnic characteristics when performing aesthetic rhinoplasty (Aung et al. 2000).

2.2.3.3.6. Moiré Stripes

This photographic based system uses moiré stripes generated from photography of the subject with a moiré camera. The stripes are automatically analysed to produce facial dimensions. Differences in the face appear as asymmetries in the Moiré stripes and are easily identified (Kawai et al 1990). This imaging technique has not been adopted widely, probably because it lacks the wider applicability of other structured light methods. Moiré stripes work well only where the shape of the structure is highly variable.

2.2.3.3.7. Stereophotogrammetry

In the early years of the 20th century, the mathematical foundations of analytical photogrammetry were established which led to the development of stereocomparators, optical instruments that allowed an operator to measure the parallax or disparity between two images (Urquhart 1997). Computational restrictions meant that from 1910 to 1955 most photogrammetric research turned to the development of opto-mechanical stereo plotting instruments called stereoplotters. These allowed maps to be plotted from stereopairs of photographs. The development of digital computers in the early 1950's, which could perform rapid calculations, lead to further development of analytical photogrammetry. The first analytical stereoplotter that used a digital computer was patented by Helava of the National Research Council of Canada in 1964. Abdel-Aziz and Karara developed direct linear transform (DLT) in 1972 which simplified the relationship between world and comparator co-ordinates. In the early 1980's digital photogrammetric instruments were developed which allowed measurement of image co-ordinates to be performed inside the computer rather than directly on photographs.

In 1967, facial contour mapping by photography was introduced to the medical literature (Burke & Beard 1967). Ultraviolet light was used as illumination. An early example of stereophotogrammetry applied to the study of the cleft face is a study of growth in monozygotic twins, one with bilateral cleft lip and one with unilateral cleft lip and palate who were examined over twelve years (Burke & Hughes 1987). A stereoplotter operated by a trained stereophotogrammetry technician was required to extract the 3D co-ordinates of the anthropometric landmarks.

It was over 15 years before computerised stereophotogrammetry was applied to analysis of facial morphology, with calibration of camera geometry described in a clinical study of a single subject undergoing facial osteotomy (Rasse et al. 1991). Use of computerised stereophotogrammetry to assess the results of face lifts reported a very large system error of 3.8 mm in the X axis, which, it was suggested, was a factor of large voxel size (Vannier et al. 1993). A published validation of the technique applied to facial growth in 59 subjects 5 years later, used a speckled pattern placed between the cameras and subject which appeared in all the monochrome images produced (Ras et al. 1996).

Full automation of the stereophotogrammetry process was described in 2000, when commercially available stereophotogrammetry equipment had become widely available for hospital use which did not require specialist operators (Siebert & Marshall 2000). Colour images of clinical subjects were also available as the images used to record the air-surface interface were supplemented by colour images without any light patterning. The colour images allowed more consistent identification of anthropometric landmarks. In digitisation of canine skulls, the reproducibility of computerised stereophotogrammetry was found to be comparable to caliper measurements and slightly superior to that of direct 3D digitisation (Stevens 1997).

Using a liquid crystal shutter, which generated coded light patterns for space encoding of the recorded image, rather than a random speckled light pattern, an initial attempt was made to automate landmark extraction (Yamada et al 1999). A study of the shape of the nostrils in cleft and control subjects used a mixture of manually located type I landmarks and automatically generated type II landmarks.

2.2.3.4. Four-Dimensional Imaging

Adding recording of time, the fourth dimension, to imaging permits dynamic assessment of mobile facial structures. At present, 4D imaging techniques are either very cumbersome or of low dimensional accuracy. Ultrasound, which has been applied to cardiovascular medicine as echocardiography for many years, requires obliteration of air spaces between the ultrasonic sensor and the tissue of interest. Studies of mimetic expressions to allow mapping of the dynamic orofacial morphology required the lower face to be submerged in a water bath with only nasal breathing possible (Deng et al. 2000). In this published study of just 4 subjects, one required training in breathing through her nose while keeping her lips under water so the high level of subject co-operation required would significantly limit applicability. Video tracking of mimetic expressions requires analysis of distortion of markers of known diameter placed on the skin to quantify the movement of the markers towards and away from the camera (Trotman et al. 1998a). Any technique requiring repeat placement of skin markers introduces systemic additional error to the imaging process.

In summary, “appropriate” and “accurate” have emerged as the key words in the field of morphometrics and anthropometry. No single combination of mathematical model and measurement technique is universally applicable. The accuracy of a measurement technique, in ideal circumstances, may not be possible to replicate in field studies, especially in potentially unco-operative subjects like children.

2.3. Results of Studies of Facial Morphology

The challenges facing the researcher in the field of facial morphology have been neatly summarised by an author whose work has been a reference point for the work of many others. In a discussion of the factors affecting accuracy and validity of anthropometric studies, whether cross-sectional or longitudinal, the closing comments listed criteriae that should be adhered to in the ideal methodology (Farkas 1996). A representative sample reflecting the ethnic composition and socio-economic profile of society should be used; the measuring skill of the examiner must be optimised as the reliability of the anthropometric norms depends on this factor; the subject must be willing to co-operate and a detailed analysis of all factors influencing the test results is necessary. Subject compliance is the factor that has limited research into paediatric facial morphology. Young children will not co-operate with direct anthropometry thus preventing the collection of large series of normative data. Indirect anthropometry reduces the level of co-operation required but the use of invasive techniques such as radiography or sedation is unacceptable where no individual clinical need exists.

A review of the literature pertaining to paediatric facial morphology demonstrated a preponderance of studies with small sample size and many childhood studies which did not examine young children. In the field of cleft-related facial morphology, there has been a lack of case control studies. The studies discussed are summarised in Table 2.1.

2.3.1. Case Control Studies

An investigation of the long term effects of alveolar bone grafting in adults, had a control sample of only 60 compared with a sample of 86 cleft affected individuals (Trotman et al. 1997). The benefits were limited to the immediate area of the cleft and not generalised to craniofacial growth.

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Ishiguro et al (1976)	To compare facial skeletal growth rates between cleft types	Prospective longitudinal	51 UCLP, 27 BCLP, 62 CPO at 3 months, 6 months and from 1 to 6 years	PA cephalograms	Direct linear measurement on films	Authors	BCLP cases had broader faces than UCLP and CPO cases Nasal and maxillary widths grew less in UCLP and BCLP cases There was no crossbite seen in CPO The effect of clefting was seen in the mandible
Burke & Hughes (1987)	To study growth in monozygotic twins, one with OFC	Prospective longitudinal	1 set of monozygotic twins, 1 with bilateral cleft lip and unilateral cleft palate	Serial stereophotogrammetry images from age 7 to 18 years	Linear measurement between facial landmarks and surface area estimation	Authors	The upper lip did not grow after age 11 in the affected twin Lip revision surgery increased the surface area of the lip
Mars & Houston (1990)	To investigate the effects of surgery on facial growth and morphology in Sri Lankan males	Retrospective	56 UCLP cases, (divided into no surgery / cheiloplasty / cheiloplasty and palatoplasty) 23 unaffected males (All over 13 years of age)	Cephalograms and dental study models	Cephalometric analysis and dental arch relationship analysis	Authors	Subjects who have had lip repair in early infancy show relatively normal maxillary growth Maxillary hypoplasia is common when the palate has been repaired early

Table 2.1. Summary of Studies of Facial Morphology at Rest

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Farkas et al (1992)	To study growth and development of regional units of the head and face	Prospective	Details of sample for younger children not published	Direct anthropometry of the facial surface	Linear dimensions	Authors	By 5 years the head width, length and circumference approached 100% maturity Facial maturity was reached at 12-15 in boys and 2 years earlier in girls The intercanthal width is mature in girls at 8 years and boys at 11 years By 5 years nasal and cutaneous lip dimensions approach maturity except for nasal protrusion
Ras et al (1994)	To analyse facial asymmetry in UCLP	Prospective	49 UCLP (aged 7.4 years) and 80 controls (aged 9.2 years)	Facial surface images acquired with stereophotogrammetry	Asymmetry measured relative to constructed reference plane, the bisection of the outer interorbital distance	Authors	There were gender related differences in asymmetry There was more asymmetry in the region of the cleft Males had more nasal asymmetry
Han et al (1995)	To analyse facial growth following cheiloplasty in cleft subjects	Retrospective	10 UCL, 33 UCLP, 14 CPO at 4 months, 4 and 8 years 33 Controls at age 8 years only	Serial PA and lateral cephalograms	Linear dimensional cephalometric analysis	Authors	Wide upper face in UCLP reduced after surgery but persisted at age 8 Less forward maxillary growth occurred after palatoplasty There was mandibular compensation in cases of UCLP and CPO

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
McCance et al (1997)	To analyse soft tissue morphology in cleft subjects after Le Fort I advancement	Prospective	13 UCLP, 6 BCLP and 5 CPO cases (mean age 20)	Laser scanned facial surface images	Linear dimensions and colour coded whole face superimpositions	Authors	Surgery did not correct the nasal retrusion The nasal complex should be accepted or higher level surgery should be performed
McCance et al (1997)	To analyse soft tissue to bone movement ratios following Le Fort I advancement	Prospective	13 UCLP, 6 BCLP and 5 CPO cases (mean age 20)	Laser scanned facial surface images CT scanned images of the facial skeleton	Linear dimensions and superimposition of the images	Authors	There was a 1.25 soft tissue to bone movement ratio in the midline and chin There were no differences in the soft tissue to bone ratios between the cleft and non-cleft sides of the face
Ras et al (1997)	To investigate left right differences in the sagittal position of the maxillary segments in children with CLP	Prospective	16 CL cases,27 UCLP cases and 17 BCLP cases (mean age 9 years)	CT scans of the facial skeleton	Linear dimensions	Authors	UCL and UCLP cases did not have significantly different sagittal maxillary segment findings

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Trotman et al (1997)	To analyse the effect of alveolar bone grafting on craniofacial growth	Retrospective	85 adult UCL cases (58 had not received bone grafts) 60 controls	PA cephalograms	Linear dimensional	Authors	The long term effects of alveolar bone grafting on craniofacial growth are minimal and limited to the immediate area of the cleft
Ferrario et al (1998)	To examine longitudinal changes in facial regional volumes	Prospective mixed cross sectional		Stereo videoimages of the facial surface	Measurement of facial regions as tetrahedrons	Authors	
Leonard et al (1998)	To compare soft tissue and skeletal form of Northern Irish children with published results for 6 European centres	Retrospective	25 UCLP cases (mean age 9.4 years)	Dental study models and cephalograms	GOSLON Yardstick and cephalometric analysis	Authors	Dental arch relationships were acceptable There were no skeletal differences but the soft tissue facial profile was more convex than in the best European centre
Feragen et al (1999)	To establish if right cleft disfigurement is due to physiognomic asymmetry or perceptual	Prospective	160 children with UCL, UCLP, BCLP or CPO (aged 6 or 16)	Projected photographic images of faces, normal and mirror reversed.	Rating of disfigurement on visual analogue scale	37 psychology students	The differences between right and left clefts are due to facial factors rather than perceptual processes

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Mauli et al (1999)	To determine the effect of presurgical nasoalveolar moulding on 3D nasal shape in unilateral clefts	Retrospective	10 UCLP cases (aged 4.5. years) who had received nasoalveolar moulding 10 UCLP cases (aged 9 years) who had not received moulding	Images of nasal casts scanned with structured light imaging system	Global nasal asymmetry scoring	Authors	There was a significant reduction in nasal asymmetry with nasal moulding maintained into early childhood
Duffy et al (2000)	To identify and assess differences in facial soft tissue morphology in cleft children	Prospective	39 CLP subjects 25 non-cleft orthodontic patients (aged 8 - 11)	Laser scanned images of the facial surface	Linear dimensions	Authors	Significant differences existed between cleft and non cleft subjects The cleft face was narrower with a deeper submandibular area
Heliovaara et al (2000)	To examine the soft tissue response following Le Fort I osteotomy in cleft subjects	Retrospective	25 males and 13 females with UCLP, mean age 23.5	PA and lateral cephalograms	Linear dimensions	Authors	Vertical changes were greater than anteroposterior changes Vertical changes were more obvious if V-Y plasty was performed There were post operative mandibular changes following maxillary surgery
Hermann et al (2000)	To analyse craniofacial morphology in infants with UCLP with reference to a control group with UCL	Prospective	55 UCLP and 53 UCL cases at 22 months	Three-projection cephalograms	Linear dimensions	Authors	Interorbital distances were the same in both groups The nasal cavity was increased in the UCLP cases The dental arch was wider posteriorly and narrower anteriorly There had been no consistent response to palatoplasty

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Kyrkanides et al (2000)	To examine the development of cranial base asymmetries in subjects with UCLP	Retrospective	30 UCLP cases 64 control cases (orthodontic patients) Grouped into 6-10, 11-14 and 15-16 years	PA cephalograms	Linear measurement	Authors	Lower face vertical asymmetry correlated with vertical maxillary dentoalveolar measurements and not cranial base There was insignificant cranial base asymmetry in UCLP cases relative to the control sample
Ferrario et al (2001)	To assess the effects of age and sex on 3D soft tissue asymmetry	Prospective cross sectional	314 subjects from 12 to 56 years	Direct digitisation of the facial surface	Linear measurement from midline to constructed "centre of gravity" for each side of the face	Authors	Females and adolescents are more asymmetric No dominance of either side of face noted
Yamada et al (2002)	To analyse 3D facial form of normal children	Prospective Mixed cross sectional	97 children at 3 months 54 children at 18 months 80 children at 3.5 years 37 adults	Images acquired with liquid crystal range finder	Semi-automated landmark extraction and interlandmark distance measurement	Authors	There are few gender differences in young children's faces Facial dimensions not at full maturity at 3.5 years
Yamada et al (2002)	To evaluate morphological problems for children with UCLP	Prospective Mixed cross sectional	8 children at 3 months 18 children at 18 months 20 children at 4 years All with UCLP	Images acquired with liquid crystal range finder	Semi-automated landmark extraction and interlandmark distance measurement	Authors	Wider intercanthal width, nose and mouth width in UCLP cases at 3 and 18 months - within normal at 4 years Deviated columella, alar expansion, flattened nasal tip and widened nasal tip angle were not within normal range at 4 years

As subjects requiring orthodontic assessment routinely require cephalometry, these have been a source of control data for many researchers but the requirement for a representative sample of the population is not fulfilled if the subjects who act as controls are limited to those requiring treatment for malocclusion. Orthodontic assessment is rarely performed in the primary dentition so control data of this variety is lacking in the infant and young child.

A commonly encountered lower age limit of 6-7 years was found in a study of the development of cranial base asymmetries in patients with UCLP (Kyrkanides et al 2000). No difference in cranial base asymmetry was found between the two groups, the lower face vertical asymmetry correlated only with the vertical maxillary dentoalveolar measurements while horizontal asymmetry developed in parallel with the mandibular fossae of the temporal bone. This study investigated twice as many controls as cleft cases but often control samples were small.

Another study of asymmetry, but of soft tissue facial asymmetry, in older children, aged 7.4 to 9.2 years, using stereophotogrammetry, found that asymmetry in children with UCLP was predominantly vertical rather than horizontal across a midline plane (Ras et al. 1994). The asymmetry was also found to be concentrated in the immediate area of the cleft.

A study applying three projection cephalometry to infants aged 22 months used UCL subjects as the control group compared with children with complete UCLP (Hermann et al. 2000). This meant the children were matched for cheiloplasty with palatal clefting as the main significant variable when examining craniofacial morphology. The inner and outer interorbital distances were the same with slight retrusion of the lateral orbital margin in the UCLP group. Deviation of the anterior nasal spine towards the non cleft side was present, and no consistent response to surgery was noted, with traits reported to have improved, worsened or stayed the same since the pre-operative period.

A preliminary report on three-dimensional analysis of the facial soft tissue using laser scanning in bilateral cleft lip and palate (BCLP), UCLP, UCL and CPO and control subjects aged 8 –11 years (Duffy et al 2000). Narrower intercanthal widths were found in the cleft subjects. Analysis was limited to comparison of interlandmark distances and a qualitative comparison of overall face shape. The effect of altering lip pose was not investigated although the authors commented on the presence of lip incompetence in the images captured. In the developed world the age of pubertal onset is dropping and it may be that some of the children examined in this study had commenced a prepubertal growth spurt. The pairwise comparison between the five types of subjects was incompletely reported.

One serial case-control growth study that managed to follow the subjects for almost 11 years had only 2 subjects, monozygotic twins, 1 with bilateral cleft lip and 1 with unilateral cleft palate (Burke & Hughes 1987). Monozygotic twins are the ideal control subjects, but while relatively large series can be enrolled in studies of common conditions such as alcoholism or schizophrenia, the rarity of OFC means that twin studies are not feasible.

Another common motif of cleft / control studies is incomplete control data with control data available only for older subjects. This gives a mixture of cross sectional and longitudinal data, as in a study using cephalometry in 10 UCL, 33 UCLP and 14 CPO subjects compared with 33 controls (Han et al 1995). The affected individuals were radiographed at 4 months or 2 years and at 4 years and 8 years but the control subjects were radiographed only at 8 years. Less forward maxillary growth was seen in cases of palatoplasty. As control data were unavailable for the earlier years, it could not be determined when the growth deficiency relative to normal was first manifest.

There is a heavy reliance evident on the "normal-for-clefts" data such as the results of multi-centre studies, which are taken as benchmarks for surgical practice. In countries with small populations such as Northern Ireland, comparisons with North Western European centres of similar ethnic background can be useful (Leonard et al. 1998). The use of such benchmarks in distant centres should be more circumspect (Johnson et al. 2000b). Comparison of the outcome of cleft surgery in Western Australia with that in the United Kingdom suggests that individual with clefts are homogeneous to an extent that transcends all geographic limits, which has not been proven.

Two recent linked publications with an acceptable case-control model employed a liquid crystal range finder to collect data on the soft tissue facial morphology (Yamada et al. 2002a). Cross sectional data was collected from 97 four-month-old infants, 54 eighteen-month-old infants and 80 three-and-a-half-year old children together with 37 adults and 1 child with UCLP aged 4 years. No comment was made on the standardisation of the facial expression or lip pose. This is of note as the faces were scanned twice to give full facial coverage. The photograph of the UCLP child provided showed a marked lips apart pose. The landmarks were extracted semi automatically and subjected to traditional morphometric analysis of distances and angles. Infant facial proportions persisted to age 3.5 years with a proportionally greater upper face width and lesser middle and lower face widths than in the adult subjects. Only minor gender differences were noted. The one cleft child examined had facial measurements within the range of normal except for enlarged nose height and nostril length and reduced right alar length.

The related study of facial morphology in UCLP was also a mixed cross - sectional study with 8 four-month-old infants, 18 eighteen-month-old infants and 20 four-year-old children. The same imaging and analytical techniques were employed and the results compared with the published findings in slightly older normal children. At 4 and 18 months the UCLP subjects had wider intercanthal widths which were within normal range at 4 years. The nasal width was greater, the nasal tip was flattened and the base of the columella was deviated towards the non cleft side. These differences persisted at age 4 years.

Description of asymmetry was limited to a comparison of paired linear dimensions and nostril angulations and showed vertical asymmetry of the superior and inferior nostril points at all time points. It was suggested that functional forces may have normalised the deformities following reconstruction of the orbicularis oris. The alar asymmetry was attributed to a poor volume of alar tissue including cartilage but there was no evidence offered for this assumption.

Persistence of nasal deformity, despite surgical intervention, was replicated in an investigation of older subjects using laser scanning (McCance et al 1997a). Comparing controls with UCLP, BCLP and CPO subjects following Le Fort I osteotomy advancement, at age 20 years, the dorsum and cleft side of the nose were still retrusive. The authors concluded that the threshold for deciding on corrective surgery should be higher, or the level of Le Fort osteotomy performed should be higher. Twenty years is a very long time to wait for results of surgical outcome. It would appear that there is a consensus developing that cleft related nasal deformity can be assessed quite early as it does not change with growth.

This would suggest that the positive preliminary results from a study of nasoalveolar moulding on three-dimensional nasal shape in unilateral clefts are likely to be maintained (Maull et al. 1999). This study was limited to cleft subjects but used those who had not received pre-surgical moulding treatment as the control group.

2.3.2. Studies of Cleft-Affected Study Samples

There is a general consensus in the literature on the growth-retarding effects of early reconstructive surgery on the facial skeleton. Examination of skeletal morphology in adults with unoperated OFC shows that the wider facial form noted at birth persists into adulthood. In Sri Lankan adults, a larger overjet was noted in unoperated UCLP subjects (McCance et al. 1990). Confirming the strong link between growth retardation and surgery, again in the Sri Lankan population, a larger than normal SNA angle was found in lateral cephalograms of unoperated UCLP subjects (Mars & Houston 1990). A smaller angle was measured in subjects who had had cheiloplasty and a markedly smaller angle in those who received both cheiloplasty and palatoplasty (Mars & Houston 1990).

A study, covering the years when the highest rate of facial development occurs, examined UCLP, BCLP and CPO subjects using posteroanterior cephalometry at 0-3 months, 4-6 months and annually from 1 to 6 years (Ishiguro et al 1976). A broader face was seen in the BCLP group at 6 years while the UCLP and CPO groups were closer to normal Bolton standards. At 1 year of age, the nasal and maxillary widths were greater in BCLP and UCLP than in CPO but thereafter the rate of growth was very low in the former two groups while the CPO widths continued to grow. At 6 years of age, all marked skeletal structural changes had disappeared showing the effects of surgical closure. Slight crossbites were found in the UCLP and BCLP groups, perhaps suggesting differing rates of transverse maxillary growth and mandibular growth. The landmarks of the nasal aperture, maxilla and dental arch showed a slight medial displacement on the affected side.

Another radiographic study of UCL, UCLP and BCLP subjects at the older age of 9 years found that the sagittal maxillary segment positions in UCL and UCLP did not differ significantly (Ras et al. 1997). The trend identified in the preceding study by Ishiguro et al (1976) might have suggested that the combined effects of palatoplasty and cheiloplasty would have lead to a discrepancy between the sagittal maxillary segment positions in groups with and without palatoplasty.

Several studies of soft and hard tissue responses to osteotomy in adulthood have highlighted the indirect relationship between both tissue types. Combining CT and laser scanning to study perioperative changes with Le Fort I osteotomies in UCLP subjects , mean age 20 years, a 1.25 ratio of soft tissue to bone movement was noted in the midline and chin and canine regions following surgery (McCance et al 1997b). There was no difference between the cleft and non-cleft sides of the face.

A similar study highlighted the need for early assessment of primary surgery before the confounding variables of secondary procedures cloud assessment of outcome (Heliovaara et al. 2000). The aim had been to examine the soft tissue response to Le Fort I osteotomies in 25 males and 13 females, aged on average 23.5 years, with UCLP who were divided into groups according to whether or not V-Y cheiloplasty was performed with the maxillary advancement. There was no clearly stated indication for V-Y plasty; several subjects had further surgery between osteotomy and cheiloplasty and the date of outcome assessment and the mounting variable count made it difficult to draw conclusions. Facial soft tissue responded as a single unit to skeletal modification, with post-operative alteration in the soft tissue dimensions over the mandible where no surgery had been performed.

Most studies of unilateral clefting reverse images of data collected from subjects with right sided clefts so that all clefts appear on the, more commonly affected, left side. It may be that this is an overly simple approach to right / left clefting according to the findings of a study of the perception of cleft related disfigurement (Feragen et al. 1999). The aim had been to establish if the circumstantial evidence that right clefts are more disfiguring than left clefts was related to perceptual processes in face recognition or physiognomic asymmetries. Images of faces of subjects with UCL, UCLP, BCLP and CPO, in normal and mirror reversed projection, were shown to a panel of psychology students that rated them on a visual analogue scale. Left-sided clefts were found to be less disfiguring. It was concluded that inherent facial factors accounted for the preferred status of left side clefts but these factors were not identified.

2.3.4. Normative Data on Soft Tissue Facial Morphology

Two research groups can be credited with significant contributions to the normative data on soft tissue facial morphology. Farkas (1996) in Canada who relies on direct anthropometry and Ferrario et al (2001) in Italy who use either handheld 3D digitisers to locate anthropometric landmarks or photography of face with adhesive markers on the skin. However the studies of both groups have been limited to older children and young adults and both use relatively simple methods of analysis to describe the complex soft tissue facial morphology.

Farkas et al (1992) have claimed that children of 4-5 years will co-operate with direct anthropometry and repeated facial measurements for up to 30 minutes. In a study of growth and development of the regional units of the head and face between 1 and 18 years, the data collected for children under 6 years was not published, with no explanation offered for this decision (Farkas et al 1992). The conclusions, however, based on this data were published. An examination of the components of the study sample highlights the difficulties in achieving co-operation in the younger subject. Those under 4 years of age were only 10% of the study sample, yet represented 22% of the time span under investigation. By 5 years the head width, length and circumference closely approached maturity. At this age, 83% of the mature overall face height was achieved with 88% of maturity achieved in the lower third by 5 years. The intercanthal width and binocular width were at 93% and 88% of maturity by age 5 years. The upper lip was at maturity by 3 years in girls and 6 years in boys but the nasal maturity was delayed relative to other regions of the face.

3D facial morphology in significant numbers of healthy Italian children and young adults have been addressed (Ferrario et al. 1998c). Either statistical analysis limited to the tools of traditional morphometrics or very simple representations of facial regions were used to assess 3D changes with growth. The results reported were in agreement with Farkas et al (1992) with facial dimensions reaching full maturity at 13-14 in girls and 15-18 in boys. Due to the mixed longitudinal study model, some of the values recorded appeared to drop with growth, an inherent problem, not seen in true longitudinal studies. The 3DFM™ method employed by Ferrario et al records the co-ordinates of adhesive markers placed on the face rather than anthropometric landmarks. There is a significant error associated with repeat placement of such skin markers. Two millimetres has been recorded for inter-operator error and intra-operator error has not been published (Ferrario et al 1996).

2.4. Facial Function

The normal anatomy of the face consists of five, bilaterally symmetrical, facial muscle slings, perinasal, perioral, oro-mandibular, zygomaticomaxillary -oro-zygomaticomaxillary and buccopharyngeal - oro - buccopharyngeal. In clefting of the muscles slings, their symmetry and power vectors are disturbed. Cheiloplasty aims to restore anatomical continuity and, by extension, correct these disturbed power vectors. The importance of a lip repair that restores normal expressive function is appreciated and a so-called functional lip repair exists (Adcock & Markus 1997). Most assessments of soft tissue reconstruction in OFC are limited to examination of the face at rest. There is little evidence in the literature of dynamic tests of the repaired cleft face and very few reports of research into facial function in children.

So, the number of studies of facial function in young children with OFC is very small. No studies published in English have been found. A review of the methods of facial function explains why paediatric studies are limited. Objective investigative methods usually require high levels of co-operation, long attention spans and may require advanced receptive communication skills which are not generally afforded by the paediatric population.

The large body of work dealing with facial expression related to emotional state will not be discussed. The mainstay of studies of facial function is maximal expression, such as effortful smile. These are gross simplifications of the expressive variability of the human face but standardisation of facial function is a recurring challenge for researchers in this field. Plastic surgeons dealing with facial paralysis and orthodontists have made the largest contribution to this area of research. Much of the work of the latter group is concerned with the aesthetic value of the smile and how it may be affected by orthodontic treatment.

2.4.1. Methods of Assessing Facial Function

2.4.1.1. Systematic Observation

The Facial Action Code (FAC) is a system devised to describe the components of facial expression based on muscle actions (Ekman & Friesen 1976). The authors claim to have taught themselves to isolate the action of single muscles of facial expression, building on the earlier work of an anatomist who trained himself to isolate muscles by mimicking the effect of electrical stimulation of named muscles. Using this skill, the authors described a range of facial expressions in terms of single action units (AUs) which was intended to be globally applicable to all studies of visible facial movement. While the system can be taught to others, few have replicated the process and instead rely on the work by Ekman and Friesen (1976) as a point of reference.

An example of a study relying on the work of Ekman and Friesen (1976) was carried out in 1994, which aimed to implement a method to use facial expressiveness in diagnostic assessment (Nafziger 1994). Subjects were recorded, using video imaging, creating expressions to match emotional experiences such as anger and surprise which the researchers analysed using the FAC describing expressions as AU histograms. Such a laborious and flawed approach to a very complex research question demonstrates the challenges of research in facial function.

2.4.1.2. Direct Facial Anthropometry

Direct measurement of the face during the performance of maximal expressions is often combined with other methods of assessment such as electromyography (Burres 1985). In a study designed to quantify facial motor function during mimetic expressions, the distance between landmarks placed on the skin with a grease pen were measured as 30 adult subjects, with no facial pathology, performed mimetic expressions with stepped increases in effort. The percentage of displacement of points was compared with the simultaneously acquired electromyographic readings. A logarithmic relationship was found between muscle electrical activity and skin motion. It was also found that distances from a variety of expressions were stable rather than a single expression or single interlandmark distance.

A patented sliding caliper which recorded two sets of 3D co-ordinates simultaneously and generated the linear distance between those points called a Faciometer™ was introduced in a study which also employed a videoimaging system to compare static and dynamic assessment of facial function (Frey et al. 1994). The data collection process took one hour to complete. It was reported that the only static points on the face were the tragii and the mid dorsum of the nose with synkinetic movement of the eyelids occurring in all expressions. Maximal teeth showing and lip purse were reproducible excursions but submaximal expressions were only suitable for intraindividual comparisons.

Other authors, using direct anthropometry, would disagree with the notion of lip purse being reproducible (Peck et al. 1992). In their study of the upper lip to tooth to jaw relativity in the vertical dimension, the range of expressions was limited to rest and maximal smile, maximal smile after coaching, not a spontaneous effortful smile. Descriptive statistics were used to analyse the linear distances measured which detected sexual dimorphism in the study sample of 42 males and 46 females with a mean age of 15 years. The same authors, who are orthodontists, studied the appearance of a high smile line, a gingival smile line (GSL), at maximum effort using direct anthropometry. GSL is one of the features of a smile that is considered by some, including patients seeking correction, to be unattractive. Another study claimed that the flattened smile, that may occur with orthodontic treatment, is considered unattractive (Hulsey 1970).

In an investigation of the range of mimic movements after surgical treatment of burns, direct anthropometry using a Faciometer ™ was combined with application of the Vancouver scar scale (Koller et al. 2000). The more mobile areas of the face such as the perioral region showed the greatest functional deficit even when the whole facial surface had been burned and repaired with skin grafting and keratinocyte application.

2.4.1.3. Photography

Photography, including video imaging, has been used for quantitative and qualitative investigation of facial function. A seminal piece of work employing plain photography that has become a reference point for other researchers was the development of the Maximal Static Response Assay (MSRA) of facial nerve function (Johnson et al. 1994). Subjects with, and without, facial movement abnormalities were photographed, with adhesive dots and a rule graduated in centimetres attached to the face, performing maximal expressions. The photographs of the maximal expressions were projected onto a digitiser board and adjusted until 1 cm on the rule photographed on the face and 1 cm on the board were equivalent. The movement from rest of each landmark was measured in 2D and presented as a profile of normal movement for each expression. The advantage claimed for this system was that it characterised global facial change.

Videoimaging was used in a project which aimed to summarise the MSRA into a numerical index for use in tracking recovery from unilateral facial paralysis (Bajaj-Luthra et al. 1997). The ratio of movement of the normal side of the face to the movement on the paralysed side was calculated by summing the vectors of movement of infra-, supra- and modiolar movements during maximal expressions.

One project which concluded objective assessment was outstripped by a subjective index like the House Brackmann scale of facial nerve weakness, was based on microscaling of video images to examine test, retest, side-to-side and day-to-day variability in two facial movements in normal subjects (Wood et al. 1994).

2.4.1.4. Laser Scanning

Surface scanning allowed changes in linear distances between landmarks during action relative to distances at rest to be measured in 3D space (Cacou et al. 1997). The subjects' heads were restrained and they were required to hold end points of maximal expression for 15 seconds to allow scanning to be completed. Landmarks were placed on the face over the major muscle groups but large variation in landmark placement was noted at successive visits. Automated facial landmark tracking would improve this sort of assessment by removing the error associated with subjective landmark placement. Such automation, which has been described in videotaped images but not surface scanned images, would greatly advance the capabilities of 3D imaging (Wachtman et al. 2001).

2.4.1.5. Four-Dimensional Ultrasound

The limitations of this cumbersome technique, requiring immersion of the subject in water for the collection of data, have already been outlined, although the added dimension of time introduces the possibility of dynamic studies incorporating the facial surface and the muscle layers (Deng et al 2000).

2.4.2. Studies of Facial Function in Children and / or Subjects with Orofacial Clefting

The studies discussed in this section are summarised in Table 2.2. One of the very few studies of facial function in younger subjects was the combined use of lateral cephalograms and electromyographic profiling of the superior orbicularis oris muscle at rest and swallowing (Carvajal et al. 1994). This was conducted in 13 children, median age 11 years, with UCLP and abnormal lip seal to assess the benefit of a removable appliance designed to eliminate the restrictive effect of the upper lip. To investigate the results of bilateral segmental gracilis muscles transplantation to the face as a treatment for Mobius syndrome, video images of 10 children, average age 7.5 years,

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Offerman et al (1964)	To determine which side of the mouth had greater mobility in subjects with repaired cleft lip	Prospective	24 UCL cases and 22 control cases aged 7 - 15	Cine camera images	Distance from midline	Author	UCLP cases had greater lip asymmetry at rest and in function The affected side of the mouth had greater mobility in the UCLP cases
Susami et al (1993)	To evaluate the shape and elasticity of repaired cleft lip quantitatively	Prospective	41 subjects with CL(P) 54 control cases Aged 7 - 15 years	Subjects with weighted lip extension device in situ	Linear measurement between the arms of the extension device Digital caliper measurements of linear lip dimensions	Authors	CL(P) patients had lower lip elasticity than controls, more marked in bilateral than unilateral clefting. CL(P) patients had shorter upper lip height.
Carvajal et al 1994	To assess the effect of a removable appliance designed to eliminate the restrictive effect of the upper lip	Prospective longitudinal	13 children with UCLP and abnormal lip seal (median age 11 years)	Lateral cephalograms. Electromyographic (EMG) profiles of the superior orbicularis oris muscle at rest and swallowing (pouting)	Cephalometric analysis Comparison of pre and post treatment EMG profiles	Authors	Improvement in the sagittal maxillary and dentoalveolar positions following fifteen months of continuous wear of the URA. Elimination of the restrictive effect of the superior orbicularis oris muscle may promote normal growth potential in children with UCLP
Trotman et al (1996)	To compare the amplitude of facial motion obtained using 2D and 3D methods	Prospective	4 adults with landmarks placed on face	Video images of the face during five maximal expressions	Amplitude of motion in 2D and 3D	Authors	3D amplitudes were significantly larger than 2D amplitudes, especially during smile animation.

Table 2.2. Summary of Studies of Facial Animation In Children and / or Cleft Subjects

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Trotman et al(1998)	To determine which facial landmarks show the greatest movement during animation To determine the sensitivity of stereo videoimaging to landmark displacement in animation	Prospective	5 adults with no facial pathology 6 adults with facial pathology (4 with repaired UCLP)	Stereo video images of the subjects wearing adhesive facial markers and head bands as stable reference points	3D displacement of skin markers from resting position	Authors	3D video tracking has the potential to detect and characterise clinically significant defects Upper face markers moved most in eye opening and closing Lower face markers moved most with smile and cheek puff
Trotman et al (2000)	To explore a mathematical model for landmark superimposition To compare adjusted data between cleft and control cases	Prospective	4 UCLP cases (14 - 16 years) and 5 BCLP cases (13 - 21 years) 50 control cases (mean age 27.3)	Stereo video images of resting face and maximal mimetic expressions No headbands in situ	3D displacement of skin markers from resting position	Authors	Markers on maxillary splint or headband unnecessary as reference points Greater within and among subject variability in cleft subjects Greater asymmetry in cleft cases in maximal expression
Zuker et al 2000	To investigate the results of bilateral, segmental gracilis muscle transplantation to the face	Prospective	10 children with Mobius syndrome average age 7.5 years	Video images of resting face and full smile	Excursion of oral commissure in 2D in millimetres	Authors	Segmental gracilis muscle transplantation innervated by the motor nerve to the masseter is an effective method of treating patients with Mobius syndrome.

performing a "full smile" were recorded (Zuker et al. 2000). The excursion of the commissures in 2D was measured in millimetres and the results following surgery supported the transplantation described as a valid treatment option. There was no description of how the "full smile" was defined. Horizontal commissure excursion is a very crude assay of perioral function. The vector of a smile differs from simple lateral displacement of the commissures and the magnitude of excursion in 3D is far greater than that detected using 2D measurement (Paletz et al. 1994); (Gross et al 1996)

In 1964, a study which aimed to determine which side of the mouth had greater movement in subjects with UCL was conducted (Offerman et al. 1964). Cine camera images of 22 UCL patients and 22 control cases, aged 7 to 15 years, were recorded during maximal smile, plosive production and sibilant articulation. The method of maximal smile acquisition was not described. Symmetry was crudely scored based on distance from the midline at the point of maximal displacement. There was greater lip asymmetry in the lips of the UCL patients at rest and this became more obvious in function. The affected side moved more and the authors' speculated that the unaffected side was drawn to the midline, synkinetic rather than anatomic movement.

A series of papers publishing the results of studies of tracking of facial movements using video cameras and interactive facial markers, lead up to a specific study of facial function in UCLP and BCLP in teenagers (Trotman et al. 1996); (Trotman et al. 1998b); (Trotman et al 2000). Maximal movement in 3D in a range of maximal facial expressions was compared on a case by case basis for the cleft subjects with the control sample results. In agreement with Offerman (1964), even greater asymmetry was noted in the cleft subjects during maximal expression. There was greater variability of expression within and among the cleft subjects but these subjects were significantly younger than the control subjects.

In a study of 41 subjects with CL(P) and 54 control cases, aged 7 to 15 years, the lips with a weighted extension device in situ were photographed at rest and maximal mouth opening (Susami et al. 1993). The aim was to evaluate the shape and elasticity of the repaired cleft lip. Cleft subjects were noted to have lower lip elasticity than controls, more marked in the bilateral than unilateral clefting.

2.5. Dental Arch Analysis

2.5.1. Study Models

The use of study models in orthodontic management of cleft and non-cleft children is well accepted, yet no papers appear to discuss, in isolation, the study model. Impression taking and bite registration are relatively easily acquired clinical skills. The use of impression materials and trays according to manufacturers' instructions and the pouring of the cast by an experienced technician produce a standardised, robust and objective record that does not degrade with time, if correctly handled. Sequential records of the dental arches are valuable in assessing changes with growth and development (Johnson et al 2000a). The challenges of maintaining an adequate archive of dental casts for clinical, research and medico-legal purposes and alternatives to the study model have been discussed. It has been calculated that for a British unit which sees 100 new patients each year, 17 m^3 of storage space are required to retain study models for the period recommended by the British Association of Orthodontists (McGuinness & Stephens 1993). A wide range of hospital policies for storage times, with little cognisance of medicolegal requirements, has also been recorded (McGuinness & Stephens 1992). The British Orthodontic Society recommends that study models for children with cleft lip and/or palate should be retained for 10 years from the date of completion of treatment or until the 26th birthday (British Orthodontic Society 1999). There are no more recent recommendations of the British Orthodontic Society on this subject on record.

2.5.2. Impression Techniques and Materials

Impression technique in children with OFC has not been written about extensively other than to document and suggest methods of minimising airway risk. In a technical note describing a technique of impression taking to protect the nasal airway, the addition of cottonoid patties to the tray was recommended but no other recent papers address this issue (Zarrinnia et al. 1993).

The authors of all the cast studies discussed either used alginate, irreversible hydrocolloid, or elastomeric material to record impressions or did not mention the impression material used.

Newer impression materials such as polyvinylsiloxanes are more expensive but, as well as being easier to work with, are well tolerated by young children due to texture and colour, more accurate and dimensionally stable indefinitely. Low density preparations are suitable for recording of soft tissue detail but higher density putties are well tolerated by the young child for recording of occlusal surface detail (Barghi & Ontiveros 1999).

2.5.3. Schedule of Impression Recording

In management centres where pre-surgical orthopaedics is performed or passive feeding plates are provided, neonatal palatal casts are clinically indicated. In the neonate with cleft lip and palate, several untoward respiratory events have been reported during impression taking (Ash & Croft 1989); (Chate 1995). However, in children with repaired complete clefts of lip and palate, the primary occlusion has been recorded adequately at 5 years (Robertson & Fish 1975). In the anaesthetised child, at times of surgery, complete palatal impressions can be acquired without additional risk to the child, whose airway is maintained by endotracheal intubation and protected by cuffing and packing. As it would be unacceptable to record impressions in unaffected neonates and professionally negligent to anaesthetise such children to acquire impressions, there has tended to be a deficiency of control data in studies of the neonatal cleft palate. The need for general anaesthesia to acquire combined midface and maxillary arch impressions accounted for the disparity between number of control observations (10), and study group observations, (120) in a study of the effect of surgery (Wada et al 1984).

In the individual patient with cleft lip and palate, sequential recording of dental arch form and occlusal relationship assists management, maps growth and monitors orthodontic treatment outcome. Routine data collection at 5 years of age was discussed earlier.

2.5.4. Study Model Analysis

2.5.4.1. Direct and Indirect Data Acquisition

Interlandmark measurement with sliding digital calipers has been the cornerstone of arch analysis, being accurate, readily available, inexpensive and reproducible. This technique is being extended and superseded by the use of handheld or microscope digitisers which allow the location of landmarks in three dimensional space as employed in the following selection of studies from the last decade - (Derijcke et al 1994); (Mishima et al 1996); (Heidbuchel & Kuijpers-Jagtman 1997); (Prasad et al 2000); (Stellzig et al 1999).

Rather than rely on direct contact digitisation, indirect measurement techniques have been developed. Digital photography of study models has been applied to assess the effect of growth hormone supplementation on dental arch development, (Richey et al 1995). Flat-bed laser scanning of study models has also been described. Questions about the validity of this technique, applied to uneven occlusal surfaces or the palatal cleft area, have already been raised in the earlier section on anthropometry (Ishikawa et al 1999); (Johnson et al 2000a).

Three-dimensional imaging systems were described initially for archiving study casts and assessing arch relationships (Keating et al. 1984); (Harradine et al. 1990); (Ayoub et al. 1996). Holographic views showed a tendency to underscore the severity of the malocclusion as opposed to study models (McGuinness & Stephens 1993). The accuracy of computerised stereophotogrammetry, a field imaging technique, applied to study models was not measured but estimated to be 0.2mm based on validation of the imaging technique in other clinical models with larger and far less complex surfaces (Ayoub et al 1996).

Surface scanning techniques, such as laser scanning, provide better subjective and objective results but the technique is time consuming and availability is limited by the high cost of customised dental imaging kits (Kuroda et al 1996); (Sohmura et al. 2000). Image acquisition times of up to 45 minutes per cast have been reported.

CT scanning of study models was discussed earlier. Accuracy of spiral CT scanning of bone deteriorates exponentially with increasing slice thickness (O'Brien et al. 2001). Errors of greater than 4mm were demonstrated in cranial base landmark location with a scanning slice thickness of 2 mm. Bone is less radiodense than dental stone, so prior to application of CT scanning to dental casts validation of the process would be required. In an object as small as a paediatric dental cast, an error of 4mm would not be acceptable.

2.5.4.2. Dental Arch Data Analysis

2.5.4.2.1. Arch Relationship

In addition to indices such as Angle's classification and incisor relationship, which define deviation from normal dental occlusion, indices exist that describe the dental arch relationship in subjects with cleft lip and palate without reference to the population norm. The Goslon Yardstick, as a measure of the quality of results of surgical repair and subsequent management up to the age of 10, was first described in 1987 and has been widely accepted by orthodontists and other health professionals working in multidisciplinary cleft management teams (Mars et al. 1987). Based on features of the dental relationship, dental study models for subjects with UCLP were categorised into five groups, graded poor to excellent. This grading system showed high inter- and intra-rater agreement after training of the raters. It was concluded that the Yardstick was highly reliable and capable of discriminating between the quality of results in different centres when raters had been calibrated.

However, just as for the 5-Year-Olds' Index which places significant importance on the need for osteotomy as an indicator of poor outcome, the results of Goslon assessment should be interpreted with reference to local osteotomy requirement rates.

The 5-Year-Olds' Index was introduced 10 years after the Goslon Yardstick (Atack et al. 1997). Based on features of dental occlusion, this index demonstrated excellent intra-rater reliability and good inter-rater agreement on assessing outcome of primary surgery at the age of five. The length of time required for a single centre to acquire a sufficiently large number of cases for meaningful statistical analysis of outcome was calculated. It was suggested that an index such as the 5-Year-Olds' Index would allow centres to compare outcome with other centres and detect deficiencies in clinical practice early enough to provide a rational justification for modification of practice. The goal was again to find a non-invasive assessment technique.

The 5-Year-Olds' Index and the Goslon Yardstick have been applied to multi-centre studies of treatment outcome. When applied to 149 cases of UCLP from six European centres, the Goslon Yardstick was found to be more sensitive than cephalometric analysis in discerning differences in dental arch relationship between centres (Mars et al. 1992). It was admitted by the authors that the Yardstick is a crude method of differentiating centres but that its use in the retrospective multicentre approach would provide the basis for further prospective studies. They were applied in the previously mentioned CSAG report (Sandy et al 1998).

Atack et al (1998) compared surgical outcome, based on dental cast analysis using the 5-Year-Old's Index, of two European centres, Bristol, UK and Oslo, Norway. The authors highlighted the deficiency of techniques for early outcome assessment in their introduction. While differences could be detected between the profile of grades of dental malocclusion at 5 years in the two centres, the question of whether children with clefts resemble their affected peers in distant centres more than their local unaffected peers was not addressed. If the rate of osteotomy requirement is accepted as a measure of UCLP surgical outcome, the baseline rate of osteotomy need in the general population should be considered. This ensures that what may appear to be a poor outcome in one centre is not simply a reflection of regional tendency to Class III malocclusion.

The Goslon Yardstick and 5-Year-Olds' Index categorise the spectrum of outcome within the cleft population but do not address deviation from normal, which should be the ultimate outcome of treatment. Huddart's scoring of buccolingual dental relationship, first described to evaluate the arch form and occlusion in unilateral cleft palate subjects, can be applied to the normal subject with modification (Huddart & Bodenham 1972). The limitation of this technique is that it describes just one aspect of the features of a cleft dental relationship, teeth in crossbite, while the former indices address all aspects and allow relatively simple classification by trained practitioners. Huddart scoring has also been criticised for failing to differentiate between the clinical importance of crossbites in different segments of the arch (Mars et al 1987). Anterior crossbites are indicative of deficient anterior maxillary growth and pose a much greater challenge to the orthodontist than posterior crossbites.

2.5.4.2.2. Dental Arch Landmarks

Two-dimensional assessment of the dental arch based on direct caliper measurement of interlandmark distances has been the accepted gold standard of arch analysis. The choice of individual landmarks on teeth as anchors for interlandmark distances or for creating arch forms is usually based on specific cusp tips or defined contact points. There are more landmarks to choose from on posterior teeth and this has lead to a variety of landmark schemata that provide results that are not directly comparable. Many have chosen the mesio-buccal cusp tips as landmarks (Woodworth et al. 1985); (Battagel 1996); (Cassidy et al. 1998). Others have chosen the most prominent palatal point at the junction of the tooth and gingival margin (Laine & Hausen 1985). However the molars may be rotated, tipped or have variable morphology with differing numbers of cusp tips or the teeth may be worn, hypoplastic or restored. All such changes and variously chosen landmarks may affect the measured arch dimensions.

2.5.4.2.3. Dental Arch Dimensions

Arch width is agreed to be inter-dental distances across the midline but variation has existed in the landmarks chosen on each tooth (Bishara et al. 1997). Varying definitions of arch length cause confusion, preventing more widespread meta-analysis. In one extensive study of arch length changes from 6 weeks to 45 years, arch length was defined as the sum of the right and left anterior and posterior arch segments (Bishara et al. 1998). This summation of distances is termed the arch circumference by other authors. Others have defined arch length as the sum of linear lengths taken from the mesial contact point of the first molars on left and right sides to the contact point of the central incisors (Sinclair & Little 1983). Arch length has also been defined as the length of a perpendicular line drawn from the contact point of the central incisors to a line joining the distal contact points of the second primary molars (Mills 1987). Others have calculated a very similar distance to that defined by Mills but have termed it arch depth (Cassidy et al 1998). These discrepancies highlight the need to clarify definitions of arch dimensions before comparing absolute values of study outcomes.

It is common to find spacing of the incisors in subjects with clefts but most of the literature on arch dimensions makes no mention of this problem. In cases where there is a median diastema, one author has suggested estimating the midpoint between the central incisors (Woodworth et al 1985).

Two studies of dental arch development in bilateral cleft lip and palate highlight difficulties posed by differing definitions of arch dimensions and research methodology (Heidbuchel & Kuijpers-Jagtman 1997); (Heidbuchel et al. 1998). Both studies were carried out in the same centre. Subjects were examined from birth to 17 years in two cohorts. In the study of children with

BCLP from birth to 4 years, the control subjects were Dutch boys, the arch landmarks were as defined by Sillman (1964) and the analysis was limited to the maxillary arch. In the other study with similar cleft type, subjects were examined from 3 to 17 years, British non-cleft subjects acted as controls, the study population was of mixed gender, arch width and depth were based on landmarks defined by Moorees (1969) and both maxillary and mandibular arches were analysed. The findings of wide posterior arch width during the first four years of life and subsequent reduction of maxillary arch depth and width with end to end occlusion becoming more marked with time are not directly relevant to the UCLP population. What is noteworthy is the opportunity for a mixed longitudinal study lost by applying differing methodologies to two very closely related studies.

2.5.4.2.4. Dental Arch Shape in Three Dimensions

The analytical techniques described in the earlier section on morphometrics have been applied to 3D co-ordinate data from study models and will not be repeated here.

2.5.5. Results of Studies of Dental Study Models in Non-Cleft Subjects

The studies discussed in this section are summarised in Table 2.3.

2.5.5.1. Control Population Formulation

The choice of control populations for dental arch analysis produces the same challenges for the researcher as in facial anthropometric studies. There are certainly differences in dental arch development between races so samples must be matched for this variable. A mixed cross-sectional study of dental arch development between the ages of 4 and 20 years in Caucasian, Mongoloid and Negroid races showed that the three ethnic groups exhibited differing growth patterns (Lavelle 1975). Arch growth spurts were similar in all three groups

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Lavelle (1975)	To compare dental arch age changed between three ethnic groups	Retrospective mixed cross sectional	40 subjects of Caucasian Mongoloid and Negroid ethnic origin at each yearly interval from 4 to 20	Study models of the dental arches	Caliper measurement of dental arch width, length, index and area	Author	A different growth pattern was shared by the three ethnic groups
Wada and Miyazaki (1976)	To clarify the character of maxillary arch deformity and compare growth between cleft and control subjects	Retrospective mixed cross sectional	87 UCLP and 62 controls from 6/12 to 4 years	Maxillofacial model including patient's upper face and upper dental arch	Caliper measurement of interlandmark distances (Landmarks projected onto single horizontal or sagittal planes)	One of the authors (orthodontist)	Prior to lip repair cleft and normal subjects showed similar growth patterns. Antero-posterior growth inhibition of the maxilla appeared to occur after lip repair and to persist at 4 years
Huddart et al (1978)	To compare the maxillary arches of normal and cleft children before any treatment	Prospective	30 UCLP 30 controls at 14 weeks	Photocopies of study models of the maxillary arches	Linear measurements with Vernier caliper gauge	Author (orthodontist)	Tissue deficiency in the posterior palatal plane in UCLP Cleft width due to displacement of bony segments laterally and anteroposteriorly
Wada et al (1984)	To compare the growth changes of the maxillary dental arch in different types of cleft and to obtain information about the possible role of the nasal septum in the growth of the maxilla	Retrospective	15 UCLP 15 BCLP 15 CP0 at 5/12, 19/12 and 4 years 10 controls at 4 years	Maxillofacial model including patient's upper face and upper dental arch	Caliper measurement of interlandmark distances	One of the authors (orthodontist)	The relation of the nasal septum to the palatal processes may play an important part in the underdevelopment of the maxilla in cleft lip and palate patients

Table 2.3. Summary of Studies of Dental Casts

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Mars et al. (1987)	To develop a clinical tool to categorise the dental relationship according to the severity of the malocclusion	Retrospective Assessments repeated after one week Cases were separated into 5 groups according to the severity of the dental arch malocclusion	55 UCLP at 12 years	Study models of the dental arches	Ranked subjectively Goslon Yardstick	Four experienced orthodontists Four assessors	High intra- and inter-examiner agreement Goslon Yardstick is highly reliable and capable of discriminating between the quality of results of different centres
Nystrom et al(1990)	To compare the effect of two different ages and two different methods of palatal repair on the development of dental arches	Retrospective longitudinal	180 children with UCLP, BCLP or CP 50 non-cleft children Not contemporary sample age 3	Study models of the dental arches	Sliding caliper linear measurements after Moorrees and dental arch relationship	Authors	Timing of palatal repair did not affect arch dimensions in children with CP, UCLP or BCLP Arch dimensions in all affected groups were smaller than in non cleft children
Mars et al (1992)	To assess dental arch relationships in the collaborating centres	Retrospective cross sectional	149 subjects with complete UCLP from six European cleft palate centres	Study models of the dental arches	Goslon Yardstick	5 orthodontists	The Goslon Yardstick was capable of discerning dental arch relationships and by inference facial morphology outcomes between centres more sensitively than Cephalometric variable
Mazaheri et al (1993)	To evaluate the early changes of maxillary alveolar arches of operated UCLP patients	Retrospective mixed longitudinal	88 patients with complete UCLP	Study models of the dental arches	4-point classification	First author	Significant long-term improvement in segment relationship is possible through simple conservative and functional surgical reconstruction of lip and palate tissues

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Derijcke et al (1994)	To evaluate the effect of intrinsic and functional factors on arch development	Retrospective cross sectional	22 adult cases of unoperated UCLP and 15 with UCL	Study models of the dental arches	Reflex microscope digitisation of casts after Moyer	Orthodontists	Arch width and depth were smaller in the complete cleft group
Nystrom and Ranta (1994)	To compare the effect of two different ages and two different methods of palatal repair on the development of dental arches	Retrospective longitudinal	120 children with CP and 50 non-cleft children ages 3 and 6	Study models of the dental arches	Sliding caliper linear measurements after Moorrees and dental arch relationship	Authors	Dental arches of children with CP were significantly smaller than those of NONC children, with the discrepancy increasing form age 3 to 6 and more in the maxilla than the mandible
Honda et al (1995)	To analyse maxillary arch growth changes from the time of cheiloplasty to 4 years of age	Retrospective	95 children with UCLP, BCLP, CPO or CL pre-op to age 4	Study models of the dental arches	Sliding caliper measurement of arch width and depth after Sillman	Author (Maxillofacial surgeon)	Palatoplasty affected transverse and anteroposterior arch growth Cheiloplasty influenced anterior but not posterior arch width Surgery not the only factor affecting growth
Richey et al (1995)	To determine pre-treatment arch dimensions of short children and to evaluate the response to rhGH treatment by measuring arch change over 5 years	Prospective longitudinal with retrospective study of controls	28 short subjects 18 receiving immediate treatment 10 receiving delayed 28 controls from archived data	Digital photographic images of study models of the dental arches	Landmark digitisation on screen and automatic measurement of interlandmark distances	Authors (orthodontist)	The influence of rhGH on arch dimensional changes over time remains equivocal

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Mishima et al (1996)	To compare the palatal forms in infants with UCLP who were fitted with Hotz' plate with those not fitted with the plate up to cheiloplasty	Prospective longitudinal	20 infants with UCLP	Wire frame models of palate constructed from 3-D digitiser	Wire frame model superimposition	Authors	Both major and lesser maxillary segments moved mesially in Hotz' plate wearing infants. Degree of palatal curvature was also less in these subjects
Mishima et al (1996)	To compare the palatal forms in infants with UCLP who were fitted with Hotz' plate with those not fitted with the plate from cheiloplasty to palatoplasty	Prospective longitudinal	20 infants with UCLP	Wire frame models of palate constructed from 3-D digitiser	Linear measurement Superimposition of constructed wire meshes	Authors	The appliance could guide the growth of the maxillary segments to narrow the cleft width until 18 months of age
Atack et al. (1997)	To develop an index for early assessment of repair of UCLP	Retrospective	27 UCLP at 5 and 10 years	Study models of the dental arches	5-point scale	Four orthodontists	Excellent intra-examiner agreement was demonstrated Inter-examiner agreement was good
Heidbuchel and Kuijpers-Jagtman (1997)	To study occlusion and arch dimensions in BCLP and compare the variables with those of non cleft individuals	Retrospective longitudinal	22 Dutch BCLP 42 British non cleft subjects Aged 3 to 17	Study models of the dental arches	Optocom digitiser measurements of arch width and depth Huddart scoring and Angle classification of inter arch relationship	Orthodontists in two centres	Maxillary arch depth and width were significantly smaller in BCLP end to end occlusion was found becoming more marked with time

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Opitz and Kratzch (1997)	To trace the changes in maxillary dimension in UCLP and BCLP up to time of palate surgery at four stages	Prospective longitudinal	44 UCLP and 28 BCLP from birth to age 3	Study models of the maxillary arches	Reflex microscope linear measurements including slope of palatal segments	Authors	Continuous reduction in cleft dimension parallel to increasing transverse and sagittal dimensions Differences between the two types of clefts could only be found in arch depth Pre surgical orthopaedic plates prevented anterior transverse collapse Lip surgery caused only temporary inhibition of sagittal growth
Atack et al. (1998)	To examine whether differences in dental arch relationships in 5-year old children could be detected between two centres	Retrospective	46 UCLP 54 UCLP	Study models of the dental arches	5 - point scale	Two orthodontists	It was possible to detect differences in surgical outcome at 5 years of age
Bishara et al (1998)	To evaluate changes in maxillary and mandibular total arch length over a 45 year span between 6 weeks and 45 years	Retrospective mixed cross-sectional	61 subjects at 6 weeks, 1 and 2 years 30 subjects at 8,13,26 and 45 years	Study models of the dental arches	Digital caliper measurement of arch length Arch length - (sum of right and left anterior and posterior segmental arch length)	2 investigators	Greatest incremental increases occurred during the first 2 years of life. Arch length continued to increase until 13 years in the maxillary arch and 13 years in the mandibular arch Then significant decreases occurred in both arches mesial to the permanent molars

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Heidbuchel and Kuijpers-Jagtman (1998)	To describe the development of maxillary arch dimensions in children with BCLP during the first 4 years of life and compare it with that in non-cleft children	Retrospective mixed longitudinal	26 BCLP (Nijmegen) 34 non cleft subjects (Amsterdam) birth to 4 years	Study models of the dental arches	Reflex Microscope digitisation of casts arch width and depth after Sillman	1 examiner	During the first 4 years of life, maxillary arch dimensions are shorter and narrower anteriorly and wider posteriorly than in non cleft children
Mishima et al (1998)	To analyse the configuration of the pre-maxilla and vomer in infants with BCLP and to clarify factors causing malalignment	Retrospective longitudinal	10 infants with complete BCLP	Study models of the palate Wire frame models of palate constructed from 3-D digitiser	Linear measurement Superimposition of constructed wire meshes	Authors	At 1month of age, a greater inclination and a smaller deviation of the vomer and a longer distance between the cleft edges of the lateral segments had a tendency to be associated with bending of the vomer or twisting of the pre-maxilla
Sandy et al (1998)	To examine the national standard of cleft care and outcomes in the United Kingdom To examine the training of recently appointed consultant orthodontists	Retrospective	326 5-year old UCLP and 321 12-year old UCLP	Study models of the dental arches	5-year old index and GOSLON index	Two orthodontists calibrated in the use of both indices	Standards of care not raised in last decade Recommendation that fewer orthodontists will need to be involved in centralised care model for children with clefts
Ishikawa et al (1999)	To determine the effect of mucoperiosteal denudation of the bone on maxillary alveolar growth	Retrospective	2 subjects aged 12 with CPO	Live subjects Laser scanned study models of the dental arches	Laser Doppler flowmetry of palatal blood flow Intra subject comparison of dental arch, alveolar arch and basal arch forms across constructed midline	Orthodontists	Bone denudation in the premolar region appeared to result in less than 3mm of palatal dental displacement and no change in lingual inclination

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Stellzig et al (1999)	To investigate whether growth increments until six months were influenced by particular factors To analyse relationship between cleft size at birth and anterior cleft reduction To examine the correlation between maxillary measurements at birth and cleft width at six months	Prospective longitudinal	34 subjects with complete UCLP	Maxillary plaster casts of the subjects	Computer controlled 3D digitising system	Orthodontists	Gender plays a role in growth changes in the first six months A correlation was demonstrated between maxillary measurements at birth and growth increments in the first 6 months
Friede et al. (2000)	To find out if the expected improvement in maxillary development had actually occurred with new operation	Retrospective longitudinal	64 UCLP with repair with minimal denudation of bone 32 UCLP following push back repair	Study models of the dental arches	Digital caliper measurement of maxillary arch width and depth	Team orthodontists	Differences were significant only for patients with velar clefts
Johnson et al (2000)	To determine the importance of initial cleft severity in determining patient outcome	Retrospective	49 children aged 6 with UCLP	Flat-bed scanned images of study models of the dental arches Study models of the dental arches	5 - point Birth Severity Index 5 - point scale	2 orthodontists	No evidence found that initial cleft area determined quality of surgical outcome at age 6
Johnson et al (2000)	To evaluate complete UCLP repair outcome in Western Australia by assessment of dentoalveolar relationships	Retrospective	54 children with UCLP	Study models of the dental arches	5 - year old study model index	2 orthodontists	Results compared favourably to results in United Kingdom but unfavourably to results in Norway

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Lindsten et al (2000)	To test the hypothesis that lateral arch spaces are greater in present-day 9-year-olds than they were in 9-year-olds born 20 to 25 years ago	Retrospective	127 children born in 1960s 125 children born in 1980s	Study models of the dental arches	Sliding caliper measurement between contact points of the first molar and lateral incisor in four quadrants	Authors	Children born in the 1980s had significantly larger maxillary and mandibular lateral arch spaces than those born in the 1960s
Morris et al (2000)	To assess the dental arch relationships of children with UCLP in region of UK. To compare this result with previously published with previous intercentre European study	Retrospective	35 subjects with UCLP	Study models of the dental arches	Goslon yardstick	First author	Useful baseline data against which progress in achieving improved outcomes can be determined by further research
Prasad et al (2000)	To determine whether two different prepalatoplasty protocols quantitatively affect maxillary arch morphology in infants with UCLP	Retrospective longitudinal	59 infants with UCLP and 29 unaffected infants (archive from the 1930s - the Sillman collection-pre-op to 18 months)	Study models of the dental arches	Directly digitised and derived 3D features	Authors	Two different regimens for the initial management of UCLP can significantly affect maxillary alveolar arch growth with respect to the treatment used and in comparison with unaffected controls
Nojima et al (2001)	To clarify morphological differences between Caucasian and Japanese mandibular arch forms in different malocclusions	Retrospective	320 subjects of Japanese and Caucasian origin	Photocopied study models of the dental arches	Linear measurements and 3 point arch shape classification	Authors	No statistically significant difference was found between the two ethnic groups within each arch form sample

Author	Aims	Design	Sample	Stimulus Media	Measurement System	Raters	Conclusions
Peltomaki et al (2001)	To examine possible associations between severity of clefting in infants and maxillary growth in UCLP	Retrospective longitudinal	24 UCLP at age 1/12 and 5 years	Study models of the maxillary arches at 1/12 and lateral cephalograms at 5	Digital sliding caliper linear measurement after Friede Linear measurement of traced lateral cephalograms	Author (orthodontist)	Patients with large clefts and small arch circumference, arch length or both demonstrated less favourable maxillary growth
Prahl et al (2001)	To evaluate the effect of infant orthopaedics on maxillary dimensions in infants with UCLP	Prospective randomised trial	49 UCLP at 15, 24, 48 and 78 weeks with or without infant orthopaedic plates	Study models of the maxillary arches (elastomeric impressions)	Reflex microscope digitisation of maxillary arch study models after Shaw and linear measurement	Orthodontists who had not treated the children Not blinded	Infant orthopaedics only has a temporary effect on maxillary arch dimensions that does not last beyond surgical soft palate closure Infant orthopaedics as a tool to improve maxillary arch form could be abandoned
Warren and Bishara (2001)	To investigate changes that may have occurred in dental arch dimensions over five decades	Retrospective	112 contemporary children 175 historical counterparts	Study casts of dental arches	Digital caliper measurement of interlandmark distances	Author (orthodontist)	The average arch may be smaller n contemporary children than in past generations

occurring between 5 and 7 years and again between 11 and 13 years. Sexual dimorphism levels varied between the three groups and arch circumference also varied to a significant level. Form is a function of size and shape. In a study aiming to clarify differences between Caucasian and Japanese mandibular arch shape in Class I, II and III malocclusions, mandibular arches were classified into ovoid, square or tapered in order to determine the frequency distribution of the 3 arch forms for each ethnic group (Nojima et al. 2001). It was concluded that there is no single arch shape unique to any of the Angle classifications or ethnic groups and that it is the frequency of arch shape that varies between these groupings. Dental arch dimensions were found to differ significantly between the two ethnic groups but ethnic differences in dental arch relationship were not addressed.

As archives of study models can be examined repeatedly and data collected using a variety of techniques, there is evidence in the literature of a heavy reliance on historical, rather than contemporaneous, archives for control data. It has not been established how long study model archives remain valid as population samples. After analysing the study models of two cohorts of nine year olds, one born in the 1980s and one born in the 1960s, it was concluded that children born in the 1980s had significantly larger maxillary and mandibular lateral arch spaces than those born in the 1960s (Lindsten et al. 2000). It was suggested that a recent reduction in the prevalence of caries could account for the difference found between the two samples. This study would suggest that a 20 year gap between data from controls and research subjects is too long to ensure the results of direct comparison are valid.

In a study over a longer time span, the casts of children enrolled in the Iowa Growth Study and born between 1946 and 1948, were compared with those of children from the same ethnic background in North America born between 1992 and 1995 (Warren & Bishara 2001). Maxillary and mandibular arch lengths were smaller in the contemporary sample. Arch widths were smaller in the contemporary sample in boys but not in girls. The presence or degree of dental crowding was not recorded. This study showed that in fifty years more marked differences between population norms occur than were seen in cases born twenty years apart. The authors postulated that increased use of pacifiers, increased consumption of food requiring less mastication and reduction in rates of breast feeding might account for the differences between the two samples. The maximum length of time for which archives remain valid has not been established yet.

2.5.5.2. Longitudinal Arch Changes

Among the studies of non-cleft individuals that demonstrated the problems of arch dimension definitions, one also reported findings of significance to the cleft population and highlighted some of the other problems associated with longitudinal studies. This study of normal dental arch length changes over 45 years used two groups to create a longitudinal sample (Bishara et al 1998). 61 subjects were followed from 6 weeks to 2 years but only 28 cases were studied from 3 to 45 years. The latter subjects were enrolled in the Iowa growth study and some of these subjects had been born 50 years before the subjects examined at 6 weeks to 2 years. The low number of complete data sets from the original 175 children highlights the difficulty of subject retention. Confirming Sillman's (1964) work on arch development from birth to adolescence, it was found that the greatest incremental increase in arch length occurred in the first 2 years. Maxillary arch growth continued to 13 years and mandibular arch length growth ceased at 8 years of age. Thereafter, arch length decreased mesial to the first permanent molars in both sexes.

2.5.6. Results of Studies of Dental Study Models in Cleft Subjects

Dental study models have been used to examine the intrinsic dental arch growth patterns in different cleft types, responses to various surgical regimens or pre surgical orthopaedic treatment and outcome prediction.

In subjects in the developing world with cleft lip or cleft lip and palate who had not been operated on before adulthood, it was possible to evaluate the intrinsic and functional factors affecting dental arch growth without iatrogenic (surgical) factors (Derijcke et al 1994). Qualitative evaluations of the dental casts of 22 adult cases of unoperated UCLP and 15 cases of unoperated UCL were carried out using the Goslon Yardstick Analysis and modified Huddart scoring. The width of the cleft in the anterior arch was evaluated. Dental casts were digitised with a Reflex Microscope according to landmarks defined by Moyers et al (1976). The cleft edge, the centroid of each tooth, the centroid projected onto the occlusal plane and the cleft edge points were calculated. Arch width and depth, palatal inclination and deviation of the midline were also calculated. The average age of the subjects in both groups was approximately 24 but the ages ranged from 15 to 40. A 25 year span would introduce age-related changes in the arch dimensions anterior to the first permanent molars but this factor was not taken into account by the researchers. No differences were found between the mandibular arches of both cleft types. The maxillary arches of subjects with UCLP were narrower in the canine and premolar areas and the arch depth was smaller that in UCL. The maxillary arch segments were either closer or overlapping to a significant degree in UCLP with no overlapping or spacing of up to 5 mm occurring in UCL.

The relatively normal dental arch found in unoperated UCL and UCLP subjects supported the hypothesis that developmental disturbances are due to surgery. This hypothesis was undermined by the finding of differences between arch forms in UCL and UCLP. It was postulated that surgical procedures influence further growth aggravating the intrinsic tissue deficit. Dental arch characteristics were found in unoperated UCLP that are usually present to a more significant degree in operated cases.

This study looked at the inherent factors intrinsic to each type of cleft by comparing growth of the maxillary arch in children with one of four types of clefting, unilateral cleft lip or cleft lip and palate, bilateral cleft lip and palate and cleft palate alone (Honda et al. 1995). Unlike the study of unoperated adults, these subjects had early cheiloplasty and palatoplasty. The former surgery had an effect on anterior but not posterior arch width. The latter procedure affected transverse and anteroposterior arch growth. Surgery alone did not explain the differing growth patterns in the first four years after birth nor did the cleft type strictly determine growth patterns. This finding agrees with researchers who studied dental cast changes from age 4 to 8 and found variation of growth patterns within groups categorised according to cleft type (Ohishi et al. 1992).

An example of two papers drawing opposing conclusions on the benefit of presurgical orthopaedic treatment demonstrates the ongoing controversy surrounding this aspect of cleft management. Three-dimensional imaging and automated landmark identification were used to acquire data on the palatal arch form, and later the dentate arch form, at the time of cheiloplasty and of relatively late palatoplasty in subjects who had received Hotz plate treatment in the neonatal period (Mishima et al 1996). The authors concluded by supporting the use of such plates. A randomised control trial on 49 UCLP cases took an opposing, and equally well supported, view on the benefits of maxillary plate wear (Prahi et al. 2001). The authors concluded that plate wear had only a temporary effect on maxillary arch dimensions that did not last beyond soft palate closure, so infant orthopaedics as a tool to improve maxillary arch form could be abandoned. Spontaneous improvement in palatal form has been reported in cases of palatal closure delayed to age 3 years (Opitz & Kratzsch 1997). The authors accepted that speech development could be adversely affected by delayed palatal closure but did not address the ethical issue of deciding between adverse speech outcome and adverse anatomical outcome.

Recent studies of dental study arches have considered the effects of the trend towards palatoplasty procedures with less bony denudation. Follow up studies of the "push-back" palatoplasty had shown deleterious effects on transverse maxillary arch growth (Ross 1970). A study correlating the amount of avascular scar tissue of the palate, measured with laser doppler flowmetry, and dental arch form, described after laser scanning, concluded palatal scarring after push-back palatal repair did not appear to inhibit vertical maxillary growth (Ishikawa et al 1999). In a larger group of subjects with UCLP, it was concluded that only in cases of velar clefting did the switch to a procedure requiring less palatal denudation confer benefit in terms of arch width gain (Friede et al 2000). The subjects were examined at 12 years of age which shows how long the audit cycle for surgical regimens could be.

Prediction of outcome can be based on the initial assessment of the cleft or it can be based on later developmental stages but robust dental arch predictors of eventual outcome are still lacking.

It has been suggested that examination of the occlusal relationship from dental study casts without clinical or radiographic examination of the soft tissue profile and skeletal base, could be used to detect treatment outcome of primary surgery in children as young as 5 years of age (Atack et al. 1997). This was 5 years earlier than the previously widely accepted Goslon Yardstick scale which is applied at 10 years. When the same cases were assessed at the age of 16 to 18 years, following orthodontic treatment but prior to orthognathic surgery, there was a significant difference between the number of subjects requiring osteotomy at eighteen and the number of subjects at five falling into the category deemed likely to need an osteotomy in the future. The predictive power of this index was especially poor on an individual basis as the group subsequently requiring osteotomy was smaller than predicted. This group also included subjects who had been predicted likely to have a fair or better long-term outcome. The authors accepted this weakness and pointed out that the decision to perform orthognathic surgery was not made on the basis of dental occlusion alone but would also include consideration of facial appearance and cephalometric analysis.

Using an approximate measure of palatal cleft area taken from a flat-bed laser scanned image of neonatal palatal casts and correlating this with the dental arch relationship at 5 years of age, it was concluded there was no correlation between the two factors (Johnson et al 2000a). Rather than taking the dental arch relationship as a synonym for midface growth as Johnson et al (2000a) did, a more recent study correlated neonatal maxillary arch measurements and cephalometric measurements at 5 - 6 years of age (Peltomaki et al. 2001). A significant correlation was found between the two variables. If the relationship between dental arch relationship and midface growth was accurately quantified, radiography that was not clinically indicated could be avoided.

2.5.7. Results of Cleft Case - Control Studies of Dental Study Models

Literature pertaining to such study models is not extensive and in some cases the number of control subjects are very low, undermining the significance of the findings.

Results from one of the few studies that included contemporaneous and local controls and used a prospective study design are now almost 25 years old (Huddart et al 1978). Arch form was compared between a UCLP group and a control group at 14 weeks of age using photocopied images of the maxillary casts. The contribution of tissue deficiency to the width of the palatal cleft was quantified as 22% with lateral segment displacement and increased slope of the palatal shelves accounting for the remainder of the differences. A more recent case control study, which aimed to evaluate two different infant management techniques for UCLP used, what must be regarded as, a redundant control group (Prasad et al 2000). The control population was composed of dental casts from an archive collected 60 years before the study populations were born. It has been documented that after fifty years the frequently utilised Iowa Growth Study archive of dental casts differed significantly from the dental casts of contemporary subjects. Using casts from subjects born one decade earlier cannot provide an acceptable control population. The results of the study will not be discussed, the feature of note being the poor study design.

Comparing a small (50) but contemporaneous control sample to a study population of 180 subjects with a mixture of UCLP, BCLP and CPO cleft types, significant differences from normal in both maxillary and mandibular arch dimensions were reported for all affected subjects (Nystrom & Ranta 1990); (Nystrom & Ranta 1994). At the age of 3 years, all arch dimension were smaller in cleft subjects and at 6 years of age the discrepancy in size had increased, more markedly in the maxillary arch than in the mandibular. Another study of maxillary arch growth, with an even smaller control population, compared 135 observations from a study sample of 15 ULP, 15 BCLP and 15 CPO examined at 5 months, 19 months and 4 years with data from 10 control subjects examined only at 4 years. The study required combined midface and maxillary arch impressions to be recorded under general anaesthesia, which explains the tiny number of normal observations (Wada et al 1984).

2.6. Summary of Literature Review

The review of the literature can be summarised as follows:

- The multiple problems associated with orofacial clefting require extended multidisciplinary management. The outcome of individual aspects of treatment is difficult to assess, especially in children.

- Outcome of treatment in Britain has compared unfavourably with outcome in other European countries.

- Radiographic assessment of cleft related deformity and normal facial morphology is being superseded by non-invasive imaging techniques, which are often three-dimensional.

- Linear dimensional analysis is the common denominator of all dentofacial shape analysis. This does not have a universally applicable counterpart in the analysis of three-dimensional shape.

- Normal reference values for facial soft tissue morphology and dental arch morphology are deficient for the paediatric population.

- Paediatric soft tissue facial morphology in subjects with orofacial clefting has not been extensively investigated.

- Palatal reconstruction may have an adverse effect on the growth of the nasomaxillary complex.

- Objective assessment of facial function in co-operative adult subjects is difficult. Assessment of paediatric facial function remains qualitative.

2.7. Aims

The aims of this study were, therefore:

1. To apply computerised stereophotogrammetry and three-dimensional morphometric assessment to soft tissue facial morphology in 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

2. To measure the statistically significant differences in soft tissue facial morphology at rest between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

3. To measure the statistically significant differences in soft tissue facial morphology in maximum smile between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

4. To measure the statistically significant differences in dental arch size and shape between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

The following null hypotheses are tested:

1. Computerised stereophotogrammetry and three-dimensional morphometric assessment cannot be applied to soft tissue facial morphology in 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

2. There are no statistically significant differences in soft tissue facial morphology at rest between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

3. There are no statistically significant differences in soft tissue facial morphology in maximum smile between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

4. There are no statistically significant differences in dental arch size and shape between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

Chapter 3

Materials and Methods

3.1. Materials

3.1.1. Data Acquisition

3.1.1.1.Stereophotogrammetry Equipment

The stereophotogrammetric camera system is not available commercially. It is an experimental custom-built product. (Figure 3.1.) The system consists of two pods each containing two monochrome cameras, one colour camera, one texture slide projector / flash and one white light flash arranged on a rigid frame around a dental chair which allowed movements of the subject along the X, Y or Z axes in isolation. The camera pods and the dental chair form an approximate equilateral triangle.

A calibration target of discs of known dimensions and locations on a contrasting background is captured by the cameras in a variety of poses before the subject is photographed (Figure 3.2.). Images of the target from all the cameras are processed to find the central location of the discs and these co-ordinates are used to create an approximate geometric model of each camera and the camera's respective relative orientation to the target.

The cameras are connected to a personal computer via two universal serial bus (USB) cards to minimise interference from electrical noise and are driven by an uninterruptable power supply unit connected to mains electrical supply.

When the slide projector / flash and four monochrome cameras are activated, four images of the face with a speckled light overlay are captured simultaneously during a 10 millisecond exposure. After a 30-millisecond delay, the colour cameras and white light flash are activated and two further images of the face are captured during a second 10-millisecond exposure. This adds up to a total capture time of 50 milliseconds. The images captured are shown in Figure 3.3.

Figure 3.1. Subject's view of Stereophotogrammetry Equipment

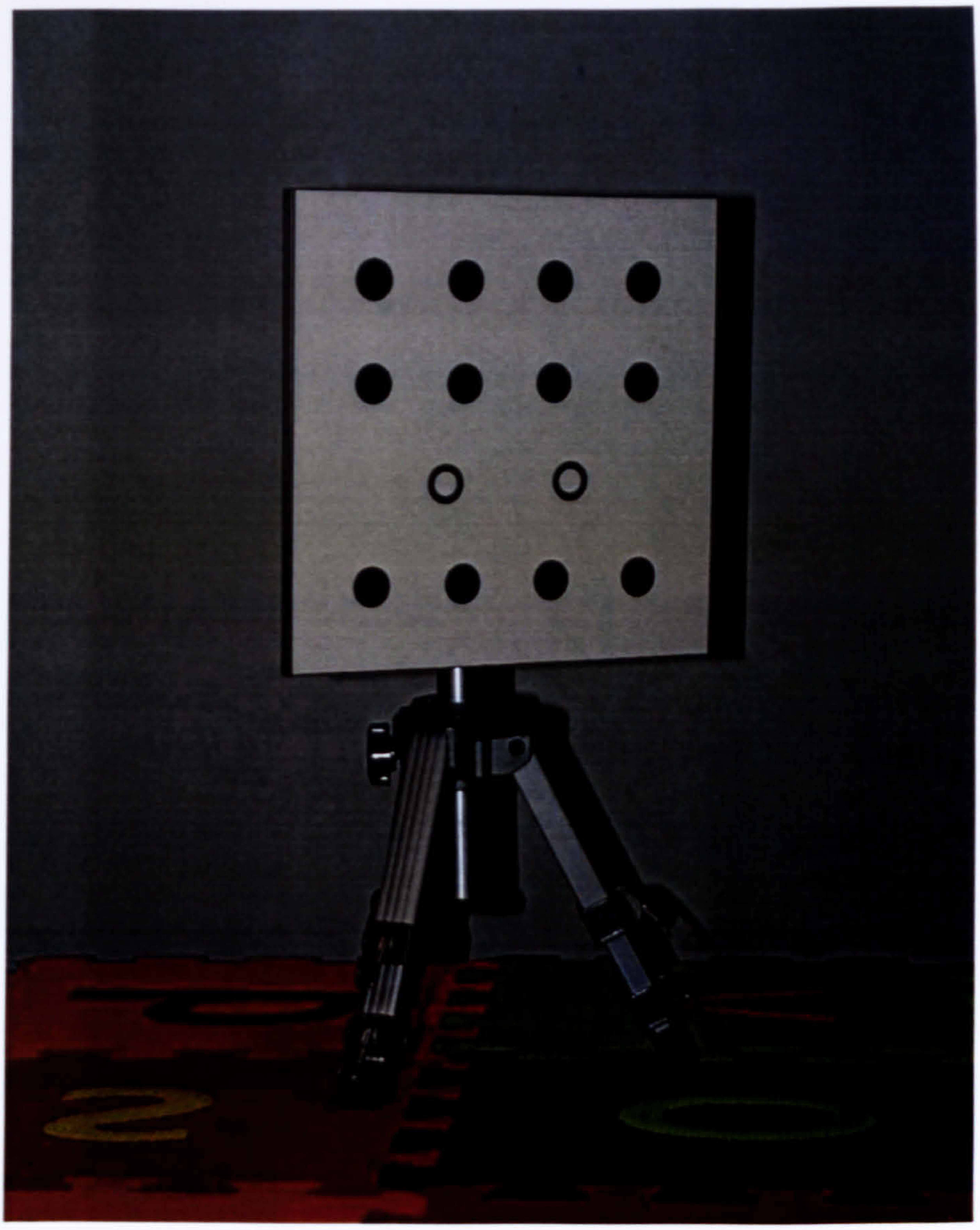


Figure 3.2. Calibration Object for Stereophotogrammetry Equipment

Figure 3.3. Multiple Images Captured by 6 Cameras in 50 milliseconds

The two monochrome images of either side of the face contain slightly differing views of the face and of the speckled pattern. The geometry of the camera set up is known from the earlier calibration process and this allows the projection of notional rays from each pair of corresponding pixels in the stereoimages and their intersection in 3D space to be computed. This process generates a point cloud, which has no undercuts. The point cloud captured from each pod of cameras can then be transformed into the same co-ordinate frame and merged into a single triangulated polygon mesh with true 3D information. This is the range data, which appears as a homogeneous facial representation. The colour images from the two pods are also merged and attached to the range data to allow anthropometric landmarks to be identified in the constructed 3D image of the subject's face (Figure 3.4.).

The computerised stereophotogrammetry equipment consists of the following components:

Slit lamp locating devices - Two 3 volt DC bulbs, projected through 25 mm photographic lenses, are mounted on 2 Spindler and Hoyer Microbenches.

Four monochrome cameras - These were custom manufactured by Gadarian, Edinburgh. Image resolution is 1000 x 800 pixels with a USB communication and data link to the personal computer controlling them. The cameras contain compound metal oxide semiconductor (CMOS) chips, which refers to the method by which the chip is manufactured.

Two colour cameras - Produced by the same manufacturer with single CMOS chips.

White light flash - This is a Centon FH95 Flashgun, available from all photographic retailers.

Texture flash and monochrome slide - The texture flash is a Centon FH95 Flashgun unit adapted by Gadarian to allow the basic flash unit to illuminate the texture slide which is focused onto the subject's face using a Schneider 25 mm projection lens. The monochrome texture slide is a 35 mm slide mounted photographic reproduction of a tagged image file (.tif) created by 3D-MATIC to have a random (non-repeating) pattern.

Figure 3.4. Images Merged to Create 3D Map of Facial Surface

Both flash units are synchronised to the cameras using Flash Control Boxes, custom manufactured by Gadarian. These power the flashes, receive trigger signals from one monochrome and one colour camera and relay this trigger signal to the two pairs of flash units.

The images were downloaded onto a personal computer with C3D™ software and a Windows 98™ operating system installed. The data are transferred to the personal computer through a USB connection using quadroport peripheral component interconnect (PCI) cards. These provide 4 USB ports in a single PCI port.

3.1.1.2. Recording of the Dental Arches

The occlusal surfaces of the upper and lower dental arch were recorded in addition-cured polyvinylsiloxane impression material (Aquasil, Dentsply, DeTrey GMBH) on thermoplastic customised special trays (Erkodur thermoforming discs, Erkodent, Erich Kopp GmbH) and the occlusal relationship was recorded in softened wax (Modelling Wax, Dentsply, DeTrey GMBH). The impressions and occlusal registration records were sterilised according to Glasgow Dental Hospital cross infection control policy protocol (Smith et al, 1999). Impressions were soaked in surface disinfectant (Virkon (1%), Antec International Ltd., U.K.) for 5 minutes. The impressions were poured in synthetic dental stone (Goldstone, Bracon Ltd., U.K.) by an experienced orthodontic laboratory instructor. Direct measurement of the study models was performed using a digital caliper (Digimatic Caliper, Mitomuyo Corporation, Japan)

Digital Imaging and Communications in Medicine (DICOM) images of the study models were recorded using a Marconi Mx 8000 quadslice spiral CT scanner (Phillips Medical Systems, U.K.). DICOM is a network protocol for the transmission of medical images and ancillary information. Batch scanning was employed with the models arranged on polystyrene ceiling tiles with plastic cup spacers (Figure 3.5.).

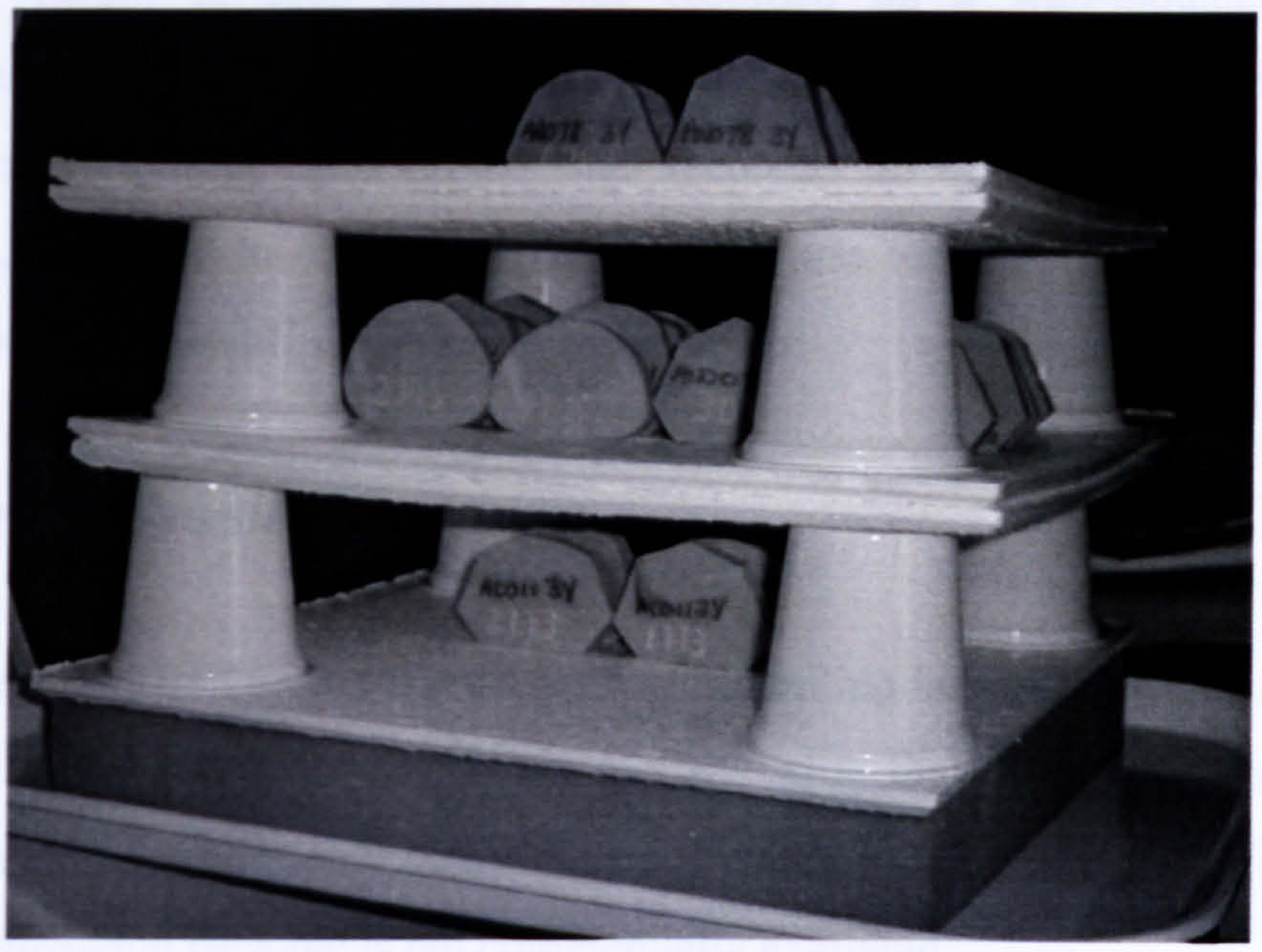


Figure 3.5. Close up view of study models in CT scanner

3.1.1.3. Recording of Weight and Height

The standing height of each subject was recorded on a wall-mounted telescopic measuring rod (seca 220, seca Vogel und Halke GmbH & Co. Germany). The weight of each subject was recorded on digital baby and toddler scales, calibrated to zero before each weight was recorded (seca 835, seca Vogel und Halke GmbH & Co.).

3.1.2. Data Processing Software

Landmarks were digitised on the Virtual Reality Modelling language (VRML) images of the facial soft tissue and study models using the Open Graphics Library (Open GL) based Facial Analysis Tool created for this project. This display and digitisation package is not commercially available.

The DICOM images of the dental study models, produced by spiral CT scanning, were segmented and volume rendered using AMIRA™ software (Version 2.3, TGS Inc., U.S.A.) and exported to VRML.

3.1.3. Statistical Software

Basic statistical analysis, ANOVA and non-parametric analysis of linear dimensions was carried out using MINITAB version 13 software (Minitab Inc., U.S.A.). Three-dimensional analysis employed S-PLUS software (Statistical Sciences Corporation, MathSoft, U.S.A.)

3.2. Method

3.2.1. Study Design

This was a prospective, cross-sectional, case-control cohort study.

3.2.2. Ethical Approval

Ethical approval to proceed with this study was granted by the Local Area Dental Ethics Committee of North Glasgow University Hospitals NHS Trust.

3.2.3. Subject Recruitment

3.2.3.1. Sample Size Calculation

The Scottish Association for Cleft Lip and Palate (SCALP) registry and database for the year 1997 to 1998 provided a profile of children with orofacial clefting at birth, limited to gender, diagnosis and geographic location of primary registration, who might be eligible for inclusion in the study. The database did not record ethnic origin or associated congenital anomalies or subsequent mortality. It was projected that 30 children with unilateral cleft lip or unilateral cleft lip and palate could be recruited to the study at 3 years of age. This was based on the documented number of children born with UCL or UCLP in 1997 and 1998 and likely rates of non-Caucasian ethnic origin or other birth defects or other exclusion criteria.

To calculate a control sample size, the projections for the number of children with clefts who could be recruited was multiplied by a factor of three. A control sample of 90 children was deemed to be sufficient to reduce the contribution from the unaffected group to the variability of difference between the groups. Further increases in the control sample size would have had diminishing returns because of the unchanging contribution from the cleft groups. See Appendix 1 for a graphic representation of the effect of increasing the control sample.

There was no published work available indicating the magnitude of any possible differences between the group which could have further informed the sample size calculation.

3.2.3.2. Study Sample

Two groups of 3-year-old children were recruited and examined at Glasgow Dental Hospital.

Group One

This group of 36 Caucasian Scottish children with cleft deformities was recruited with the assistance of the clinical staff in each of the surgical centres in Scotland. The group was divided into two subgroups; one subgroup of 13 children with unilateral cleft lip (UCL) and another of 21 children with unilateral cleft lip and palate (UCLP). There was some reluctance on the part of parents of children with UCL to take part in the study. Most children with UCL required no further contact with the multidisciplinary care team after initial corrective surgery and parents reported the cleft deformity to be a historic event in the child's life rather than an ongoing problem.

All of the affected children had primary reconstructive surgery in either Glasgow or Edinburgh performed by one of three surgeons following the same surgical protocol. Passive feeding appliances with lip strapping were fitted in neonates with palatal clefts and worn up to the time of lip surgery. At the age of 3 months, a modified Millard cheiloplasty and a Mc Comb nasal correction were performed. At the age of 9 months, children with cleft palate had palatoplasty with minimal bony denudation performed. "Push-back" palatoplasty was not performed.

Children born before full term, those with orofacial clefting as part of a craniofacial syndrome and where management deviated from the surgical protocol outlined above, were excluded.

3.2.3.3. Control Sample

Ninety Caucasian children living in the Greater Glasgow and surrounding areas constituted the control group. Glasgow City Council Education Department denied direct access to the nurseries under its jurisdiction but gave permission to ask head teachers to disseminate recruitment leaflets to the parents of children born between October 1997 and August 1998. This mailshot was followed by the recruitment of 5 subjects.

Permission was obtained from Councils adjacent to Glasgow City to recruit 3-year-old children through Council run nursery schools. The mailshot and nursery based recruitment campaign covered 29.7% of the population of Scotland. The researcher wrote to all nursery schools managed by councils to explain the study to the teaching staff and request their assistance. With agreement of staff, the first names of enrolled children born from June 1997 to June 1998 and the surnames of their parents or guardians were obtained. A personalised letter and a participation acceptance / rejection form was prepared for each child by the researcher and posted to the guardian by the nursery school staff. (Appendix 2)

Guardians who expressed an interest in participation in the study, by providing their address and phone number on the acceptance form, were contacted directly by the researcher to arrange an appointment for data collection. The following exclusion criteria were applied: -

- premature birth
- homozygous twin (other twin included)
- poor co-operation and /or anxiety
- current history of digit / pacifier sucking habit
- dental caries or early loss of primary teeth

The socio-economic profile of the population of Glasgow and surrounding area is similar to that of the Scottish cleft population. It was possible to recruit a deprivation category matched sample by applying the systematic approach outlined above to recruitment over a wide geographic area.

At the child's visit to the Dental Hospital, written informed consent was obtained (Appendix 3).

3.3. Pilot Studies

3.3.1. Study One - Stereophotogrammetry Process

3.3.1.1. Preliminary Set Up of Camera System

The size of the subject dictated the physical arrangement of the cameras. As two separate studies were utilising the same cameras, the working volume had to accommodate the average head size of young children (aged 3 months to 5 years).

A doll, approximately the same size as a 2-year-old child, was posed in the dental chair and the cameras arranged around it such that the composite views from the colour cameras showed the surface of the doll's face from tragus to tragus and from the upper forehead to the lower part of the neck (Figure 3.6.). Composite monochrome camera views also delivered full coverage with all cameras from both pods showing the vertical mid-face. The doll occupied half of the visual field of each camera.

The camera aperture and focus were set to create a focal trough deep enough to accommodate the doll's head. The white and patterned light flashes were positioned to ensure illumination of the doll's head from tragus to tragus. The patterned light projector was focused to ensure discrete random pattern projection onto the skin surface throughout the focal trough. Photographing a range of volunteer subjects, aged from 3 months to 6 years, tested the camera position to ensure full-face data recording and illumination were reproducible.

3.3.1.2. Estimation of Camera Target Zone and Working Volume

An arbitrary point zero was determined from the position of the doll's head relative to the posterior wall, floor and the perpendicular bisection of the inter camera pod distance when the head appeared in the centre of each camera's visual field. A rigid tape measure arranged horizontally and vertically in turn was attached to the doll located at point zero and the resultant set up

Figure 3.6. Doll posed for Customisation of Camera Set Up

photographed. The doll was moved in 2.5cm increments away from point zero along the positive and negative arms of each axis until it disappeared from the camera field of view. The doll's head was 10cm from ear to ear and could be visualised when moved up to 5cm from the starting point along the positive and negative limbs of the X-axis. The limits of movement along the Y and Z-axes were 2.5cm and 5cm respectively with the dimensions of the head being 14 cm anterior to posterior limits and 10 cm from superior to inferior limits. Therefore, the target zone was calculated to be 20cm x 19cm x 20cm.

3.3.1.3. Computed Confirmation of Target Zone and Working Volume

To confirm the above calculations, the C3D™ internal frame of reference was applied to 3D models built from composite camera images of the doll and also demonstrated point zero to be as measured with a working volume of 20cm x 19cm x 20cm. This procedure of working volume definition was repeated once monthly as a data quality control measure.

3.3.1.4. External Location of the Target Zone

It was necessary to quickly place the subject in the target zone to allow optimum use of a young child's limited attention span and short period of co-operation. Low intensity slit lamps were attached to the colour camera on each pod and positioned such that the intersecting beams formed a cross on the subject's forehead when the subject was correctly located in the target zone.

3.3.1.5. Software and Additions

The C3D™ software developed by the Turing Institute and Faraday Laboratory in Glasgow University Computer Science Department required further modification to allow it to be used with young children. As the subject was not restrained during imaging, the time lag between monochrome and colour camera exposure had to be shortened to 30 milliseconds from 200 milliseconds. This required changes to be made to the activation and co-ordination of the cameras and flashes. No changes to the calibration process or matching of stereoimages were required.

The images that produced the range data, the 3D map of the facial surface, were captured 30 milliseconds before the colour images that were used to identify anthropometric landmarks. It was necessary to ensure, therefore, that unobserved movement of the subject had not occurred in the intervening milliseconds. To ensure that both sets of images were co-incident an additional programme, Check Align™, was written to allow the colour image to be laid over the monochrome image on screen. If intrinsic or extrinsic movement of the face had occurred the overlying images appeared blurred and that set of images was discarded. It was also possible to digitise the same landmark on the monochrome and colour images separately and then superimpose the images to ensure that the crosses marking each landmark co-incided.

3.3.1.6. Validation of the C3D™ System

A three phase procedure was performed to estimate the system error for landmark extraction.

Prior to the commencement of this study, basic performance tests had been performed but accuracy of the C3D™ system had not been verified in the field. The design and implementation of an active videometrics system for an imaginary clinical study of craniofacial surgery had included testing of the matching algorithm of the C3D™ software and accuracy analysis of the complete static calibration process. The conclusion had been that "the obtained results give encouragement that the required accuracy of 1.0 mm RMSE (root mean square error) can be obtained" (Urquhart, 1997).

3.3.1.6.1. System Error Estimation - Phase 1

Aim – to assess the accuracy and reproducibility of the computerised stereophotogrammetry process from image capture to landmark co-ordinate extraction

Material and Methods - Twenty-one casts of infant faces with 5 inkspots/landmarks were photographed twice in four positions using the C3D™ computerised stereophotogrammetry system. The position of the casts relative to the cameras was varied along the X-axis in 10 centimetre increments or rotated around the Y-axis in 20-degree increments. A single image of each cast in each of the four positions was digitised by three operators once (84 images x 3). Landmark digitisation was performed in a random subset twice (8 images x 3). A further set of random duplicate images was also digitised (8 images x 3).

Co-ordinate configurations recorded by the C3D™ system were compared with those recorded by a co-ordinate measuring machine (CMM) in the Department of Mechanical Engineering, University of Birmingham, England. The latter data collected by this piezo-electric, semi-automated, rigid-framed digitiser were taken as the gold standard.

Using Generalised Partial Procrustes analysis, the 3D co-ordinate configurations were compared and displacement from the CMM recorded configurations estimated. Procrustes Analysis is a method of superimposition that aligns configurations of landmarks to a position of maximal agreement by rotating, translating and scaling the configurations (Dryden & Mardia 1998). The geometrical relationship of the landmarks, the shape, is preserved in this superimposition method. The system error and operator error in landmark identification were quantified.

Results - The system error (accuracy) of the C3D™ method of landmark co-ordinate identification was 0.79 mm (SD 0.57). The operator error was 0.23 mm (SD 0.35). Details of error estimation are recorded in Appendix 4

3.3.1.6.2. Assessment of the Effect of Head Position - Phase 2

Aim – to assess the effect of head tilt on the integrity of the range data

Materials and Methods - A doll's face was marked with 12 crosses using a ballpoint pen of 1 mm tip diameter (*Bic*, medium). The crosses covered a wide area of the face (Figure 3.7.). The doll's head was photographed twice in each of 9 positions. The starting position was with the Frankfort plane parallel to the floor. Then the head was tilted around an axis in the doll's neck above and below the starting position in 10-degree increments, covering a range of 80 degrees. The landmarks were digitised twice by a single operator and the 3D co-ordinates extracted. Interlandmark distances were also calculated

The distances and 3D co-ordinate configurations recorded at different degrees of head tilt were compared. The alteration in image quality with alteration in head tilt was assessed subjectively. The alteration in the range data with alteration in head position was quantified.

Results - The operator error in landmark digitisation was 0.20 mm (SD 0.09). Relative to the configuration recorded with the head positioned with the Frankfort plane parallel to the ground, the error in landmark location including the above operator error was 0.34 mm (SD 0.13) over 60 degrees of variation. At 40 degrees rotation away from the starting point, the error increased to 1.12 mm (SD 0.22) at minus 40 degrees and 0.79mm (SD 0.45) at plus 40 degrees. The operator error also increased to 0.64 mm (SD 0.22) and 0.40 mm (SD 0.17). The distances calculated between landmarks remained constant at all degrees of rotation. See Appendix 4 for full details of error estimation.

For the main study of facial morphology, the subjects were photographed with the Frankfort Plane parallel to the floor.

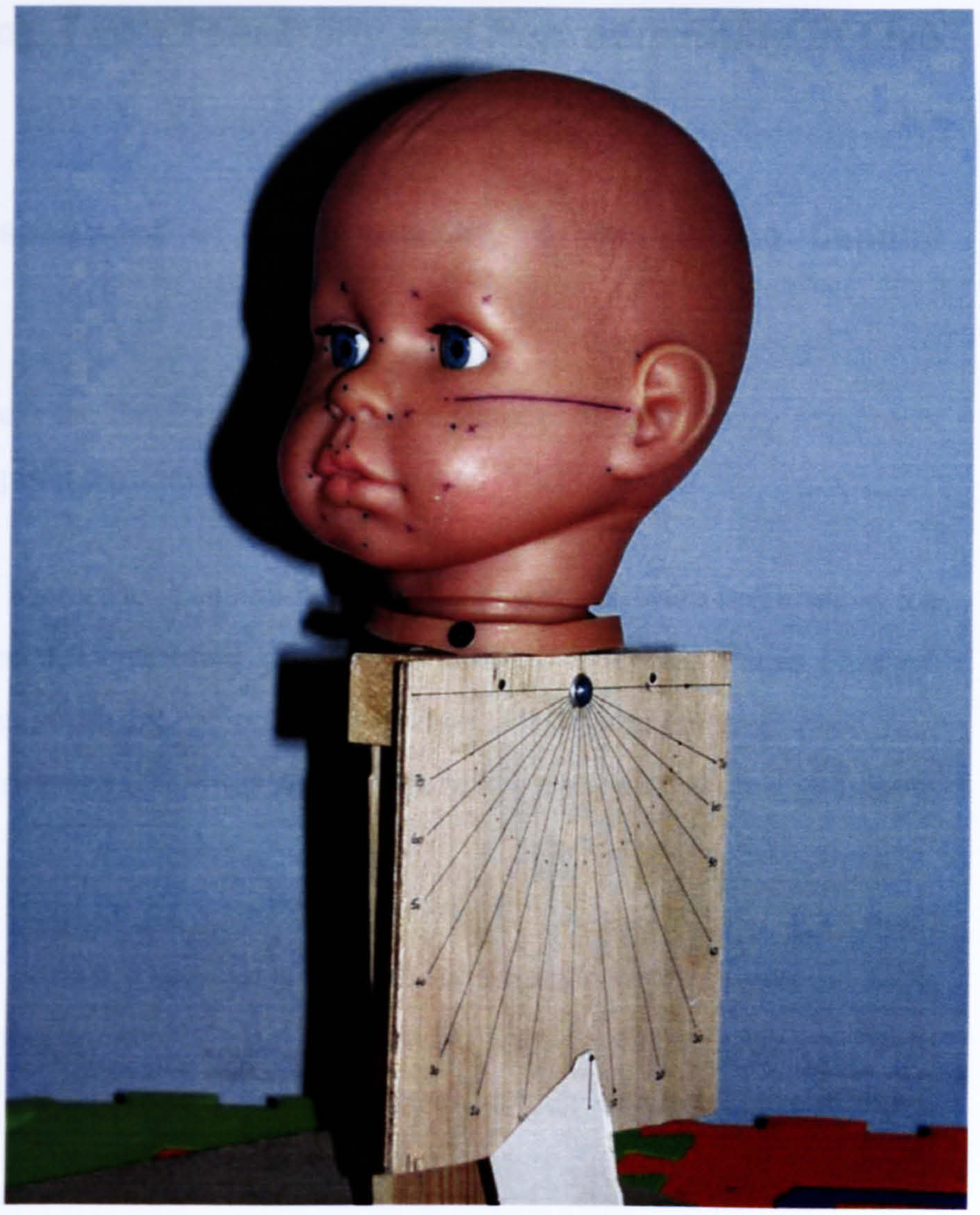


Figure 3.7. Doll's Head with Landmarks in situ - Pilot Study - Phase 1

3.3.2. Study Two - Feasibility and Reproducibility in Live Subjects

3.3.2.1. Assessment of the Feasibility of the Image Capture Process

Aim - to assess the feasibility of using the stereophotogrammetry equipment in the imaging of a paediatric study population

Materials and Methods - Ten children, aged 3 to 6 years, were recruited in the Paediatric Dental Department of Glasgow Dental Hospital. These children were not included in the project study groups. They were photographed four times at rest using the C3D™ system, twice before and twice after a 30-minute break.

Results - All children co-operated with the image capture process with minimal encouragement. Children showing low levels of anxiety were photographed sitting on a guardian's knees, which ameliorated any overt resistance. No child declined to be photographed.

Following this data set acquisition trial, a light safety harness was attached to the dental chair to be worn by the child during photography (Figure 3.8.). This was to ensure that if a child attempted to leave the chair unaided, his / her progress would be slowed by the harness until the researcher or guardian could step from behind the camera pods to assist him / her.

3.3.2.2. Assessment of the Stability of the Resting Face in Young Children

Aim – To assess the stability of the resting face in young children

Materials and method – The same children described above (Section 3.3.2.1.) had 20 cross inkmarks placed on their faces with an eyeliner pen of 1mm

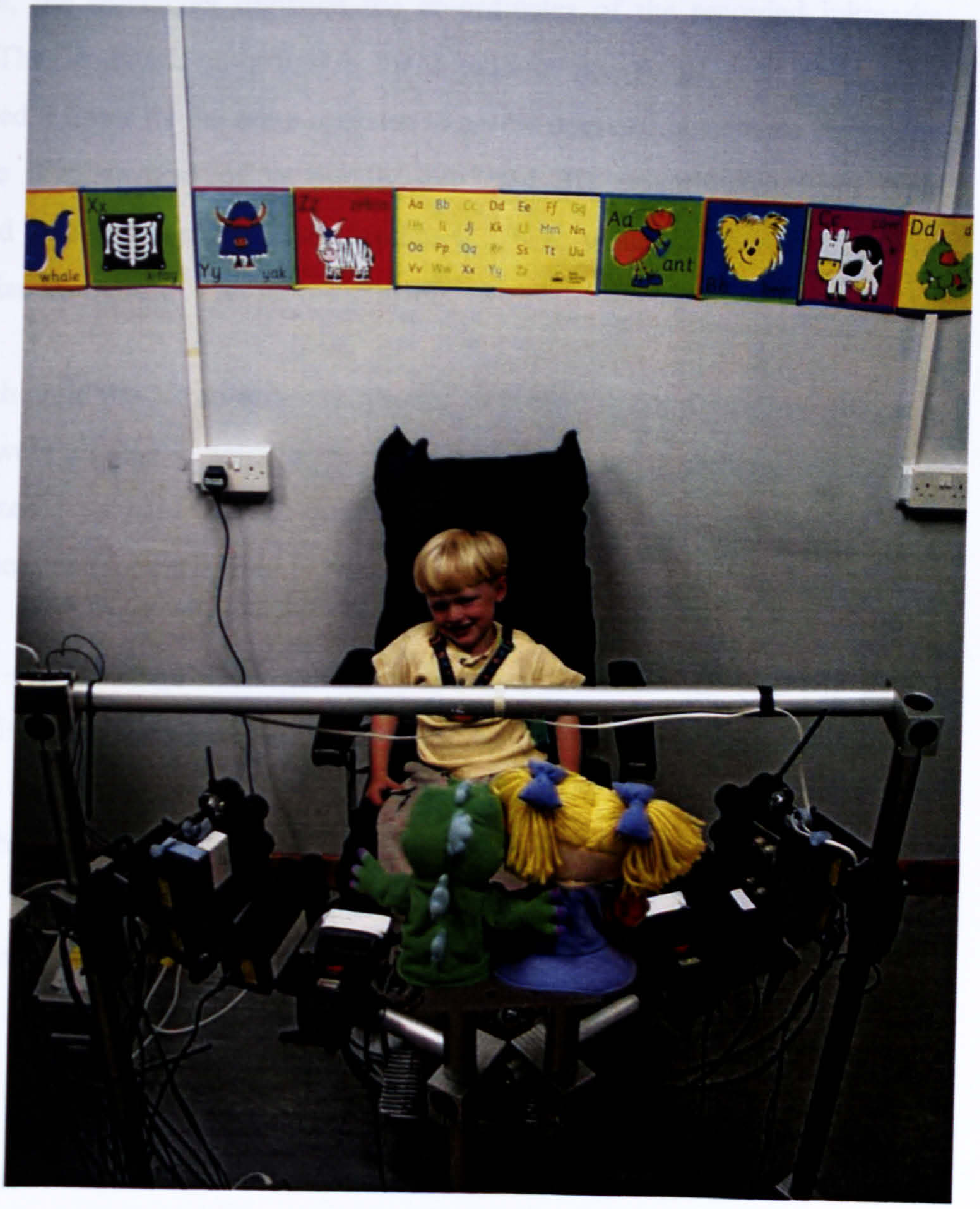


Figure 3.8. Subject seated for Image Capture

diameter (Rimmel, U.K.) (Figure 3.9.). On the images collected by this process, the researcher digitised the co-ordinates of the recorded inkmarks once. The landmark co-ordinates for 2 random images of each child were extracted 3 times by the same operator to assess operator placement error. The average displacement of repeatedly extracted 3D co-ordinates from their centroid was calculated for each landmark. These values were averaged over all the landmarks and over all the subjects.

For each child the 3D co-ordinate systems or landmark configurations for each image were aligned in pairs to maximum agreement using Generalised Partial Procrustes Analysis. The co-ordinate systems were compared within and across sessions for each child.

Results - The average within landmark standard deviation was 0.78 (SE 0.44) for the images taken 1 minute apart and 0.63 (SE 0.36) for images taken 30 minutes apart. The overall within landmark standard deviation was 0.77 (SE 0.29) which was equivalent to an average distance of 1.09 mm. The associated operator error was 0.22 mm (SD 0.10). See Appendix 4 for details of error estimation.

3.3.2.3. Assessment of the Reproducibility of the Smiling Image in Children

Aim – To assess the reproducibility of the maximal smile expression in children

Materials and Method - Fifteen cases, 5 cleft and 10 non-cleft, of multiple smiling images were chosen at random from the study samples.

Recorded smiles were considered acceptable if the following conditions were fulfilled:-

- The exocanthion points were obscured indicating narrowing of the

Figure 3.9. Subject with Inkmarks in situ - Pilot study

palpebral fissures.

- The christae philtrae were flattened and not discernible.
- The teeth were in occlusion.
- The first primary molars were visible between the open lips.

A set of 13 anthropometric landmarks, known to have a low element of operator inconsistency, was chosen. For each child, 2 images of similar expressions were digitised twice by the researcher. The anthropometric landmarks are shown in Figure 3.10. The recorded configurations of 3D co-ordinates were compared between duplicate mark-ups and duplicate images for each child.

Results - The reproducibility of maximal smile in young children was comparable to that in adults with 1.03 mm (SD 0.58) being the average displacement of landmarks on repetition of facial expression (Trotman et al. 1998a)The operator error was 0.45 mm (SD 0.27). See Appendix 4 for details of error estimation.

3.3.2.4. Assessment of Facial Landmark Digitisation Reproducibility

Aim – To assess anthropometric landmark digitisation reproducibility

Materials and Methods - Images of 12 research subjects, at rest, were chosen in each of the following categories, 3-year-olds with clefts and 3-year-olds without clefts. The researcher digitised 50 anthropometric landmarks 3 times on each image. The average displacement of repeatedly extracted 3D co-ordinates from their centroid was calculated for each landmark. The landmarks were ranked in order of ascending operator placement inconsistency (Table

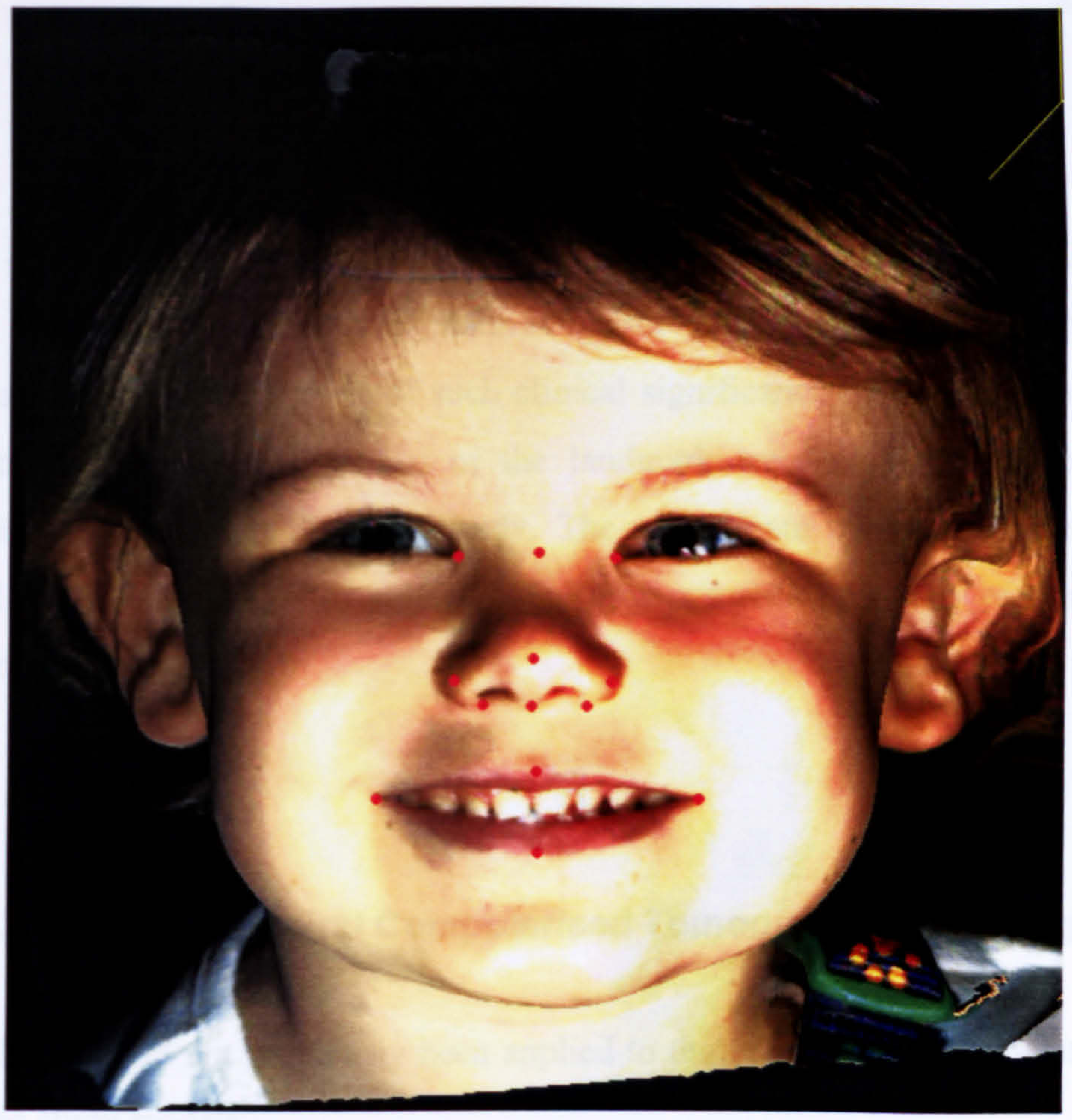


Figure 3.10. Landmarks utilised in Assessment of Smile Reproducibility

3.1.).

Results - A set of 25 landmarks was chosen with operator inconsistency of 0.5 mm or less to be part of the definitive data set. In cases where the anthropometric landmarks were of such clinical significance that it could not be omitted, duplicate digitisations of the landmark were performed when identifying landmarks in the main study. The co-ordinate of the calculated centroid was extracted to reflect the approximate position of the anthropometric landmark.

3.3.3. Study Three

3.3.3.1. Validation of Spiral Computerised Tomographic Scanning

This technique had not previously been applied to the investigation of dentate paediatric study models.

Aim – To assess the accuracy and reproducibility of spiral CT scanning of dental study models.

Materials and Methods - The dental study models were arranged on a framework of polystyrene sheets (Figure 3.11.) with rigid plastic cups as spacers for scanning.

The following settings were selected:

- Field of view - 250 mm
- Matrix - 512 x 512 pixels
- Imaging time - 80 to 100 seconds
- Slice size - nominally 0.5mm, effectively 0.6mm
- Reconstruction slice size - nominally 0.25 mm, effectively 0.3mm.
- Pitch - 0.873 (overlapping helix)
- Rotation time - 0.75 seconds.

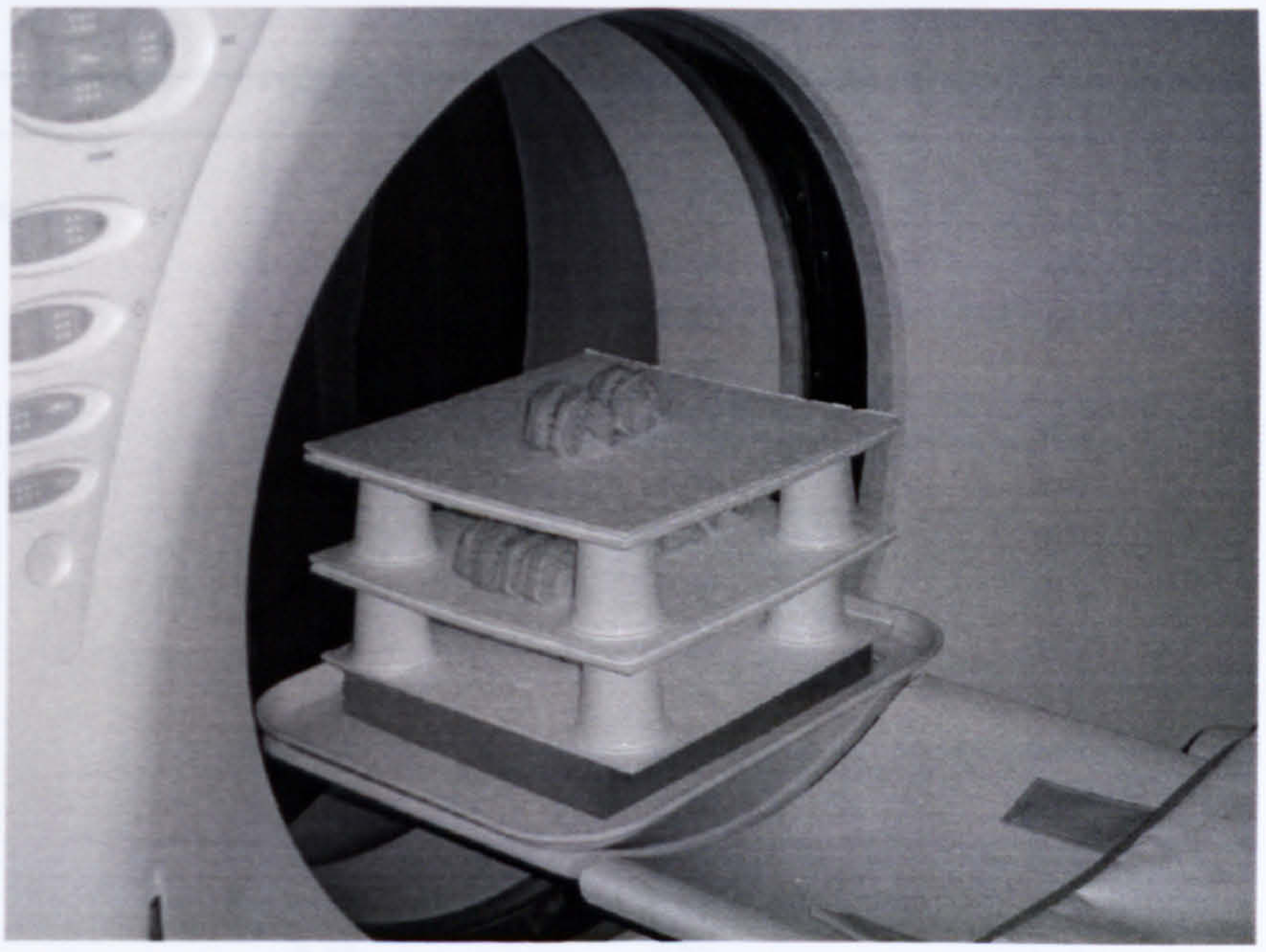


Figure 3.11. Study models in CT scanner

Landmarks	Clefts	Controls	Landmarks
< 0.5 mm inconsistency			
cR	0.23	0.26	cR
li	0.31	0.29	cL
al'Li	0.31	0.30	li
al'Ri	0.32	0.32	stoi
enL	0.32	0.33	sn'L
cphR	0.36	0.33	chL
enR	0.38	0.34	stos
cL	0.39	0.34	al'Li
exR	0.39	0.35	enL
chR	0.39	0.35	enR
stos	0.42	0.36	sbalR
sn'L	0.44	0.36	al'Ri
stoi	0.45	0.36	chR
exL	0.45	0.37	sbalL
sn'R	0.49	0.38	exR
sbalR	0.49	0.38	cphR
chL	0.49	0.39	cphL
sn	0.49	0.39	ls
obiL	0.49	0.40	obiR
cphL	0.50	0.41	sn'R
ls	0.50	0.42	exL
sl	0.50	0.44	prn
sbalL	0.50	0.45	sl
		0.46	acL
		0.46	sn
0.5 - 0.71 mm inconsistency			
acL	0.50	0.53	alR
al'Lo	0.51	0.53	obiL
obiR	0.60	0.55	acR
n	0.61	0.62	alL
acR	0.63	0.62	al'Lo
al'Ro	0.64	0.68	n
prn	0.64	0.69	al'Ro
alR	0.68		
alL	0.71		
> 0.71 mm inconsistency			
pg	0.93	0.82	pg
tL	0.95	0.87	tL
tR	1.10	1.41	gn
gn	1.25	1.66	chkR
chkR	2.01	1.99	chkL
chkL	2.39	2.42	tR
goR	4.87	5.02	goR
goL	5.02	5.12	goL

Table 3.1. Operator Inconsistency in Digitisation of Anthropometric Landmarks

- Kilovoltage - 120
- Ma per slice - 150.
- Scanning was at ultra high resolution and reconstruction used a bony algorithm filter.

One batch of 12 model pairs was scanned twice and the standard landmark set digitised. Another batch of scanned models was digitised twice. The 3D co-ordinate configurations were compared for duplicate scans and duplicate digitisations to quantify the system reproducibility and the operator reproducibility in digitising landmarks.

One sample Student's *t* tests were used to compare the intercanine distances, calculated for the scanned study models, with the intercanine distances measured directly using digital calipers. The aim was to test if each difference was statistically different from zero and quantify the accuracy of landmark extraction from spiral CT scanned study models with the values measured directly, the latter taken as the gold standard. Regression analysis of the output of both forms of measurement was performed.

3.4. Main Study

3.4.1. Data Collection for Each Subject

All the subjects were examined by the researcher at Glasgow Dental Hospital. Each visit took approximately 30 minutes and the data were collected according to the following protocol. The order of collection was designed to ensure optimal use of the child's short attention span and limited co-operation. The dental impressions were recorded at the end of the session, as this was the procedure with which the child was least likely to comply.

3.4.1.1. Photography of the Face at Rest

For photography each subject was positioned as follows: -

- Sitting alone wearing a safety harness which was secured to the heavily padded dental chair
- Head resting against a soft padded back support with a concavity to accommodate the occiput
- Facing straight ahead with the Frankfort plane parallel to the floor
- Lips, approximated lightly, with no flattening of the chin. Some of the children were obligate mouth breathers, either due to cleft-related ENT problems or malocclusion and short lips, so it was necessary to settle for close approximation of the lips, instead of a lip seal.

Four to five images of the subject at rest were recorded, including one with the dental chair rotated to face a single camera pod only. This frontal view of the face allowed subsequent subjective comparison of the 3D reconstruction of the face with a 2D full-face colour photograph taken with the single pod.

3.4.1.2. Recording Maximal Smile

The subject remained positioned as above and was requested to perform a maximal smile in response to a standard verbal cue: "big smile for the camera, show me all your front teeth". Where possible, 4 to 5 images of a maximal smile were recorded to allow assessment of reproducibility. For a description of this assessment, see section 3.3.2.3. While the researcher sought the subject's full attention and co-operation, there was no attempt to encourage the subject to laugh spontaneously.

The recorded images were considered acceptable if the conditions described in section 3.3.2.3. were fulfilled. The success rate of recording an acceptable smiling image acquisition was 73%.

3.4.1.3. Height and Weight Measurement

The clothed subject, without shoes, was weighed in kilograms (to 1 decimal place) using the stand-on scales as detailed previously. The standing height of each subject was measured in centimetres using a wall-mounted telescopic measuring rod.

3.4.1.4. Dental Arch Recording and Bite Registration

Not all the children enrolled in the study co-operated with this aspect of the data collection which was performed last during the child's visit to the Dental Hospital.

The success rate of acceptable impression acquisition was 85%.

3.4.1.5. Spiral CT Scanning of Study Models

This last stage of data collection was carried out when all of the children in the study had been seen and stone study models were available, and not after each individual visits.

The study models were scanned in a single session in batches of 24, comprising 12 sets of maxillary and mandibular casts. The settings of the scanner were as listed in section 3.3.3.1.

3.4.2. Data Editing

3.4.2.1.Building and Editing of Three-Dimensional Soft Tissue Images

The set of images of each child was built to a resolution of 0.005mm using the C3D™ software. The extraneous detail surrounding the child's head was cropped from the image on screen and the remaining area was rebuilt to a resolution of 0.002 mm. The images were converted to VRML .

3.4.2.2. Processing of Three-Dimensional CT Scanned Casts

The DICOM output of the spiral CT scanner was segmented and volume rendered using AMIRA™ and converted to VRML. The software code was rewritten for each VRML image to render it compatible with the experimental Facial Analysis Tool.

3.4.2.3. Landmark Extraction

3.4.2.3.1. Facial Soft Tissue Images

A single operator digitised the VRML images. The co-ordinates of the anthropometric landmarks shown in Figures 3.12. - 3.14. were recorded on images of the resting face.

Facial Analysis Tool

The position of the face on screen was standardised by creating a plane, using the inner canthi and the midline of the sub labial fold, and setting this created plane parallel with the screen surface. The landmarks were identified according to the following schedule of planar tilts, :

Plane parallel to screen	ex, en, n, prn, ls, ch, cph, sto, sl
15 degrees positive rotation around X axis	sbal, sn

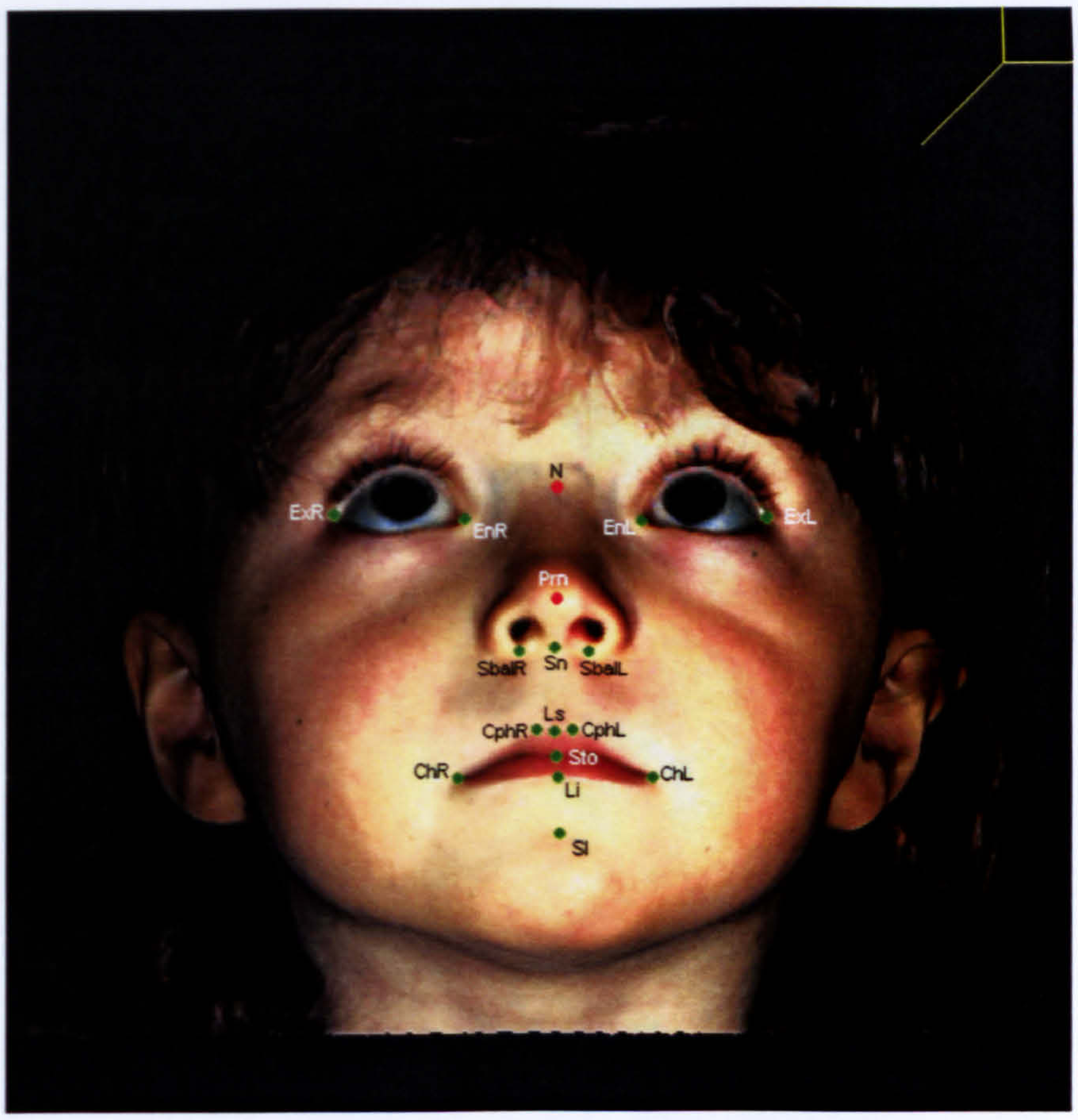


Figure 3.12 Face at rest with anthropometric landmarks in situ – frontal view

Illustrated in green – digitised once

ExR	Right exocanthion	ExL	Left exocanthion
EnR	Right endocanthion	EnL	Left endocanthion
SbalR	Right subalare	SbalL	Left subalare
CphR	Right christae philtri	ChR	Right cheilion
CphL	Left christa philtri	ChL	Left cheilion
Sn	Subnasale	Ls	Labrale superiorus
Sto	Stomion	Li	Labrale inferiorus

Illustrated in red – digitised twice

N	Nasion	Prn	Pronasale

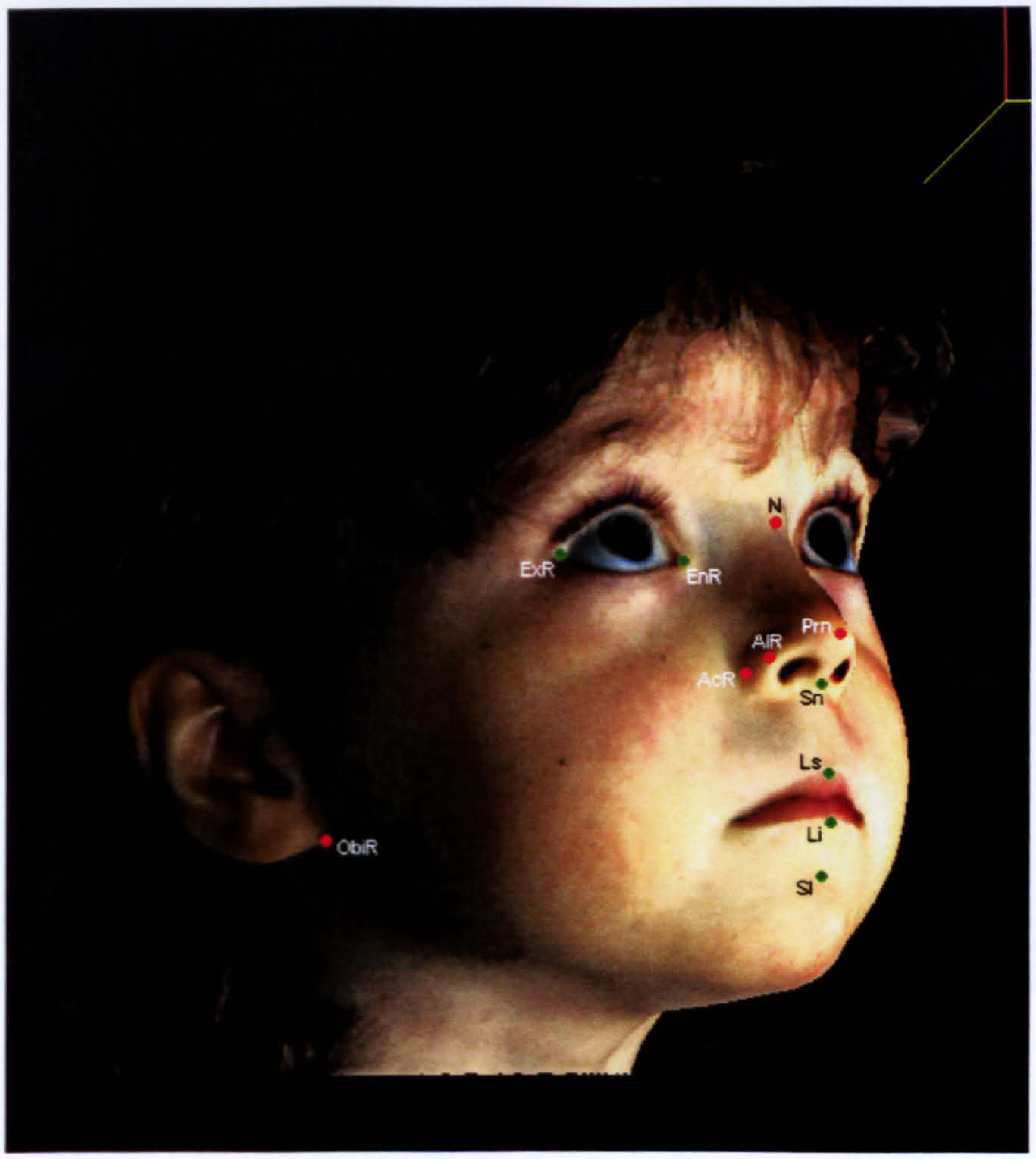


Figure 3.13 Face at rest with anthropometric landmarks in situ - from right

Illustrated in green – digitised once

ExR	Right exocanthion	EnR	Right endocanthion
Sn	Subnasale	Ls	Labrale superiorus
Li	Labrale inferiorus	Sl	Sublabialis

Illustrated in red – digitised twice

N	Nasion	Prn	Pronasale
AlR	Right alare	AcR	Right alar curvature
ObiR	Otobasion inferorus		

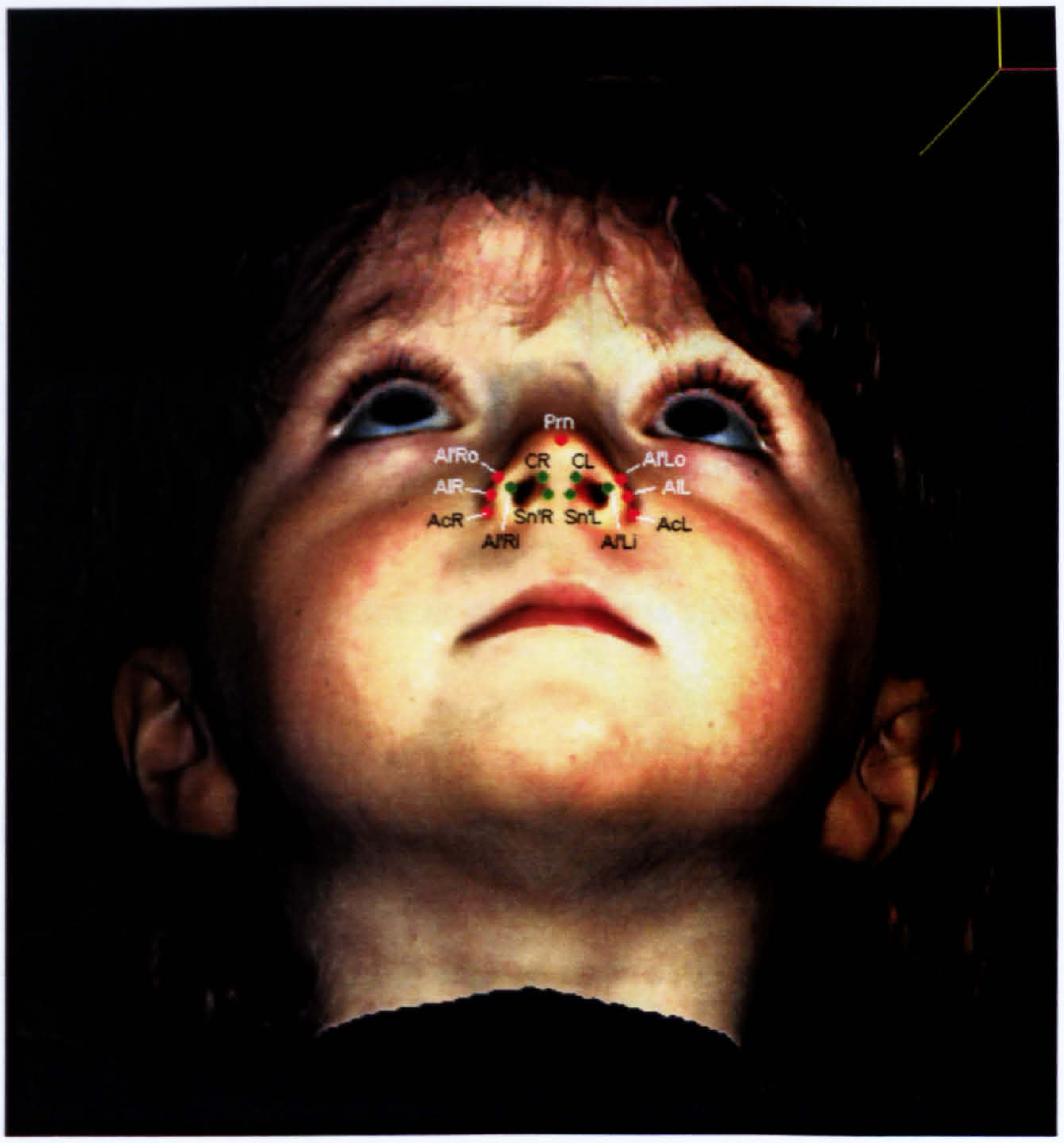


Figure 3.14. Face at rest with anthropometric landmarks in situ – worm's eye view

Illustrated in green – digitised once

CR	Right columella high point	Sn'R	Right subnasale'
CL	Left columella high point	Sn'L	Left subnasale'
Al'Ri	Right inner alare'	Al'Li	Left inner alare'

Illustrated in red – digitised twice

AcR	Right alar curvature	AlR	Right alare
AcL	Left alare curvature	AlL	Left alare
Al'Ro	Right outer alare'	An'Lo	Left outer alare'
Prn	Pronasale		

30 degrees positive rotation around X axis	al', ac, al
45 degrees positive rotation around X axis	c, sn'
90 degrees positive and negative rotation around Y axis	obi

Landmark placement was aided by simultaneous display of two smaller views of the face on screen. The first smaller image was displayed at 90 degrees rotation around the Y-axis and the second smaller image was shown rotated 90 degrees around the X-axis relative to the position of the main VRML image on screen.

An example of the screen during landmark placement is shown in Figure 3.15.

On images of the smiling face, the landmarks illustrated in Figures 3.16. - 3.18. were digitised and the 3D co-ordinates recorded. The landmarks illustrated in blue were digitised once, those illustrated in yellow were digitised twice and the calculated centroid was recorded as the landmark co-ordinates.

3.4.2.3.2. Dental Study Model Images

On images of the study models, the landmarks illustrated in Figure 3.19. were digitised and the 3D co-ordinates recorded. The details of the occlusal surface between the cusp tips was not sufficient to permit identification of the fossae, so cusp tips and incisal edges of all erupted teeth were chosen to define the dental arch shape. The landmarks were digitised with the study model images arranged on screen, in pairs, with the occlusal plane parallel to the plane of the screen. The location of the digitised landmarks was reviewed through two other windows on screen showing the models from the labial and buccal viewpoints.

For right-sided clefts, the maxillary and mandibular study models were digitised such that right-sided landmarks were labelled left and vice versa. This was to ensure that all clefts appeared to occur on the left side of the maxillary arch. Thus, details of the dental arch in the area of the cleft were not

Figure 3.15. Images on Screen during Landmark Digitisation

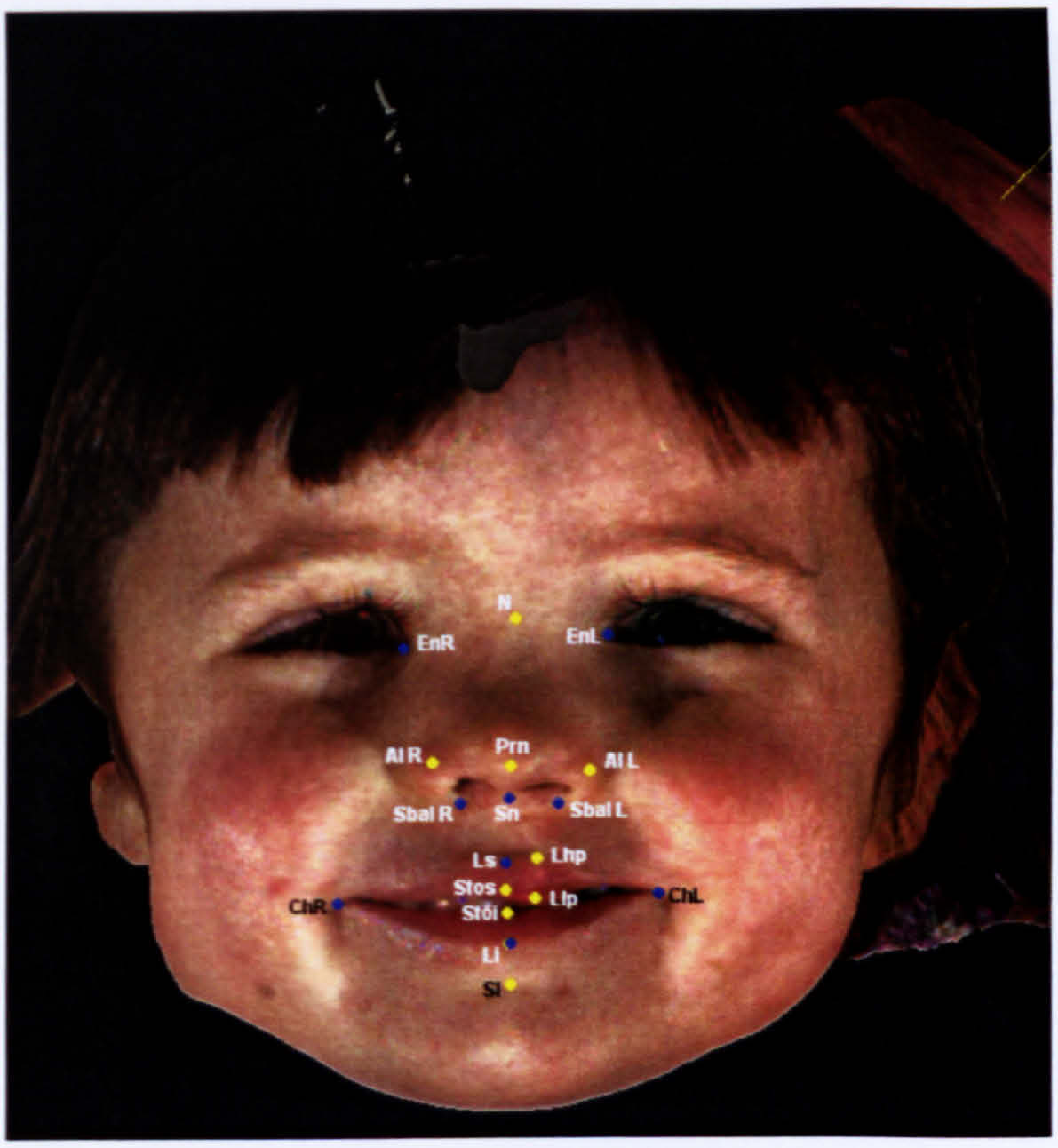


Figure 3.16 Smiling Face with anthropometric landmarks in situ – frontal view

Illustrated in blue – digitised once

EnR	Right endocanthion	EnL	Left endocanthion
SbalR	Right subalare	SbalL	Left subalare
ChR	Right cheilion	ChL	Left cheilion
Sn	Subnasale	Ls	Labrale superiorus
Li	Labrale inferiorus		

Illustrated in yellow – digitised twice

N	Nasion	Prn	Pronasale
Lhp	Lip high point	Llp	Lip low point
AlR	Right alare	AlL	Left alare
Stos	Stomion superiorus	Stoi	Stomion inferiorus
Prn	Pronasle	N	Nasion
Sl	Sublabialis		

Figure 3.17. Smiling Face with Anthropometric Landmarks in situ – submento-vertex view

Illustrated in blue – digitised once

CR	Right columella high point
Sn’R	Right subnasale dash
CL	Left columella high point
Sn’L	Left subnasale dash

Illustrated in yellow – digitised twice

AcR	Right alar curvature
AcL	Left alar curvature

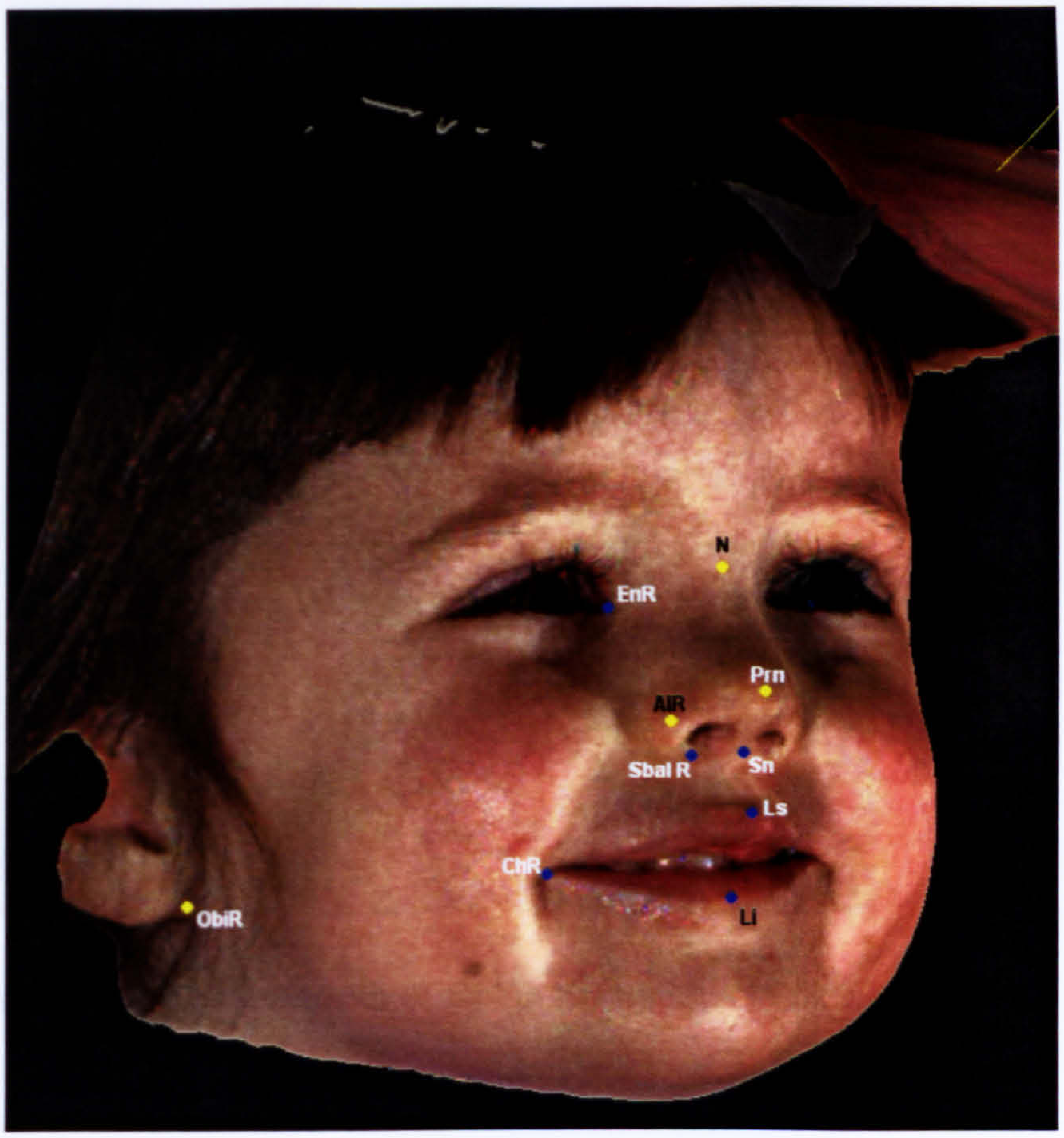


Figure 3.18. Smiling Face with Anthropometric Landmarks in situ – viewed from right side

Illustrated in blue – digitised once

EnR	Right endocanthion	Sn	Subnasale
SbalR	Right subalare	ChR	Right cheilion
Ls	Labrale superiorus	Li	Labrale inferiorus

Illustrated in yellow – digitised twice

Obi R	Right otobasion inferiorus	Prn	Pronasale
AlR	Right alare	N	Nasion

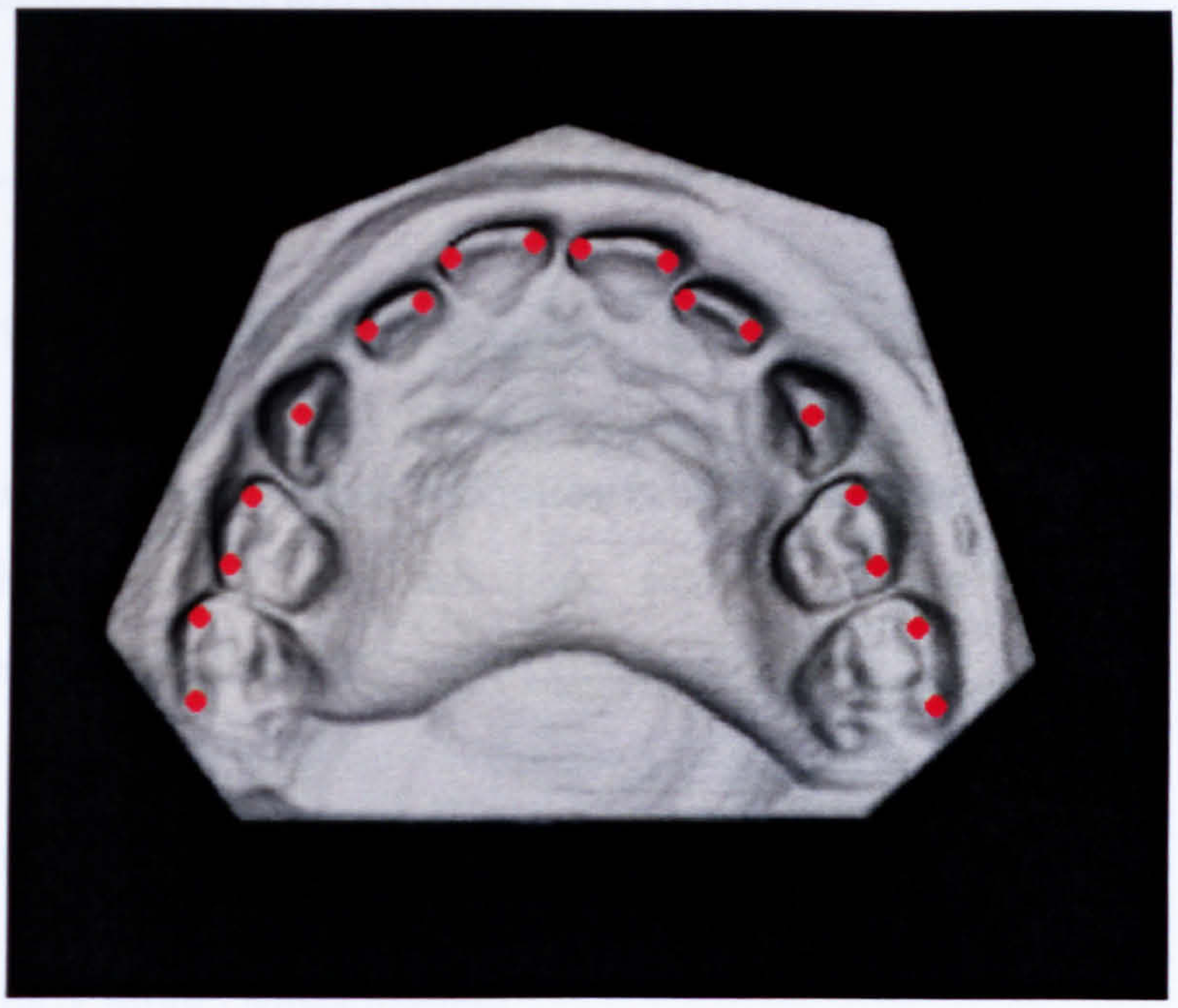


Figure 3.19. Study Model with Cusp Tip Landmarks in Situ

lost when the average arch form for each type of cleft was constructed from the collection of right and left-sided cleft arch forms.

3.4.2.4. Measurement of Linear Dimensions

3.4.2.4.1. Facial Soft Tissue Images

The linear distances, angles and ratios between landmarks on the resting face, shown in Table 3.2. were calculated. For the smiling faces, interlandmark distances listed in Table 3.3. were calculated.

3.4.2.4.2. Study Models

Direct Study Model Measurement

For the study models, the linear dimensions were measured directly. These directly measured distances were the basis for the analysis of arch size. The intercanine distances, calculated for the images of study model produced by spiral CT scanning, were employed to assess accuracy of the technique only.

The maxillary and mandibular distances were measured with a digital caliper in triplicate and the averaged dimensions were recorded.
The intercanine width was measured between the canine tips. When attrition of the tip had occurred, the width was measured between the palatal edge of the flattened occlusal surface. The first and second intermolar widths were measured between the mesial fossae of the primary molar teeth. (Figure 3.20.)
The arch depth was the shortest distance measured between the mesioincisal corner of the central incisor and a metal ruler held against the distal aspect of the second molars. The depth was measured on both sides and the average recorded (Figure 3.21.).
To generate the arch circumference, the distal quadrant lengths (from the distal aspect of the second molar to the distal aspect of the canine) and the mesial quadrant lengths (from the distal aspect of the canine to the mesial aspect of the central incisor) were added together with the distance measured between

Interlandmark Distances	Landmarks
Binocular width:	exL - exR
Right ocular width:	exR - enR
Left ocular width:	exL - enL
Intercanthal width:	enR - enL
Upper face height:	n - sto
Midface prominence:	sl - n - sn
Right lower naso-aural distance:	n - obiR
Left lower naso-aural distance:	n - obiL
Right lower subnasale-aural distance:	sn - obi R
Left lower subnasale-aural distance:	sn - obi L
Nose height:	n - sn
Nose bridge length:	n - prn
Nasal protrusion	sn - prn
Nasolabial angle:	prn - sn - ls
Nasal tip angle	n-prn-sn
Soft tissue nose width	alL - alR
Length of right ala:	acR - prn
Length of left ala:	acL - prn
Right alar thickness:	al'Ri to al'Ro
Left alar thickness:	al'Li to al'Lo
Anatomical width of nose:	acR - acL
Nasal Base Width	sbalR-sbalL
Width of right nostril floor:	sbalR - sn
Width of left nostril floor:	sbal L to sn
Width of the columella:	sn'R - sn'L
Length of columella (right):	cR - sn
Length of columella (left):	cL - sn
Soft tissue nose to mouth width ratio	alR-alL:chR-chL
Anatomical tissue nose to mouth width ratio	acR-acL:chR-chL
Mouth width:	chR - chL
Upper lip height:	sn - sto(s)
Upper cutaneous lip height:	sn - ls
Upper vermillion lip height:	ls-sto(s)
Lower lip height:	sto(i) - sl
Lower cutaneous lip height:	li - sl
Lower vermillion height:	sto(i) - li
Width of the philtrum:	cphR - cphL
Right hemiphiltrum width	cphR-ls
Left hemiphiltrum width	cphL-ls
Right upper lip length	cphR-chR
Left upper lip length	cphL-chL
Right lower lip length	li-chR
Left lower lip length	li-chL

Table 3.2. Interlandmark Distances measured on the Image of the Face at Rest

Interlandmark Distances	Landmarks
Intercanthal width:	enR - enL
Upper face height:	n - sto
Midface prominence:	sl - n - sn
Right lower naso-aural distance:	n - obiR
Left lower naso-aural distance:	n - obiL
Right lower subnasale-aural distance:	sn - obi R
Left lower subnasale-aural distance:	sn - obi L
Nose height:	n - sn
Nose bridge length:	n - prn
Nasal protrusion	sn - prn
Nasolabial angle:	prn - sn - ls
Nasal tip angle	n-prn-sn
Soft tissue nose width	alL - alR
Length of right ala:	acR - prn
Length of left ala:	acL - prn
Anatomical width of nose:	acR - acL
Nasal Base Width	sbalR-sbalL
Width of right nostril floor:	sbalR - sn
Width of left nostril floor:	sbal L to sn
Width of the columella:	sn'R - sn'L
Length of columella (right):	cR - sn
Length of columella (left):	cL - sn
Soft tissue nose to mouth width ratio	alR-alL:chR-chL
Anatomical tissue nose to mouth width ratio	acR-acL:chR-chL
Mouth width:	chR - chL
Upper lip height:	sn - stos
Upper cutaneous lip height:	sn - ls
Upper vermillion lip height:	ls-stos
Lower lip height:	stoi - sl
Lower cutaneous lip height:	li - sl
Lower vermillion height:	stoi - li
Right upper lip length	ls-chR
Left upper lip length	ls-chL
Right lower lip length	li-chR
Left lower lip length	li-chL
Maximum upper vermillion height	lhp-llp
Maximum upper vermillion height to midline	lhp-ls

Table 3.3. Interlandmark Distances measured on the Image of the Smiling Face

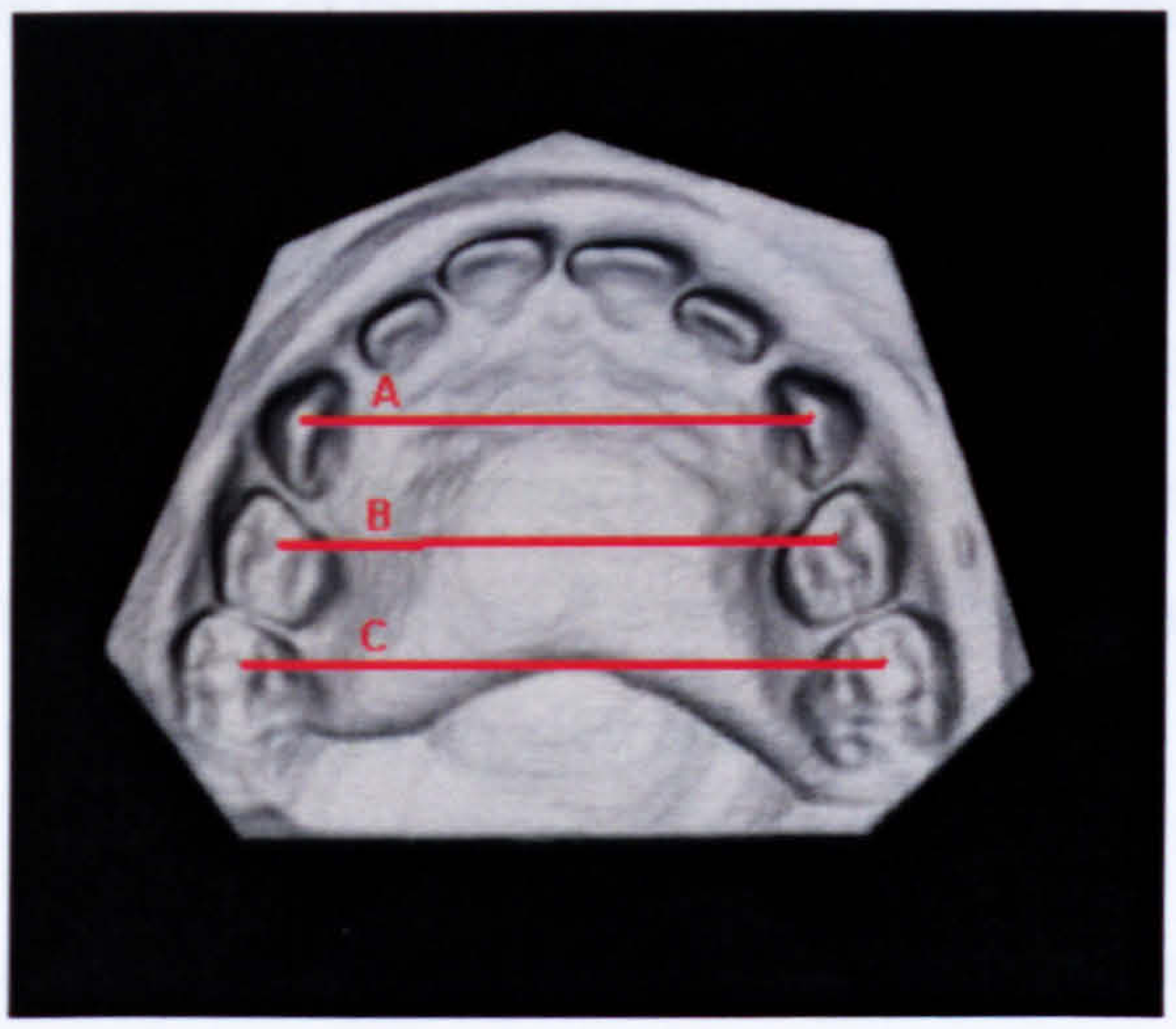


Figure 3.20. Arch Widths Illustrated on Cast

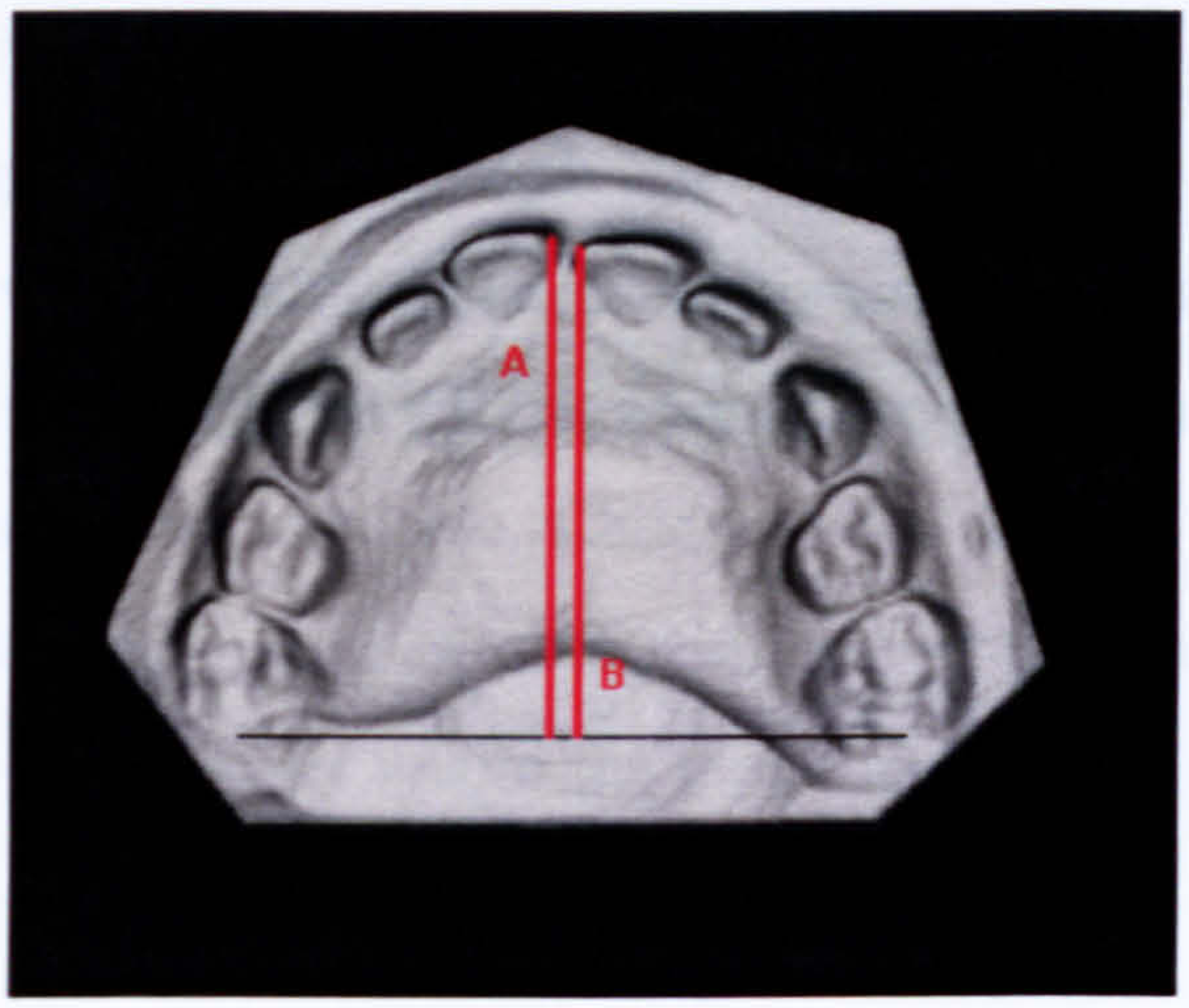


Figure 3.21. Arch Depth Illustrated on Cast

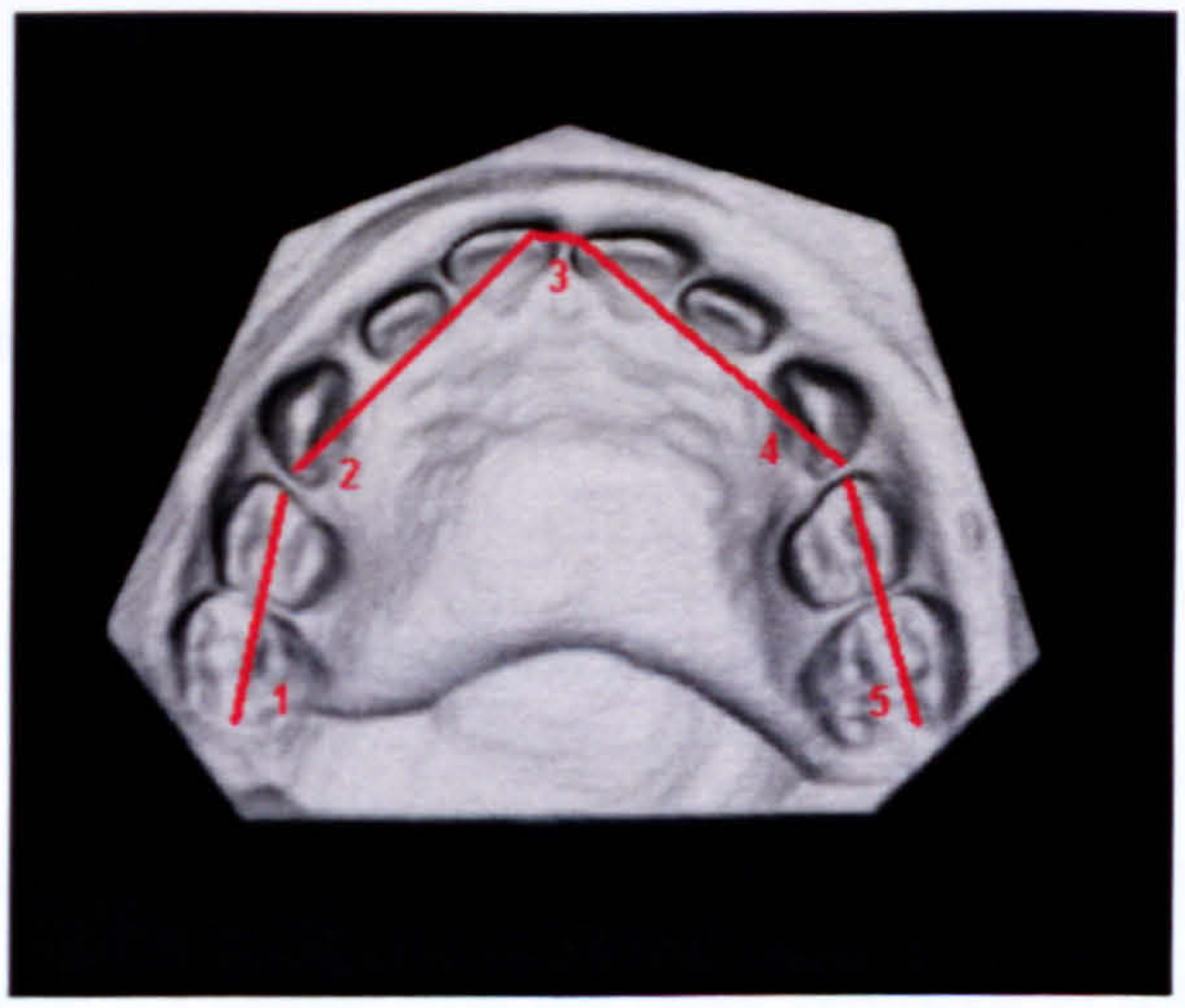


Figure 3.22. Arch Circumference Illustrated on Cast

the mesial aspects of the central incisors where a space existed.

Figure 3.22. shows the landmarks and distances and components of circumference, illustrated on a CT scanned cast. For all children with clefts, the dimensions were recorded for affected and unaffected sides. The affected side was designated left and the unaffected side designated right, regardless of the side of the face that the cleft affected.

Molar and incisor relationships were recorded for each case and classified according to Angle's classification and British Standards Institute classification respectively. The score of decayed, missing and filled teeth (DMF) was recorded. The presence of crossbites and supernumerary teeth, including the number of teeth affected were noted. Some children did not co-operate with the mandibular arch impression and for these children, the molar and incisor arch relationships were recorded as Class O as only the maxillary arch model was available. Children who did not co-operate with two arch impression did not co-operate with intra-oral examination either. It was not possible to clinically record the molar and incisor arch relationships in these children.

3.4.3. Statistical Analysis

3.4.3.1. Height and Weight of Study Samples

The height and weight of the subjects in the study and control samples were compared using a two sample Student's *t* test.

3.4.3.2. Effect of Changes in Lip Pose

A selection of linear measurements of the resting control faces, with the lips closed, was compared with the same selection of measurements of the resting control faces, captured with the lips apart, using a two-sample Student's *t* test.

Generalised Procrustes Analysis superimposition was performed for a selection of subsets of the total landmark set (Dryden & Mardia, 1998). Using this form of superimposition the 3D shape of the facial landmark configuration, the geometric relationship between the landmarks, is retained. The configurations are aligned using rotation, scaling and translation of the recorded 3D co-ordinate configurations such that there is minimal distance between corresponding landmarks and an average shape can be constructed. Mean shapes for the open and closed lip posture in the control population were generated. Hotelling's T squared multivariate testing of the data was performed (Dryden & Mardia, 1998).

3.4.3.3. Description of the Average Resting Face in Control Subjects

3.4.3.3.1. Linear Dimensions

The average resting face in all the control cases was described following one sample Student's *t* testing of a larger selection of linear distances, ratios and angles to define the degree of variation around the mean.

3.4.3.3.2. Symmetry

The asymmetry score of the image of the face at rest was calculated. The configuration of landmarks for each subject was mirror-imaged. The mirror-imaged landmark sets were superimposed on the original sets of landmarks. The 3D distances between corresponding landmarks in the superimposed sets were calculated and the average distance expressed as a score of asymmetry for each subject.

3.4.3.4. Measurement of Cleft Related Facial Deformity at Rest

3.4.3.4.1. Linear Dimensions

The same set of linear measurements was compared between the three subject types, UCL, UCLP and control cases, using one way ANOVA. Visual inspection of boxplots of the data was carried out to test for normality before deciding whether to use parametric or non-parametric tests. For measurements with statistically significant differences, Tukey's pairwise comparison was used to identify between which subject types the statistically significant difference occurred. For measurements with unequal variance, a Kruskal-Wallis non-parametric test of significance was performed. For asymmetric data, a Mann-Whitney pairwise comparison was used instead of Tukey's comparison to identify where statistically significant differences lay .

3.4.3.4.2. Three-Dimensional Analysis

Midface Relationship

To describe the prominence of the midface in 3D, Bookstein co-ordinate superimposition was employed. In this method of superimposition, two anchor landmarks are chosen and the position of other landmarks relative to the anchor landmarks is described. In this instance, the anchor points chosen were sublabialis (sl) and nasion (n) and assigned the (X, Y, Z) co-ordinates -0.5,0,0 and 0.5,0,0. These two midline landmarks were chosen to represent the upper and lower face respectively. These anchor landmarks now set a new set of axes and a new set of unit measurement. The remaining landmarks in the configuration assume new 3D co-ordinates based on their position relative to the anchor landmarks, Bookstein co-ordinates. When the third landmark is chosen, its co-ordinates will be (variable X, variable Y, 0) as it lies in the same Z plane as the anchor landmarks. The third point fixes the reference planes. The distances between landmarks with Bookstein co-ordinates are not in millimetres but in subdivisions of the distance between the two anchor points.

In this example, examining the relative position of the midface , the X axis runs vertically in the frontal and side views. The Z axis runs horizontally in the frontal and submento-vertex view and the Y axis runs horizontally in the side view.

The views of the face with the new axes illustrated are shown beside the plots showing the relative position of the subnasale (sn) landmark (Figure 3.23.). As all the points lie at the same point on the Z axis, the variation in the frontal and submento-vertex view varies only along one axis.

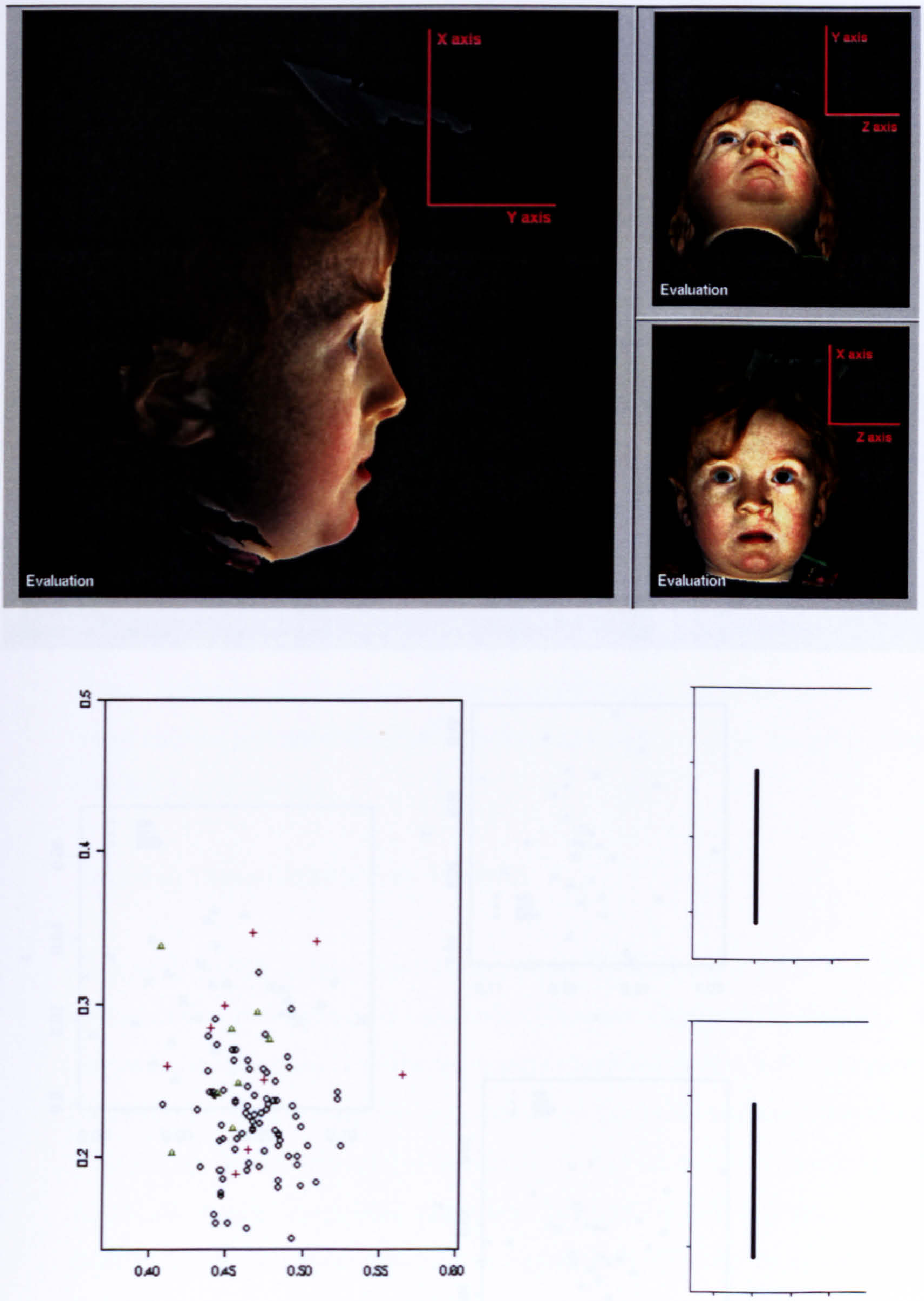
Intercanthal Area and Upper Lip

To describe the relative location of the nasal bridge, inner canthi and upper lip in 3D, Bookstein co-ordinate superimposition was again employed. In this instance, the anchor points chosen were right exocanthion (exR) and left exocanthion (exL) and assigned the (X, Y, Z) co-ordinates -0.5,0,0 and 0.5,0,0. These anchor landmarks now set a new set of axes and a new set of unit measurement. The third landmark chosen was nasion (n) , its co-ordinates became (variable X, variable Y, 0) as it lay in the same Z plane as the anchor landmarks.

In this example, examining the intercanthal area, the X axis runs horizontally in the frontal and submento-vertex views. The Z axis runs vertically in the frontal and side views and the Y axis runs horizontally in the side view.
The views of the face with the new axes illustrated are shown beside the plots showing the relative position of the right endocanthion (enR) landmark (Figure 3.24.). As there are more than 3 points in this example, not all lie on the same point on the Z axis. The greater variation in the three plots can be compared with the preceding example.

3.4.3.4.3. Symmetry

The symmetry scores for all three subject types, divided into subgroups with

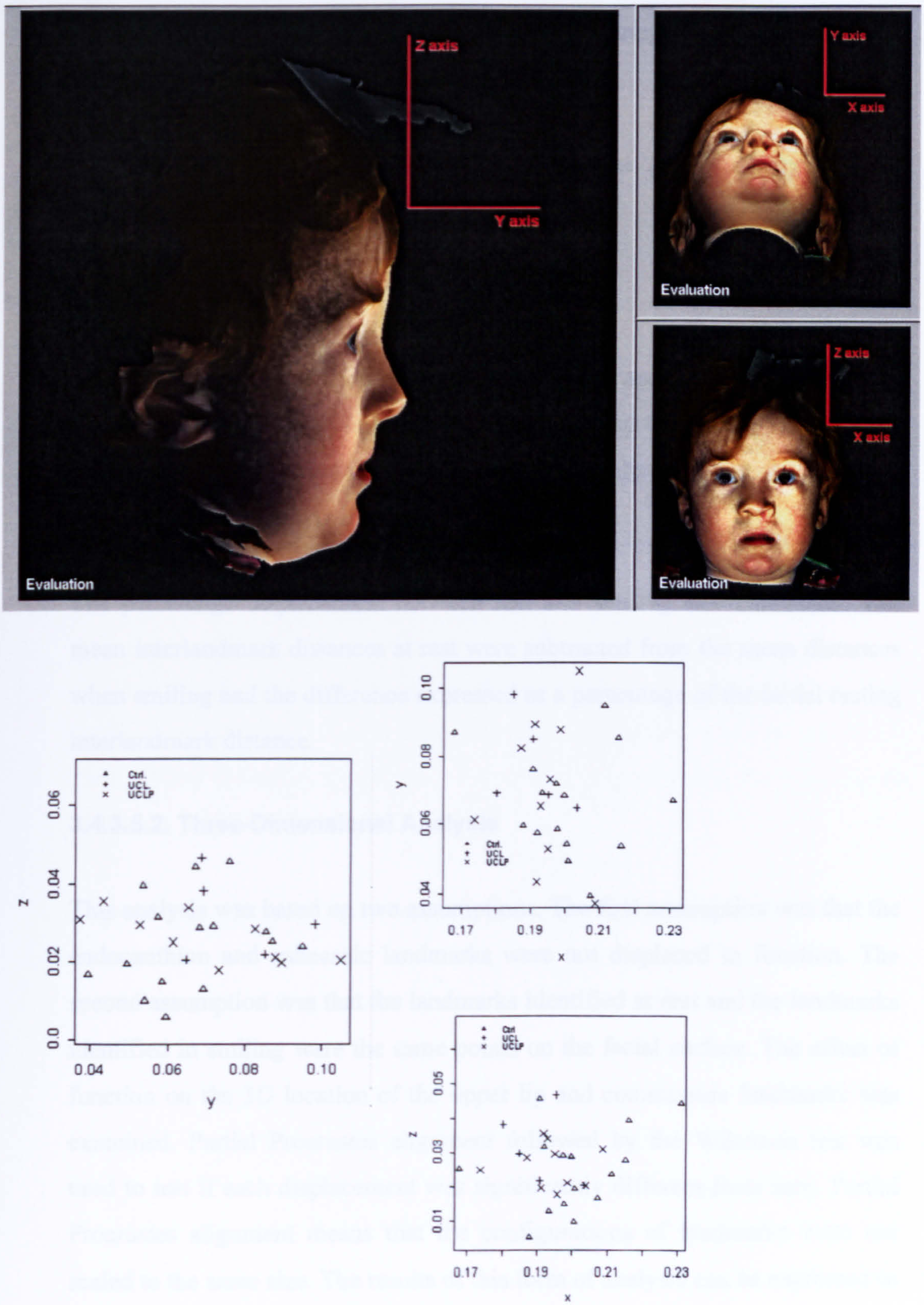


Circles = Control cases

Triangles = UCL cases

Crosses = UCLP cases

Figure 3. 23. Views of Face with Plots of Landmark Locations Aligned - Analysis of Midface Relationship



Triangle = Control Cross = UCL X = UCLP

Figure 3. 24. Views of Face with Plots of Landmark Locations Aligned – Analysis of Relative Position of Right Inner Canthus

open and closed lip pose at rest, were compared using all available landmarks. Symmetry scoring was repeated with the nasal and upper lip landmarks excluded and the scores for the three subject types compared again.

3.4.3.5. Description of Changes in Facial Morphology during Smiling in Controls

3.4.3.5.1. Linear Dimensions

For each control subject for whom a resting image and an acceptable smiling image was recorded, the linear distances, angles and ratios calculated for the resting image were compared with the same measurements calculated for the smiling image, using a two sample Student's *t* test.

The percentage displacement between rest and smiling was calculated. The mean interlandmark distances at rest were subtracted from the mean distances when smiling and the difference expressed as a percentage of the initial resting interlandmark distance.

3.4.3.5.2. Three-Dimensional Analysis

This analysis was based on two assumptions. The first assumption was that the endocanthion and subnasale landmarks were not displaced in function. The second assumption was that the landmarks identified at rest and the landmarks identified in smiling were the same points on the facial surface. The effect of function on the 3D location of the upper lip and commissure landmarks was examined. Partial Procrustes alignment followed by the Wilcoxon test was used to test if each displacement was significantly different from zero. Partial Procrustes alignment means that the configurations of landmarks were not scaled to the same size. The results of this form of analysis can be expressed in millimetres.

3.4.3.6. Measurement of Cleft Related Facial Deformity in Function

3.4.3.6.1. Linear Dimensions

For each subject for whom a resting image and an acceptable smiling image was recorded, the differences between the measurements in the resting image and the measurements in the smiling image were calculated in each subject. The mean differences between resting and smiling linear distances, ratios and angles for each of the three subject groups were compared using one way ANOVA with Tukey's pairwise comparison again employed to locate intergroup statistically significant differences. Unequal variance as determined by the boxplot of the data was not demonstrated, so it was not necessary to perform non-parametric testing of significance.

3.4.3.6.2. Three-Dimensional Analysis

The vectors of displacement of the endocanthion, subnasale, upper lip and commissure landmarks due to maximum smile production were compared for the cleft and control subject types as described above for the control subjects alone.

3.4.3.7. Dental Arch Form and Arch Relationships

3.4.3.7.1. Linear Dimensions

The linear dimensions of the study models were compared for the three subject types using one way ANOVA with Tukey's pairwise comparison following testing of data for normality. The maxillary to mandibular arch depth and circumferences ratios were calculated and compared for the three subject types.

3.4.3.7.2. Three-Dimensional Analysis

The 3D co-ordinate configurations were superimposed and scaled to unit size using Generalised Procrustes Analysis. The shapes of the maxillary and mandibular arches were compared for subject type. The majority of the subjects with UCLP had absent lateral incisors at the site of the repaired cleft so the left lateral incisor landmarks had to be excluded from the configuration. Data from subjects with unerupted molars had to be excluded from the analysis as all configurations were required to contain the same number of landmark co-ordinates.

3.4.3.8. Soft Tissue Facial Morphology and its Relationship with Dental Arch Features

A feature which was significantly different in each of the three subject types was considered to have higher power of discrimination. Two features of the dental arch and a feature of the soft tissue facial morphology at rest with the greatest power of discrimination were chosen. The correlation between the discriminant features was analysed for the three subject types.

Chapter 4

Results of Analysis of Facial Morphology

4.1. Profiles of the study groups

4.1.1. Data Collected

Table 4.1. summarises the data acquired from each of the study groups. The number of smiling images refers to the number of satisfactory smiling images acquired, not the numbers of children who could be persuaded to attempt to produce an maximal smile.

The discrepancy between the number of impressions acquired and the number of CT scanned study models suitable for analysis is due to the requirement for the selected landmark set to be present in all dental arch forms for 3D analysis. If a tooth other than the left lateral incisor was absent in the cleft maxillary arch the subject's study models were excluded from the analysis. Also, if primary molars were unerupted or partially erupted the subject's study models were excluded.

4.1.2. Socio-Economic Profiles of Study Subjects

In Figure 4.1. the socio-economic profile of the study groups is compared to the socio-economic profile of the Scottish cleft population over the last ten years. Despite a significant proportion of the control population coming from areas of social deprivation, visible dental caries was negligible. Only one child was excluded due to premature loss of primary teeth.

4.1.3.Gender Distribution of Study Subjects

The control sample was not controlled for gender but the male : female division within the cleft and unaffected samples was found to be equivalent, Figure 4.2.

	UCL		UCLP		Control	
	Number	%	Number	%	Number	%
Subjects Recruited	13		21		90	
Images						
Image of Face at Rest (Lips Closed)	8	72.7	11	52.3	73	81.1
Image of Face at Rest (Lips Open)	5	27.3	10	47.6	15	18.9
Image of Smiling Face	11	84.6	15	71.4	65	72.2
Study Models						
Upper and Lower Impressions	11	84.6	13	61.9	74	82.2
Upper Impression only	0	0	3	14.2	4	4.4
CT scanned Study Models	10	76.9	11	52.3	61	67.7

Table 4.1. Summary of Observations in Study Samples

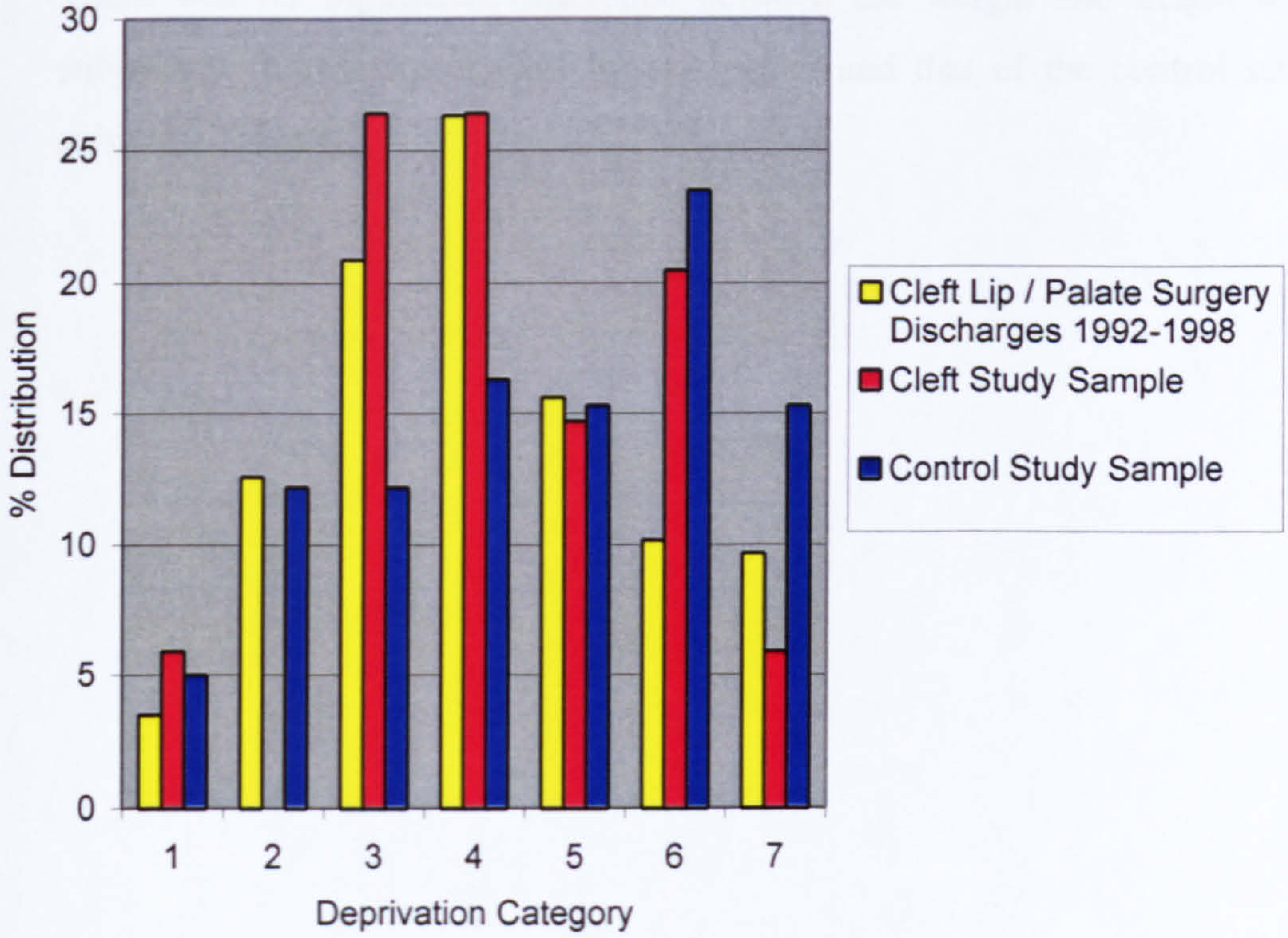


Figure 4.1. Socioeconomic Profile of Study Samples

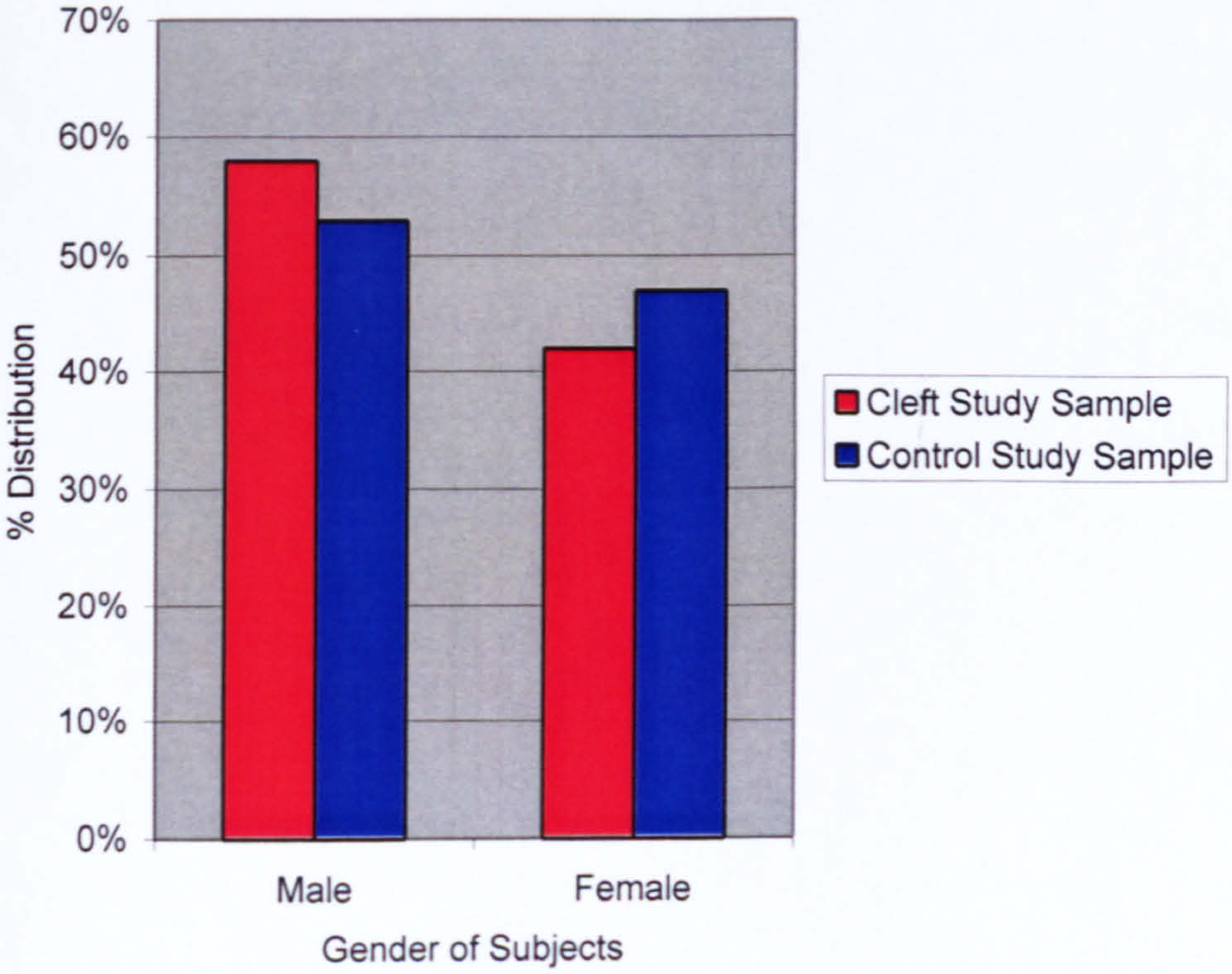


Figure 4.2. Gender Distribution in Study Samples

4.1.4.Height and Weight Profiles of Study Subjects

There was no significant difference between the weight and height of the subjects with cleft lip or cleft lip and palate and that of the control sample subjects, Table 4.2.

	UCL		UCLP		Control		
	Mean	Std Dev	Mean	Std Dev	Mean	Std Dev	p value
Height (cm)	95.44	3.63	96.24	4.3	95.75	3.29	0.82
Weight (kg)	15.87	2.24	14.85	1.49	15.33	1.91	0.4

Table 4.2. Comparisons of Height (in cm) and Weight (in kg) for 11 UCL, 19 UCLP and 86 Control Subjects

4.2. Facial Anthropometry

4.2.1. Quality Control

Images of two control subjects had to be excluded from the analysis of soft tissue morphology. Their images appeared warped which cast doubt on the integrity of the three-dimensional range data for that observation. The subjects had been co-operative, the calibration scores were satisfactory and the individual images captured by each camera in both pods were satisfactory. There was no evidence of movement in the delay between successive camera exposures but the merged images were unacceptable.

4.2.2. Effect of Lip Pose on Facial Anthropometry

There were some differences in the proportions of subjects exhibiting a closed lip pose in the images of the face captured at rest, Table 4.3. The higher proportion of cleft children with open lip pose at rest is due to the upper airway pathology and obligate mouth breathing patterns that may accompany orofacial clefting. At 3 years of age the children could not be induced to co-operate with photography to the extent that they would hold their breath to produce a lip closed pose. Alternatively, if children were trying hard to co-operate and achieve a lip seal, they produced an image showing visible effort, with flattening of the chin and lip pursing rather than a relaxed pose. Such images were discarded as unsatisfactory.

4.2.2.1.Analysis of Lip Pose Effect on Interlandmark Distances

It should be noted that all linear distances referred to in the following analyses were Euclidean distances. Euclidean distance is the shortest distance measured between two points in space. This qualifying adjective will be implied, but not stated, wherever the word distance appears.

	UCL		UCLP		Control	
	Number	%	Number	%	Number	%
Lips Together	8	61.5	11	52.3	73	83
Lips Apart	5	38.5	10	47.7	15	17
Total	13		21		88	

Table 4.3. Observed Lip Pose at Rest in Study Samples

The results of two sample Student's *t* testing of measurements compared for the variable lip pose in the control population are shown in Table 4.4. The larger control sample was chosen for analysis to increase the statistical power of the comparison.

In the upper face, around the eyes and the nasal base there were no statistically significant differences between the interlandmark distances measured in the images of children with a closed lips pose and those measured in children with lips apart at rest. The upper face height differed significantly, probably because it was measured between nasion (n) and stomion (sto) with the latter landmark location being directly affected by the alteration in lip pose.

In the lower face, the linear distances that differed significantly were the vertical dimensions directly related to the lips while the horizontal inter-commissure distance was not affected by lip pose. The smaller nasal height observed in the group with a closed lip pose may be due to subtle differences in perioral tone elevating the soft tissue subnasale (sn) landmark. Flattening of the chin was apparent when significant effort was being made to close the lips but it may have been that some level of effort, sufficient to raise the sn point and reduce the measured nasal height, was not visible and therefore images with subtle increases in perioral tone were included in the study sets.

4.2.2.2. Effect of Lip Pose on Three-Dimensional Facial Configuration

The difference between the recorded images detected by comparison of linear measurements was confirmed using 3D analysis. The results of multivariate testing of the constructed mean shapes for open and closed lip pose in cleft and control subjects are shown in Table 4.5.

Not all available landmarks were included in the comparison, as the variability of the whole facial shape could have obscured the effect of lip pose detected initially following analysis of interlandmark distances.

(Red = Upper face)		Closed		Open		
(Black = Midface)		(n= 73)		(n= 15)		
(Green = Lower Face)		Mean	Std dev	Mean	Std dev	p value
Binocular width:	exL - exR	75.75	3.33	75.17	2.43	0.44
Right ocular width:	exR - enR	23.80	1.30	23.82	0.75	0.92
Left ocular width:	exL - enL	24.02	1.31	23.87	1.31	0.69
Intercanthal width:	enR - enL	29.54	1.93	29.19	1.42	0.42
Upper face height:	n - sto	50.95	2.82	49.11	2.98	0.04
Midface prominence	sl - n - sn	12.29	2.46	13.40	2.95	0.19
Right naso-aural distance:	n - obiR	98.81	3.53	99.44	3.70	0.55
Left naso-aural distance:	n - obiL	98.67	3.69	98.94	3.13	0.77
Right subnasale-aural distance:	sn - obi R	93.85	3.20	94.54	4.80	0.60
Left subnasale-aural distance:	sn - obi L	93.87	3.50	94.11	3.78	0.83
Right lower aurochelion distance	obi R-chR	75.27	3.51	75.93	4.44	0.60
Left lower aurochelion distance	obi L-chL	75.33	3.78	75.40	4.33	0.95
Nose height:	n - sn	32.86	2.10	34.07	1.96	0.04
Nose bridge length:	n - prn	27.77	2.04	29.01	1.94	0.04
Nasal projection	sn - prn	10.80	0.90	10.96	1.32	0.66
Nasolabial angle:	prn - sn - ls	133.25	7.66	132.22	5.84	0.41
Nasal tip angle	n-prn-sn	108.75	4.25	108.50	4.76	0.86
Soft tissue nose width	alL - alR	25.48	1.76	25.18	1.61	0.53
Length of right ala:	acR - prn	19.91	1.12	20.18	1.41	0.49
Length of left ala:	acL - prn	20.00	1.18	20.27	1.37	0.48
Anatomical width of nose:	acR - acL	28.17	1.66	28.37	1.87	0.69
Nasal Base Width	sbalR-sbalL	13.29	1.82	13.62	1.77	0.53
Width of right nostril floor:	sbalR - sn	7.19	0.94	7.34	0.87	0.56
Width of left nostril floor:	sbalL - sn	7.26	1.04	7.26	1.14	1.00
Width of the columella:	sn'R - sn'L	7.26	0.56	4.90	0.70	0.83
Length of columella (right):	cR - sn	6.11	0.82	6.06	0.94	0.85
Length of columella (left):	cL - sn	6.16	0.80	5.79	0.96	0.18
Mouth width:	chR - chL	36.99	2.19	36.60	1.88	0.40
Upper lip height:	sn - sto(s)	18.80	1.76	15.39	1.76	<0.001
Upper cutaneous lip height:	sn - ls	13.70	1.97	12.50	1.99	0.05
Upper vermillion lip height:	ls-sto(s)	6.15	1.52	3.46	1.21	<0.001
Lower lip height:	sto - sl	13.51	1.38	11.17	1.49	<0.001
Lower cutaneous lip height:	li - sl	8.41	1.57	8.03	1.46	0.38
Lower vermillion height:	sto - li	5.50	1.50	3.72	1.10	<0.001
Width of the philtrum:	cphR - cphL	8.90	1.18	8.00	1.13	0.12
Right hemiphiltrum width	cphR-ls	4.64	0.64	4.20	0.57	0.01
Left hemiphiltrum width	cphL-ls	4.61	0.64	4.09	0.62	0.01
Right upper lip length	chR -cphR	19.58	1.57	21.28	1.73	0.00
Left upper lip length	chL -cphL	19.69	1.59	21.04	1.82	0.02
Right lower lip length	li-chR	19.83	1.40	19.62	1.31	0.58
Left lower lip length	li-chL	20.00	1.54	19.53	1.61	0.31

Table 4.4. Comparison of Interlandmark Distances (mm) at Rest in Controls with Varying Lip Pose.

	Hotelling's p Values	
	Cleft subjects	**Control subjects**
No significant difference		
exR, exL, enR, enL, n	0.82	0.76
exR, exL, enR, enL, n, sl	0.06	0.07
exR, exL, enR, enL, n, sn	0.92	0.40
exR, exL, enR, enL, n, sn, acR, acL	0.93	0.17
exR, exL, enR, enL, n, sl, sn, acR, acL	0.12	0.13
exR, exL, enR, enL, n, sl, sn, acR, acL, alR, acL	0.41	0.26
exR, exL, enR, enL, n, sn, chR, chL	0.24	0.22
Significant difference in one group		
sl, sn, chR, chL	0.00	0.18
sl, sn, acR, acL, alR, alL	0.03	0.40
sn, chR, chL, acR, acL	0.04	0.46
exR, exL, enR, enL, n, sl, sn	0.04	0.20
exR, exL, enR, enL, n, sl, sn, acR, acL, alR, acL ls, li	0.23	0.01
Significant difference in both groups		
exR, exL, enR, enL, n, ls, li	0.00	0.01
sl, sn, acR, acL, alR, alL, ls, li	0.01	0.00

Table 4.5. 3D comparison of Facial Landmark Configurations for Changing Lip Pose

The average configurations of upper facial landmarks did not significantly differ with changing lip pose. The inclusion of nasal base landmarks in the averaged configurations did not detect a pose effect but the addition of upper and lower lip landmarks produced significantly different face shapes for the open and closed lip poses with Hotelling's T squared p values of less then 0.05.

The effect of lip pose was, on balance, considered significant and therefore subsequent analysis was carried out with the children's images classified according to lip pose at rest as well as according to study group.

4.2.3. Anthropometry of the Face at Rest in the Control Group

4.2.3.1. Interlandmark Distance Analysis

The characterisation of the control face at rest, by describing the average interlandmark distances, ratios and angles across the face for the resting face, is summarised in Table 4.6. The coefficient of variance, or ratio of standard deviation to mean value, shows which areas of the face showed the greatest degree of variability. In the upper face the coefficient of variance was less than 0.1 with the exception of the angle describing the prominence of the midface.

The interpretation of angles calculated between 3D co-ordinates should be circumspect. A large displacement in the location of one landmark may not be reflected in the angle measured if the geometric relationship between the three landmarks creating the angle resembles that of other sets of landmarks. See section 4.2.3.3.1. for a full investigation of the midface relationship to the upper and lower face.

Greater variation, with larger coefficients of variance from 0.10 to 0.30, was seen in the lower portion of the face and in the small distances measured around nose and lips. The alar thickness, bilaterally, and the upper and lower

(Red = Upper Face) (Black = Midface) (Green = Lower Face)		Controls - closed lips at rest (n= 73)			
		Mean	Std Dev	SE mean	CV
Binocular width:	exL - exR	75.75	3.33	0.39	0.04
Right ocular width:	exR - enR	23.80	1.30	0.15	0.05
Left ocular width:	exL - enL	24.02	1.31	0.15	0.05
Intercanthal width:	enR - enL	29.54	1.93	0.23	0.07
Upper face height:	n - sto	50.95	2.82	0.33	0.06
Midface prominence	sl - n - sn	12.29	2.46	0.29	0.20
Right lower naso-aural distance:	n - obiR	98.81	3.53	0.41	0.04
Left lower naso-aural distance:	n - obiL	98.67	3.69	0.43	0.04
Right subnasale-aural distance:	sn - obi R	93.85	3.20	0.37	0.03
Left subnasale-aural distance:	sn - obi L	93.87	3.50	0.41	0.04
Right aurochelion distance	obi R-chR	75.27	3.51	0.41	0.05
Left aurochelion distance	obi L-chL	75.33	3.78	0.44	0.05
Nose height:	n - sn	32.86	2.10	0.25	0.06
Nose bridge length:	n - prn	27.77	2.04	0.24	0.07
Nasal projection	sn - prn	10.80	0.90	0.11	0.08
Nasolabial angle:	prn - sn - ls	133.25	7.66	0.63	0.06
Nasal tip angle	n-prn-sn	108.75	4.25	0.50	0.04
Soft tissue nose width	alL - alR	25.48	1.76	0.21	0.07
Length of right ala:	acR - prn	19.91	1.12	0.13	0.06
Length of left ala:	acL - prn	20.00	1.18	0.14	0.06
Right alar thickness:	al'Ri to al'Ro	3.31	0.68	0.08	0.20
Left alar thickness:	al'Li to al'Lo	3.31	0.64	0.08	0.19
Anatomical width of nose:	acR - acL	28.17	1.66	0.19	0.06
Nasal Base Width	sbalR-sbalL	13.29	1.82	0.21	0.14
Width of right nostril floor:	sbalR - sn	7.19	0.94	0.11	0.13
Width of left nostril floor:	sbalL - sn	7.26	1.04	0.12	0.14
Width of the columella:	sn'R - sn'L	7.26	0.56	0.07	0.08
Length of columella (right):	cR - sn	6.11	0.82	0.10	0.13
Length of columella (left):	cL - sn	6.16	0.80	0.09	0.13
Soft tissue nose width: mouth width	alR-alL:chR-chL	0.69	0.05	0.01	0.07
Anatomical tissue nose width : mouth width	acR-acL:chR-chL	0.76	0.05	0.00	0.07
Mouth width:	chR - chL	36.99	2.19	0.26	0.06
Upper lip height:	sn - sto(s)	18.80	1.76	0.21	0.09
Upper cutaneous lip height:	sn - ls	13.70	1.97	0.23	0.14
Upper vermillion lip height:	ls-sto(s)	6.15	1.52	0.18	0.25
Lower lip height:	sto - sl	13.51	1.38	0.16	0.10
Lower cutaneous lip height:	li - sl	8.41	1.57	0.18	0.19
Lower vermillion height:	sto - li	5.50	1.50	0.18	0.27
Width of the philtrum:	cphR - cphL	8.90	1.18	0.14	0.13
Right upper lip length	chR -cphR	19.58	1.57	0.18	0.08
Left upper lip length	chL -cphL	19.69	1.59	0.19	0.08
Right lower lip length	li-chR	19.83	1.40	0.16	0.07
Left lower lip length	li-chL	20.00	1.54	0.18	0.08

Table 4.6. Average Linear Dimensions (mm) in Resting Control Subject's Faces with Closed Lip Pose

vermillion lip heights demonstrated the largest degree of variability.

4.2.3.2. Symmetry of Interlandmark Distances

The largest available sample, the resting face in the control subjects with lips together, was analysed. The comparison of paired linear measurements for this sample is shown in Table 4.7.

There was no statistically significant difference between interlandmark distances on the right and left sides of the faces of the control sample. This technique of assessing symmetry is very rudimentary. Apparent symmetry of linear dimensions gave no information about symmetry of landmarks or whether the face would appear balanced to the observer.

4.2.3.3. Three-Dimensional Analysis

The variation of the human facial shape and its complexity challenges the capabilities of statistical analysis using shape analysis models. The research questions had to be focused based on the results of initial analysis of distances to allow meaningful statistical investigation.

4.2.3.3.1.Midface Relationship to the Upper and Lower Face

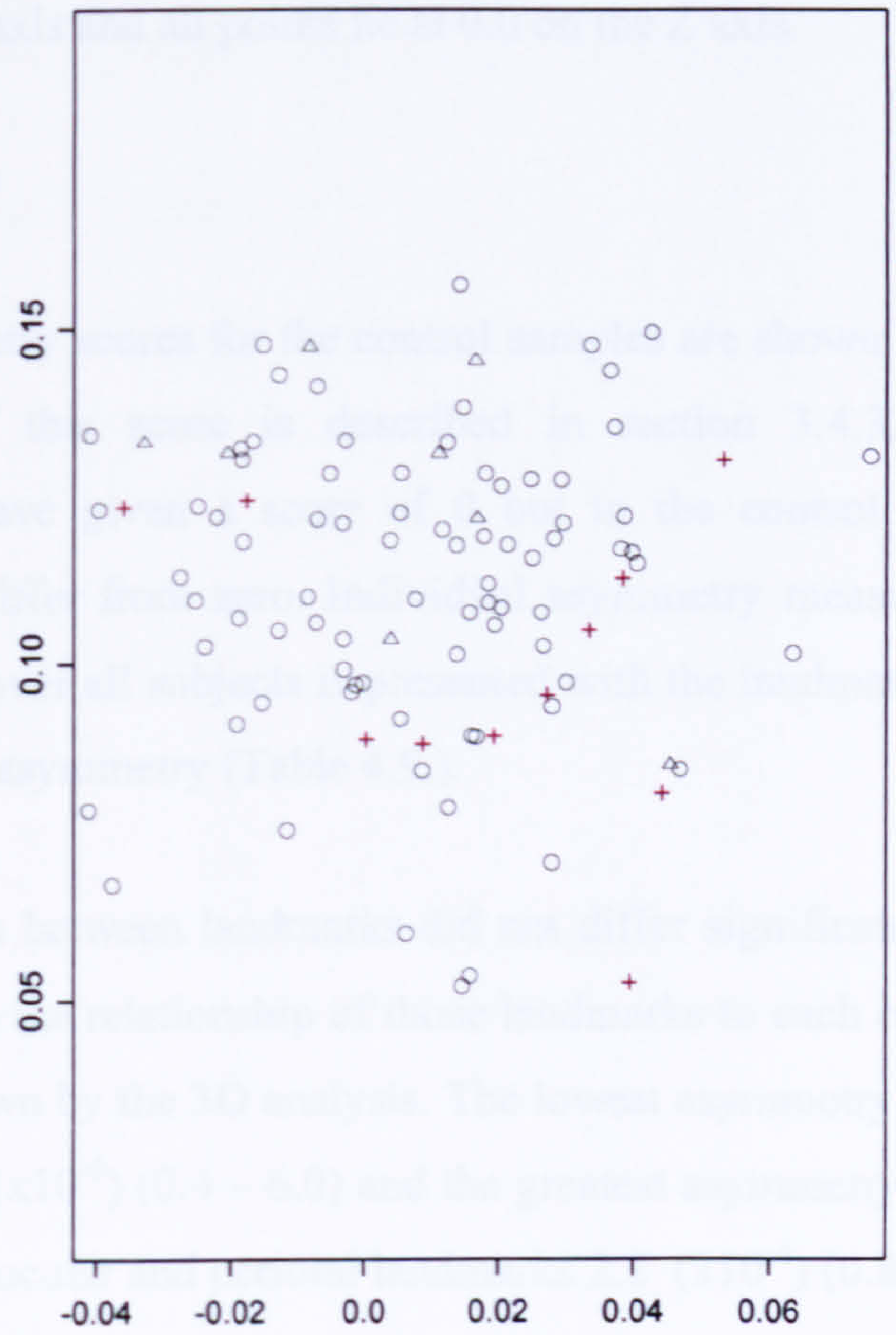
To fully describe the relative position of the midface, it was necessary to define the 3D location of the subnasale point at the base of the columella. The Bookstein co-ordinate system was employed which considered the soft tissue nasion (n) and sublabialis (sl) as fixed baseline points. Three views of the face with the new axes illustrated are shown in Figure 4.3. The X-axis became vertical in the frontal and lateral view as the anchor points lay one above the other vertically.

The range of positions of the midface, represented by (sn), is shown in Figure 4.4. The plot shows the position of (sn) relative to (n) representing the upper face and (sl) representing the lower face. There is more variation in the vertical

		Right (N=73)		Left (N=73)		
		Mean	St Dev	Mean	StDev	p value
Ocular width:	ex - en	23.80	1.30	24.02	1.31	0.31
Nasoaural Distance:	n - obi	98.81	3.53	98.67	3.69	0.82
Subnasale -Aural Distance:	sn - obi	93.85	3.20	93.87	3.40	0.97
Alar Length:	ac - prn	19.91	1.12	20.00	1.18	0.63
Alar Thickness:	al'i to al'o	3.31	0.68	3.31	0.64	1.00
Width of Nostril Floor:	sbal - sn	7.19	0.94	7.26	1.04	0.63
Columellar Length:	c - sn	6.11	0.82	6.16	0.80	0.75
Upper Lip Length	ch - cph	19.58	1.57	19.69	1.59	0.68
Lower lip length	ch - li	19.83	1.40	20.00	1.54	0.48

Table 4.7. Comparison of Paired Linear Measurements In 73 Control Subjects at Rest

Figure 4.3. Face at Rest with Frame of Reference Set for Analysis of Midface Position



Circle = Control Triangle = UCL Cross = UCLP

(Refer to axes illustrated in Figure 323)

Figure 4.4. Relative Position of Subnasale (Sn) in 3 Subject Types

direction (X -axis) than in the anteroposterior direction (Y axis). All three points are assigned the same location on the horizontal axis so there is no variation along this axis and all points lie at 0.0 on the Z axis.

4.2.3.3.2.Symmetry

The median asymmetry scores for the control samples are shown in Table 4.8. The calculation of this score is described in section 3.4.3.3.2. Perfect symmetry would have given a score of 0 but in the control subjects the asymmetry scores differ from zero. Individual asymmetry measures for each landmark averaged over all subjects is presented with the landmarks ranked in order of descending asymmetry (Table 4.9.).

The paired distances between landmarks did not differ significantly on either side of the face, but, the relationship of those landmarks to each other was not symmetrical, as shown by the 3D analysis. The lowest asymmetry score was in the ocular area 1.2 ($x10^{-5}$) (0.4 – 6.0) and the greatest asymmetry was seen in the configuration of ocular and perioral landmarks 2.2 ($x10^{-5}$) (0.8 – 3.6).

Comparing the ranked landmark asymmetry and the regional asymmetry, it can be seen that individual landmarks of relatively higher asymmetry in combination created less asymmetrical regions.

4.2.4. Anthropometry of the Face at Rest (Cleft Samples Vs. Control Sample)

4.2.4.1. Interlandmark Distance Analysis

Tables 4.10. to 4.13 show the results of the comparison of the interlandmark distances for all three subject types, for subjects with a closed lip pose at rest. Table 4.14 shows the pairwise comparison of the 25 interlandmark distances between subject types where initial ANOVA showed a significant difference.

	Minimum	Lips Closed Median	Maximum	Minimum	Lips Apart Median	Maximum
Landmark Configurations						
Li, Ls, N, Sl, AcR, AcL, AlR, AlL, CR, CL, CphR, CphL, EnR, EnL, ExR, ExL, SbalR, SbalL, Sn'R, Sn'L	1	2	4	1	2	3.6
Li, Ls, N, Sl, ChR, ChL, EnR, EnL, ExR, ExL,	1	2	5.2	0.8	2.2	3.6
Li, Ls, N, Sl, EnR, EnL, ExR, ExL,	0.9	2	5.6	0.8	2.2	3
N, Sl, AcR, AcL, AlR, AlL, EnR, EnL, ExR, ExL,	0.8	1.8	5.2	0.8	2	2.6
N, Sl, EnR, EnL, ExR, ExL	0.6	1.4	6.8	0.6	1.6	2.5
N, EnR, EnL, ExR, ExL	0.4	1.2	6	0.6	1.6	2

Table 4.8 Asymmetry Scores (Unit Size x 10^{-5}) for Control Cases

Landmark	Abbreviation	
Sublabialis	sl	**Highest Asymmetry Score**
Left Exocanthion	exL	
Right Cheilion	chR	
Left Cheilion	chL	
Labrale inferiorus	li	
Right Exocanthion	exR	
Right Christa Philtri	cphR	
Left Christa Philtri	cphL	
Right Endocanthion	enR	
Left Endocanthion	enL	
Left Alare	alL	
Left Alar Curvature	acL	
Right Alare	alR	
Left Columella High Point	cL	
Right Alar Curvature	acR	
Right Columella High Point	cR	
Right Subalare	sbalR	
Left Subalare	sbalL	
Left Subnasale'	sn'L	
Right Subnasale'	sn'R	**Lowest Asymmetry Score**

Table 4.9. Ranked Individual Landmark Asymmetry Scores (in descending order)

			UCL (N=7)	UCLP (N=11)	Control (N=73)
			Mean	Std Dev	p Value
Binocular width:	exL - exR	UCL	76.05	1.56	
		UCLP	77.12	3.34	
		Control	75.75	3.33	0.43
Right ocular width:	exR - enR	UCL	23.58	0.65	
		UCLP	23.78	1.21	
		Control	23.80	1.30	0.91
Left ocular width:	exL - enL	UCL	23.80	1.02	
		UCLP	23.74	1.36	
		Control	24.02	1.31	0.75
Intercanthal width:	enR - enL	UCL	30.24	1.31	
		UCLP	31.41	2.29	
		Control	29.54	1.93	0.01
Upper face height:	n - sto	UCL	51.60	3.29	
		UCLP	51.60	3.31	
		Control	50.96	2.82	0.71
Midface prominence	sl - n - sn	UCL	13.88	2.83	
		UCLP	10.83	2.70	
		Control	12.29	2.47	0.05
Right lower naso-aural distance:	n - obiR	UCL	100.17	3.43	
		UCLP	98.27	7.87	
		Control	98.81	3.53	0.68
Left lower naso-aural distance:*	n - obiL	UCL	99.87	59.80	
		UCLP	98.70	42.00	
		Control	98.50	44.40	0.40
Right lower subnasale-aural distance: *	sn - obi R	UCL	94.33	50.30	
		UCLP	92.02	35.00	
		Control	93.88	46.70	0.34
Left lower subnasale-aural distance:*	sn - obi L	UCL	96.32	62.40	
		UCLP	91.38	29.60	
		Control	93.73	46.10	0.04
Nose height:	n - sn	UCL	33.37	2.56	
		UCLP	33.84	2.59	
		Control	32.86	2.10	0.35
Nose bridge length:	n - prn	UCL	27.96	2.37	
		UCLP	27.69	2.48	
		Control	27.77	2.04	0.97

* Indicates asymmetric data

Underlined figures are the results of non-parametric testing

Table 4.10. Comparison of Linear Measurements (mm) in the Upper Face at Rest (Lips Closed) in 7 UCL, 11 UCLP and 73 Control Cases

			UCL (N=7)	UCLP (N=11)	Control (N=73)
			Mean	Std Dev	p Value
Nasal projection	sn - pm	UCL	11.07	1.66	
		UCLP	11.04	1.33	
		Control	10.80	0.90	0.64
Nasolabial angle: *	pm - sn - ls	UCL	133.60	111.20	
		UCLP	143.10	135.00	
		Control	133.60	83.10	<0.001
Nasal tip angle	n-pm-sn	UCL	109.92	3.77	
		UCLP	114.94	4.24	
		Control	108.75	4.25	<0.001
Soft tissue nose width	alL - alR	UCL	27.15	1.43	
		UCLP	28.21	1.99	
		Control	25.48	1.76	<0.001
Length of right ala: *	acR - pm	UCL	20.42	51.40	
		UCLP	20.77	65.60	
		Control	19.78	42.50	0.02
Length of left ala: *	acL - pm	UCL	22.63	70.30	
		UCLP	20.77	49.80	
		Control	19.77	43.10	0.03
Right alar thickness: *	al'Ri to al'Ro	UCL	2.77	30.00	
		UCLP	3.54	60.60	
		Control	3.29	45.30	0.05
Left alar thickness:	al'Li to al'Lo	UCL	3.17	0.96	
		UCLP	3.43	0.80	
		Control	3.31	0.64	0.73
Anatomical width of nose:	acR - acL	UCL	30.04	0.69	
		UCLP	31.09	1.73	
		Control	28.17	1.66	<0.001

* Indicate asymmetric data spread

Underlined figures are the results of non-parametric testing

Table 4.11. Comparison of Linear Measurements in the Upper Nasal Region at Rest (Lips Closed) in 7 UCL, 11 UCLP and 73 Control Cases

			UCL (N=7)	UCLP (N=11)	Control (N=73)
			Mean	Std Dev	p Value
Nasal Base Width	sbalR-sbalL	UCL	15.15	1.54	
		UCLP	17.76	1.60	
		Control	13.29	1.82	<0.001
Width of right nostril floor:	sbalR - sn	UCL	7.88	1.56	
		UCLP	9.83	2.05	
		Control	7.19	0.94	<0.001
Width of left nostril floor:	sbal L to sn	UCL	8.77	1.12	
		UCLP	9.56	2.59	
		Control	7.27	1.04	<0.001
Width of columella	sn'R - sn'L	UCL	5.58	0.86	
		UCLP	5.47	0.79	
		Control	4.94	0.56	0.00
Length of columella (right):	cR - sn	UCL	6.25	1.41	
		UCLP	6.43	1.95	
		Control	6.11	0.82	0.64
Length of columella (left): *	cL - sn	UCL	4.83	21.10	
		UCLP	5.27	39.50	
		Control	6.07	49.40	0.02
Soft tissue nose width : mouth width:	alR-alL:chR-chL	UCL	0.68	0.34	
		UCLP	0.76	0.06	
		Control	0.69	0.05	<0.001
Anatomical tissue nose width : mouth width:*	acR-acL:chR-chL	UCL	0.75	76.20	
		UCLP	0.85	150.80	
		Control	0.76	84.00	<0.001
Mouth width:	chR - chL	UCL	39.75	1.96	
		UCLP	37.07	2.65	
		Control	37.07	2.19	0.01

* Indicates asymmetric data spread

Underlined figures are the results of non-parametric testing

Table 4.12. Comparison of Linear Measurements (mm) in the Lower Nasal Region at Rest (Lips Closed) in 7 UCL, 11 UCLP and 73 Control Cases

			UCL (N=7)	UCLP (N=11)	Control (N=73)
			Mean	Std Dev	p Value
Upper lip height:	sn - sto(s)	UCL	19.26	1.90	
		UCLP	18.34	2.09	
		Control	18.80	1.76	0.57
Upper cutaneous lip height:	sn - ls	UCL	12.11	2.78	
		UCLP	12.42	2.05	
		Control	13.70	1.97	0.04
Upper vermillion lip height:*	ls-sto(s)	UCL	7.64	67.60	
		UCLP	5.22	42.80	
		Control	5.91	44.40	0.08
Lower lip height:	sto - sl	UCL	13.95	1.25	
		UCLP	13.44	1.22	
		Control	13.51	1.38	0.69
Lower cutaneous lip height:	li - sl	UCL	8.82	2.24	
		UCLP	8.49	1.49	
		Control	8.41	1.57	0.82
Lower vermillion height:	sto - li	UCL	5.52	1.23	
		UCLP	5.64	1.45	
		Control	5.50	1.50	0.96
Width of the philtrum:	cphR - cphL	UCL	11.79	2.17	
		UCLP	11.51	1.51	
		Control	8.90	1.18	<0.001
Right hemiphiltrum width	cphR-ls	UCL	5.20	1.03	
		UCLP	5.93	1.09	
		Control	4.64	0.64	<0.001
Left hemiphiltrum width	cphL-ls	UCL	6.76	1.47	
		UCLP	5.73	1.29	
		Control	4.61	0.64	<0.001
Right upper lip length	chR-cphR	UCL	19.90	1.60	
		UCLP	18.06	2.13	
		Control	19.58	1.57	0.15
Left upper lip length	chL-cphL	UCL	20.46	1.39	
		UCLP	18.16	2.18	
		Control	19.69	1.59	0.01
Right lower lip length	li-chR	UCL	21.01	1.37	
		UCLP	20.00	1.81	
		Control	19.83	1.40	0.12
Left lower lip length *	li-chL	UCL	21.55	73.30	
		UCLP	19.87	44.30	
		Control	20.09	43.60	0.02

* Indicates asymmetric data spread

Underlined figures are the results of non-parametric testing

Table 4.13. Comparison of Linear Measurements (mm) in the Perioral Region at Rest (Lips Closed) in 7 UCL, 11 UCLP and 73 Control Cases

		UCL vs. Control		UCLP vs. Control		UCLP vs. UCL	
Intercanthal width:	enR - enL	-2.52	1.13	-3.36	-0.38	-3.40	1.06
Midface prominence	sl - n - sn	-3.97	0.78	-0.48	3.40	0.15	5.95
Left lower subnasale-aural distance:	sn - obi L	-7.72	2.65	1.07	8.26	1.16	13.15
Nasolabial angle:*	prn - sn - ls	-10.07	0.12	-11.47	-5.00	-10.08	2.76
Nasal tip angle	n-prn-sn	-5.16	2.80	-9.45	-2.94	-9.88	-0.16
Soft tissue nose width	alL - alR	-3.34	0.00	-4.09	-1.37	-3.09	0.98
Length of right ala: *	acR - prn	-1.33	0.42	-1.96	-0.41	-0.47	1.97
Length of left ala: *	acL - prn	-3.66	-0.62	-1.48	0.72	-1.01	3.67
Right alar thickness: *	al'Ri to al'Ro	-0.13	0.90	-1.13	0.03	0.02	1.84
Left alar thickness:	al'Li to al'Lo	-1.96	2.23	-3.66	-0.24	-4.64	0.47
Anatomical width of nose:	acR - acL	-3.40	-0.34	-4.17	-1.67	-2.92	0.82
Nasal Base Width	sbalR-sbalL	-3.54	-0.19	-5.84	-3.10	-4.66	-0.56
Width of right nostril floor:	sbalR - sn	-1.79	0.42	-3.54	-1.74	-3.30	-0.61
Width of left nostril floor:	sbal L to sn	-2.74	-0.26	-3.31	-1.28	-2.31	0.73
Width of columella	sn'R - sn'L	-1.22	-0.06	-1.00	-0.06	-0.60	0.82
Length of columella (left):	cL - sn	0.24	1.98	-0.44	0.98	-1.90	0.22
Soft tissue nose width : mouth width:	alR-alL:chR-chL	-0.04	0.05	-0.11	-0.04	-0.14	-0.02
Anatomical tissue nose width : mouth width:	acR-acL:chR-chL	-0.03	0.04	-0.11	-0.05	-0.13	-0.04
Mouth width:	chR - chL	-4.79	-0.58	-1.72	1.73	0.11	5.26
Upper vermillion lip height:	sn - ls	-3.37	-0.19	-1.55	1.04	-0.41	3.47
Width of the philtrum:	cphR - cphL	-4.13	-1.66	-3.62	-1.60	-1.23	1.80
Right hemiphiltrum width	cphR-ls	-1.26	0.13	-1.86	-0.72	-1.58	0.12
Left hemiphiltrum width	cphL-ls	-2.92	-1.38	-1.75	-0.49	0.08	1.97
Left upper lip length	chL-cphL	-2.33	0.79	0.26	2.81	0.39	4.21
Left lower lip length	li-chL	-3.46	-0.57	-1.30	1.07	0.13	3.66

Blue ink indicates significant result

Table 4.14. Pairwise Comparison of Linear Measurements (mm) in the Face at Rest (Lips Closed) in 7 UCL, 11 UCLP and 73 Control Cases

The results of the same comparison for subjects with the lips apart at rest were similar, as seen in Tables 4.15. to 4.18 but have not been discussed in detail. Pairwise comparison for the interlandmark distances in the resting faces with lips open are seen in Table 4.19.

The larger number of control subjects with lips closed at rest, as preferred by the data collection protocol, increased the power of the analysis to detect differences between cleft faces and control faces with lips together. Fewer significant differences between cleft and control subjects were detected in the groups with lips apart resting poses. There were 13 significant differences in the latter group and 25 in the group with lips closed at rest. Figures underlined in Tables 4.10. to 4.13 and Tables 4.15. to 4.18. indicate a non-parametric Kruskal-Wallis test was performed because a boxplot of the data showed asymmetric data spread. In these cases, the mean should be read as the median value and the standard deviation should be read as the rank. Where the Kruskal - Wallis test results were significant, the pairwise comparison was by a Mann Whitney test rather than a Tukey's comparison.

In the upper face, the ocular and binocular widths did not differ between subject types but the intercanthal widths were greater in subjects with UCLP than in control subjects or in those with UCL. It could not be claimed that subjects with UCLP had telecanthus until the 3D position of the inner canthi was investigated.

The vertical face heights and the apparent relationships between the sublabial fold, nasal root and nasal spine were the same in the three subject types. The angle formed by the chin, nasal root and nasal spine was largest in subjects with UCLP and a significant difference was detected at the 95% confidence level. Pairwise comparison at 98% confidence to adequately test small numbers of cases did not confirm this finding. The distances from the ears to the nasal root and base were similar in all subject types with one exception. The left subnasale aural distance was greater in subjects with UCL than in the other subject types. 3D investigation was still required to describe the relative

			UCL (N=5)	UCLP (N=10)	Control (N=15)
			Mean	Std Dev	p Value
Binocular width:	exL - exR	UCL	73.06	4.40	
		UCLP	76.51	2.63	
		Control	75.17	2.43	0.11
Right ocular width:	exR - enR	UCL	22.92	1.73	
		UCLP	23.66	1.44	
		Control	23.82	0.75	0.36
Left ocular width:	exL - enL	UCL	22.63	2.35	
		UCLP	23.22	1.23	
		Control	23.87	1.31	0.25
Intercanthal width:	enR - enL	UCL	29.21	3.43	
		UCLP	31.00	1.93	
		Control	29.19	1.42	0.09
Upper face height: *	n - sto	UCL	46.25	10.20	
		UCLP	50.70	19.20	
		Control	49.33	14.80	0.16
Midface prominence *	sl - n - sn	UCL	13.12	17.00	
		UCLP	11.09	11.90	
		Control	13.46	17.40	0.28
Right lower naso-aural distance: *	n - obiR	UCL	99.19	15.00	
		UCLP	98.01	11.60	
		Control	98.92	14.00	0.72
Left lower naso-aural distance:	n - obiL	UCL	96.90	5.05	
		UCLP	97.88	3.97	
		Control	98.94	3.13	0.57
Right lower subnasale-aural distance:*	sn - obi R	UCL	93.91	15.30	
		UCLP	88.81	7.10	
		Control	94.71	16.00	0.04
Left lower subnasale-aural distance:	sn - obi L	UCL	91.24	5.05	
		UCLP	91.14	3.75	
		Control	94.11	3.78	0.15
Nose height:	n - sn	UCL	33.29	2.44	
		UCLP	34.33	2.05	
		Control	34.07	1.96	0.66
Nose bridge length:	n - prn	UCL	27.92	2.03	
		UCLP	27.88	2.47	
		Control	29.01	1.94	0.38

* Indicates asymmetric data spread

Underlined figures are results of non-parametric testing

Table 4.15. Comparison of Linear Measurements (in mm) in the Upper Face at Rest (Lips Open) in 5 UCL, 10 UCLP and 15 Control Cases

			UCL (N=5)	UCLP (N=10)	Control (N=15)
			Mean	Std Dev	p Value
Nasal projection	sn - pm	UCL	11.29	0.38	
		UCLP	11.52	1.12	
		Control	10.96	1.32	0.50
Nasolabial angle: *	pm - sn - ls	UCL	125.70	24.70	
		UCLP	143.80	45.30	
		Control	133.40	22.60	<0.001
Nasal tip angle	n-pm-sn	UCL	108.83	6.11	
		UCLP	114.87	6.66	
		Control	108.50	4.76	0.03
Soft tissue nose width *	alL - alR	UCL	26.76	18.20	
		UCLP	27.66	22.40	
		Control	25.77	10.00	0.00
Length of right ala:	acR - pm	UCL	19.74	0.52	
		UCLP	20.08	2.58	
		Control	20.18	1.41	0.89
Length of left ala:	acL - pm	UCL	20.83	1.64	
		UCLP	21.48	2.66	
		Control	20.27	1.37	0.33
Right alar thickness:	al'Ri to al'Ro	UCL	3.39	0.38	
		UCLP	3.45	0.73	
		Control	3.26	0.80	0.82
Left alar thickness: *	al'Li to al'Lo	UCL	2.92	17.20	
		UCLP	2.80	14.10	
		Control	2.85	15.90	0.79
Anatomical width of nose:	acR - acL	UCL	29.55	2.30	
		UCLP	30.33	1.72	
		Control	28.38	1.87	0.05

* Indicates asymmetric data spread

Underlined figures are results of non-parametric testing

Table 4.16. Comparison of Linear Measurements (in mm) in the Upper Nasal Region at Rest (Lips Open) in 5 UCL, 10 UCLP and 15 Control Cases

			UCL (N=7)	UCLP (N=10)	Control (N=15)
			Mean	Std Dev	p Value
Nasal Base Width	sbalR-sbalL	UCL	16.21	1.90	
		UCLP	19.30	2.18	
		Control	13.62	1.77	<0.001
Width of right nostril floor:	sbalR - sn	UCL	7.28	1.10	
		UCLP	8.75	1.81	
		Control	7.34	0.87	0.03
Width of left nostril floor: *	sbal L to sn	UCL	10.00	19.80	
		UCLP	13.11	23.20	
		Control	7.08	8.90	<0.001
Width of columella	sn'R - sn'L	UCL	5.61	1.02	
		UCLP	5.62	1.05	
		Control	4.90	0.70	0.10
Length of columella (right):	cR - sn	UCL	7.57	0.33	
		UCLP	6.46	1.79	
		Control	6.06	0.94	0.08
Length of columella (left):	cL - sn	UCL	5.35	0.51	
		UCLP	5.53	1.35	
		Control	5.79	0.96	0.69
Soft tissue nose width : mouth width:	alR-alL:chR-chL	UCL	0.74	0.04	
		UCLP	0.78	0.09	
		Control	0.69	0.06	0.01
Anatomical tissue nose width : mouth width:	acR-acL:chR-chL	UCL	0.80	0.07	
		UCLP	0.84	0.86	
		Control	0.78	0.60	0.02
Mouth width:	chR - chL	UCL	36.96	3.04	
		UCLP	36.45	3.26	
		Control	36.60	1.88	0.94

* Indicates asymmetric data spread

Underlined figures are results of non-parametric testing

Table 4.17. Comparison of Linear Measurements (mm) in the Lower Nasal Region at Rest (Lips Open) in 5 UCL, 10 UCLP and 15 Control Cases

			UCL (N=7)	UCLP (N=10)	Control (N=15)
			Mean	Std Dev	p Value
Upper lip height: *	sn - sto(s)	UCL	14.02	12.20	
		UCLP	16.82	19.60	
		Control	15.84	13.90	0.18
Upper cutaneous lip height:	sn - ls	UCL	10.49	2.01	
		UCLP	12.68	2.03	
		Control	12.50	1.99	0.12
Upper vermillion lip height:	ls-sto(s)	UCL	4.85	1.12	
		UCLP	4.38	1.24	
		Control	3.46	1.21	0.06
Lower lip height: *	sto - sl	UCL	11.84	16.80	
		UCLP	11.85	16.90	
		Control	11.16	14.10	0.70
Lower cutaneous lip height:	li - sl	UCL	8.38	1.65	
		UCLP	8.12	1.27	
		Control	8.03	1.46	0.90
Lower vermillion height:	sto - li	UCL	4.42	1.12	
		UCLP	4.88	1.35	
		Control	3.73	1.10	0.07
Width of the philtrum:	cphR - cphL	UCL	11.29	1.22	
		UCLP	11.35	1.56	
		Control	8.00	1.13	<0.001
Right hemiphiltrum width	cphR-ls	UCL	4.85	0.72	
		UCLP	5.47	1.57	
		Control	4.20	0.57	0.02
Left hemiphiltrum width	cphL-ls	UCL	6.57	1.25	
		UCLP	6.03	1.26	
		Control	4.09	0.62	<0.001
Right upper lip length *	chR-cphR	UCL	21.35	21.00	
		UCLP	17.58	6.70	
		Control	21.14	19.50	0.00
Left upper lip length	chL-cphL	UCL	21.68	1.77	
		UCLP	18.70	1.97	
		Control	21.04	1.82	0.01
Right lower lip length	li-chR	UCL	20.99	1.67	
		UCLP	20.53	2.14	
		Control	19.62	1.31	0.22
Left lower lip length	li-chL	UCL	21.43	1.48	
		UCLP	21.37	1.64	
		Control	19.53	1.61	0.13

*Indicates asymmetric data spread

Underlined figures are results of non-parametric testing

Table 4.18. Comparison of Linear Measurements (mm) in the Perioral Region at Rest (Lips Open) in 5 UCL, 10 UCLP and 15 Control Cases

		UCL vs. Control		UCLP vs. Control		UCL vs. UCLP	
Nasal Base Width	sbalR-sbalL	-5.07	-0.12	-7.64	-3.73	-5.72	-0.46
Width of right nostril floor:	sbalR - sn	-1.59	1.71	-2.72	-0.11	-3.22	0.28
Width of left nostril floor:	sbal L to sn	-4.82	-1.03	-7.05	-2.82	-1.59	5.43
Nasolabial angle:*	prn - sn - ls	-6.28	10.03	-15.15	-7.30	-19.96	-2.67
Nasal tip angle	n-prn-sn	-7.59	6.92	-12.11	-0.64	-13.74	1.65
Soft tissue nose width: *	alL - alR	-4.11	0.45	-4.82	-0.99	-3.03	2.03
Soft tissue nose width : mouth width:	alR-alL:chR-chL	-0.14	0.04	-0.16	-0.20	-0.12	0.06
Anatomical tissue nose width : mouth width:	acR-acL:chR-chL	-0.09	0.04	-0.11	-0.01	-0.10	0.03
Width of the philtrum:	cphR - cphL	-4.96	-1.62	-4.67	-2.03	-1.83	1.71
Right hemiphiltrum width	cphR-ls	-1.97	0.68	-2.31	-0.22	-2.03	0.79
Left hemiphiltrum width	cphL-ls	-3.74	-1.23	-2.93	-0.95	-0.78	1.87
Right upper lip length: *	chR-cphR	-2.35	1.41	1.54	5.37	1.36	6.85
Left upper lip length	chL-cphL	-3.03	1.75	0.05	4.23	0.45	5.52

Blue ink indicates significant result

Table 4.19. Pairwise Comparison of Linear Measurements (mm) in the Face at Rest (Lips Open) in 5 UCL, 10 UCLP and 15 Control Cases

position of the midface.

The nasal height, length and projection were similar in all subject types. The nasal base and upper lip linear measurements showed the greatest deviation from normal, and therefore the greatest deformity, in cleft subjects.

The nasolabial and nasal tip angles were approximately 10 and 5 degrees larger, respectively, in subjects with UCLP compared to the UCL and control groups. This suggests that either the upper lip was flatter or the nasal tip was higher with perhaps more of the nostrils showing in frontal view in UCLP. Interpretation of angles formed between 3D landmarks must be cautious, as explained earlier.

The nasal dimensions differed most widely between the study groups. The nose, therefore, was the location of the greatest deformity. The soft tissue and anatomical nose widths were greater in cleft subjects with the UCL and UCLP noses resembling each other and differing significantly from normal. The nasal widths were wider in UCLP than in UCL but the difference did not achieve statistical significance. The nasal base width was narrowest in control cases, wider in UCL cases and wider again in UCLP. All differences were statistically significant.

The nostril floor widths replicated these differences. The nostril floor was widest in UCLP cases on both sides and significantly different from normal. In the UCL cases the left (affected) nostril floor width was significantly different from normal but the width of the right (unaffected) nostril floor was within normal limits. In the UCL cases, there was not just widening of the nose but asymmetry between the lengths of the nostril floors. The columella was significantly wider in UCL and UCLP cases than in control cases. The median left columellar length at 5.27 mm was shorter than the control median of 6.07mm but this was not statistically significant. The left columellar length was significantly shorter than normal in the UCL cases.

The splayed cleft nose was confirmed by the larger nose to mouth width ratios in the cleft cases. The mouth width was greater in the UCL face but, as the commissures are unrelated to bone, the variability of the resting expression cannot be discounted as a significant contribution to mouth width variety. Statistical significance may not always be clinically significant in this area of the face. The upper lip height was the same across the three subject types but the cutaneous lip height was slightly shorter in the cleft cases in the midline. The upper vermillion lip height in the midline was similar in the 3 groups, the subjective observation of lip drooping and an enlarged vermillion lip height in cleft cases was not confirmed in analysis of the resting face.

The philtrum was wider in UCL and UCLP cases than in control cases. The average difference between UCL cases and control cases was 25% greater than the average difference between UCLP cases and controls. The difference in philtrum width between UCL and UCLP cases was not significant. The main source of difference was on the cleft-affected side of the philtrum. On the left, there was a significant difference in the hemiphiltrum width between the three subject types. The width was greatest in the UCL cases, least in the control cases and the width in the UCLP cases lay between that of the other two groups.

The left upper lip length was also greater in the UCL cases than in both UCLP and control cases. This would suggest that the proportions between philtrum and lip length, which are constructed at the time of cheiloplasty, are similar for UCL and UCLP cases.

The left lower lip length in the UCL cases was significantly greater than in the control cases and the UCLP cases. The left lower lip length was shorter in the UCLP cases than in the control cases but not to a statistically significant extent.

4.2.4.2.Three-Dimensional Analysis

4.2.4.2.1. Midface Relationship

The results of computation of Bookstein co-ordinates for the subnasale point in the three subject types is presented in Figure 4.4. The 3D location of the subnasale point relative to nasion and sublabialis in UCL and UCLP cases is superimposed on the graph showing the range of 3D locations for subnasale in control cases. Multivariate ANOVA analysis of the Booksteined co-ordinates confirmed the subjective impression of an overlapping range of variability for all subject types. The p-values results of the Hotelling's T squared test did not achieve statistical significance as shown in Table 4.20.

Univariate ANOVA did not detect any significant variation when the X and Y co-ordinates were considered separately. This finding would suggest that at this age there is no apparent soft tissue midface retrusion in the cleft cases.

4.2.4.2.2. Intercanthal Area

A replication of the Bookstein analysis was performed with two baseline landmarks, as for the midface. The right exocanthion (exR) and left exocanthion (exL) points were considered fixed and the positions of nasion (n) and right endocanthion (enR) and left endocanthion (enL), were evaluated.

Table 4.21. of Hotelling's T squared p values results for the multivariate MANOVA show that no significant difference between the three groups was found for the 3D positions of n and enR relative to exR and exL. The position of enL differed in subjects with a closed lip pose and the significant variation was in the X axis. The pairwise comparison detected that the significant difference lay between the UCLP cases and the control cases (Table 4.22.).

This finding agreed with the results of linear analysis which found a significantly larger intercanthal width in UCLP. Three –dimensional analysis

	Multivariate ANOVA		Univariate ANOVA
	p Values		p values
At Rest – Closed Lips	0.09	X axis	0.32
		Y axis	0.05
At Rest – Open Lips	0.12	X axis	0.13
		Y axis	0.15
Smiling	0.21	X axis	0.20
		Y axis	0.52

Table 4.20. Comparison of Three Dimensional Location of Midface relative to Upper and Lower Face for 8 UCL, 11 UCLP and 73 Control Cases

	Multivariate ANOVA	Univariate ANOVA	
	p Values		p Values
N	0.17	X axis	0.12
		Y axis	0.33
EnR	0.87	X axis	0.37
		Y axis	0.86
		Z axis	0.93
EnL	0.01	X axis	0.00
		Y axis	0.86
		Z axis	0.90

Table 4.21. Comparison of Three Dimensional Location of Inner Canthi and Nasal Bridge Relative to Right and Left Outer Canthi for 8 UCL, 11 UCLP and 73 Control Cases

	Multivariate ANOVA		Univariate ANOVA
	p Values		p Values
UCL vs. Control	0.34	X axis	0.07
		Y axis	0.84
		Z axis	0.74
UCLP vs. Control	0.00	X axis	<0.001
		Y axis	0.62
		Z axis	0.77
UCL vs. UCLP	0.57	X axis	0.18
		Y axis	0.69
		Z axis	0.76

Table 4.22. Pairwise Comparison of Three Dimensional Location of Left Inner Canthus Relative to Right and Left Outer Canthi

suggested the left inner canthus was displaced relative to the other periocular landmarks. This localised the cause of the increased intercanthal width to the enL landmark.

A significant difference was detected between the UCLP cases and the control cases on pairwise comparison. The 3D location of the left inner canthus in the UCLP cases was displaced along the X axis in the Bookstein frame of reference relative to the control cases. As the anchor points chosen were the right and left exocanthion, the X axis in the Bookstein frame of reference was the horizontal axis in the frontal view. Displacement in the X axis indicates that the inner canthus in the UCLP cases was lateral to the inner canthus in the control cases in the frontal plane.

The Bookstein plots graphically representing the relative 3D location of the landmarks discussed are presented in Appendix 5.

4.2.4.2.4. Symmetry

Table 4.23 summarises the results of comparison of asymmetry scores between the three subject types using different configurations of landmarks. The facial configuration incorporating the largest number of landmarks was significantly more asymmetric in the UCLP and UCL cases than in the control cases. There was no significant difference between asymmetry scores in the two cleft groups.

There were no significant differences between the asymmetry scores for any of the subject types when the ocular and nasal bridge landmarks were examined. When the sublabialis (sl) landmark was added to the ocular configuration, a significant difference was detected between the UCL and control cases with the lips apart, but not with the lips closed.

The addition of upper and lower midline lip landmarks to the configuration, detected a persistent significant difference between the UCL cases and the control cases with the lips apart. Recalculating the asymmetry scores for ocular

Closed Lips Pose Landmark Configurations	**UCL vs Control** **p Values**	**UCLP vs Control** **p Values**	**UCL vs UCLP** **p Values**
Li, Ls, N, Sl, AcR, AcL, AlR, AlL, CR, CL, CphR, CphL, EnR, EnL, ExR, ExL, SbalR, SbalL, Sn'R, Sn'L	0.02	<0.001	0.17s
Li, Ls, N, Sl, ChR, ChL, EnR, EnL, ExR, ExL,	0.32	0.01	0.23
Li, Ls, N, Sl, EnR, EnL, ExR, ExL,	0.59	0.06	0.44
N, Sl, AcR, AcL, AlR, AlL, EnR, EnL, ExR, ExL,	0.28	0.1	0.77
N, Sl, EnR, EnL, ExR, ExL	0.55	0.25	0.06
N, EnR, EnL, ExR, ExL	0.1	0.13	1

Lips Apart Pose Landmark Configurations	**UCL vs Control** **p Values**	**UCLP vs Control** **p Values**	**UCL vs UCLP** **p Values**
Li, Ls, N, Sl, AcR, AcL, AlR, AlL, CR, CL, CphR, CphL, EnR, EnL, ExR, ExL, SbalR, SbalL, Sn'R, Sn'L	<0.001	0.001	1
Li, Ls, N, Sl, ChR, ChL, EnR, EnL, ExR, ExL,	0.09	0.19	0.51
Li, Ls, N, Sl, EnR, EnL, ExR, ExL,	0.04	0.21	0.25
N, Sl, AcR, AcL, AlR, AlL, EnR, EnL, ExR, ExL,	0.08	0.01	0.95
N, Sl, EnR, EnL, ExR, ExL	0.02	0.19	0.25
N, EnR, EnL, ExR, ExL	0.23	0.96	0.16

Blue Ink highlights Significant Result

Table 4.23. Pairwise Comparison of Asymmetry Scores for 13 UCL, 21 UCLP and 88 Control Subjects

and lip landmarks, including the commissures, detects a significant difference between the UCLP and control cases, only with the lips closed.

Comparison of the asymmetry scores for a configuration of ocular and peripheral nasal landmarks showed 1 significant difference between UCLP and control cases with the lips apart. A larger asymmetry score in the cleft nasal area might have been expected. The aim had been to describe the overall nasal shape so peripheral landmarks on the nose were chosen rather than the nasal base landmarks.

Comparison of the asymmetry scores for a configuration of ocular and perioral landmarks showed one significant difference between UCLP and control cases with the lips closed.

The boxplots of the data reported in this section are presented in Appendix 6.

4.3. Assessment of Facial Function (Rest vs. Smiling Expressions)

4.3.1. Interlandmark Distance Analysis in Control Subjects

The comparison of the linear measurements of the face in the control subjects at rest and performing an maximal smile was the first step in analysing the changes in soft tissue morphology in function. The linear measurements were calculated between the 3D location of the anthropometric landmarks at rest and at the point of maximal effort. The change, or otherwise, of the dimensional relationship between the landmarks did not give any information about any changes in location of the individual landmarks.

Table 4.24 shows the results of two sample Student's *t* testing and the percentage displacement (pD) of interlandmark distances for resting and smiling expressions in that portion of the control sample which was initially photographed at rest with a closed lip pose. The percentage displacement is the difference between the distance at rest and the distance when smiling, expressed as a percentage of the distance at rest.

The distances measured around the eyes, the columella and between the midline and the periphery of the face changed little with alteration in expression. Percent displacements of less than 2.8% were not found to be statistically significant.

In function the upper face height reduced, the nose, nasal base and both nostrils widened, the mouth width increased and the cutaneous upper lip and upper lip vermillion height shortened. Percent displacements for these distances were 6% or greater. The differences between distances at rest and in maximal smile were statistically significant.

The subjective observation that the nose tip dropped and the lip was drawn

(pD = Percent Displacement)		Rest (N=56)		Smile (N=56)			
		Mean	Std Dev	Mean	Std Dev	p value	pD
Upper Face							
Intercanthal width:	enR - enL	29.44	1.73	29.23	1.98	0.55	-0.71
Right hemi intercanthus	enR-n	18.29	1.42	18.21	1.42	0.77	-0.44
Left hemi intercanthus	enL-n	17.98	1.45	17.84	1.37	0.62	-0.78
Upper face height:	n - sto	50.72	3.04	46.05	2.76	<0.001	-9.21
Midface prominence	sl - n - sn	12.05	2.58	11.71	2.74	0.49	-2.82
Right naso-aural distance:	n - obiR	98.57	3.67	98.40	3.23	0.79	-0.17
Left naso-aural distance:	n - obiL	98.38	3.66	97.70	3.47	0.32	-0.69
Right subnasale-aural distance:	sn - obi R	93.61	3.28	92.97	2.92	0.28	-0.68
Left subnasale-aural distance:	sn - obi L	93.65	3.49	92.57	3.42	0.10	-1.15
Mid Face							
Nose height:	n - sn	32.67	2.19	32.19	2.29	0.26	-1.47
Nose bridge length:	n - prn	27.60	2.05	27.99	2.01	0.32	1.41
Nasal protrusion	sn - prn	10.79	0.91	10.14	1.11	0.00	-6.04
Nasolabial angle:	prn - sn - ls	133.48	7.98	140.21	6.48	<0.001	5.04
Nasal tip angle	n-prn-sn	108.54	4.51	105.40	4.98	<0.001	-2.89
Soft tissue nose width	alL - alR	25.51	1.72	27.30	1.81	<0.001	7.02
Length of right ala:	acR - prn	19.84	1.12	20.73	1.28	<0.001	4.49
Length of left ala:	acL - prn	19.89	1.17	20.83	1.17	<0.001	4.73
Anatomical width of nose:	acR - acL	28.17	1.66	30.85	2.10	<0.001	9.51
Nasal Base Width	sbalR-sbalL	13.36	1.77	16.48	1.60	<0.001	23.35
Width of right nostril floor:	sbalR - sn	7.17	0.90	8.78	0.93	<0.001	22.32
Width of left nostril floor:	sbalL - sn	7.31	1.03	8.95	0.80	<0.001	22.41
Width of the columella:	sn'R - sn'L	4.89	0.56	4.91	0.52	0.86	0.37
Length of columella (right):	cR - sn	6.18	0.86	6.15	0.85	0.87	-0.40
Length of columella (left):	cL - sn	6.28	0.80	5.99	0.77	0.05	-4.64
Lower Face							
Nose Mouth ratio 1	alR-alL:chR-chL	0.69	0.05	0.56	0.04	<0.001	-17.99
Nose Mouth ratio 2	acR-acL:chR-chL	0.76	0.06	0.64	0.05	<0.001	-16.13
Mouth width:	chR - chL	37.16	2.31	48.52	3.25	<0.001	30.57
Upper lip height:	sn - stos	18.75	1.84	14.88	1.55	<0.001	-20.64
Upper cutaneous lip height:	sn - ls	13.76	2.11	11.38	1.78	<0.001	-17.30
Upper vermillion lip height:	ls-stos	6.04	1.53	4.02	1.13	<0.001	-33.44
Lower lip height:	stoi - sl	13.43	1.40	12.77	1.32	0.01	-4.91
Lower cutaneous lip height:	li - sl	8.63	1.57	8.78	1.43	0.60	1.74
Lower vermillion height:	stoi - li	5.11	1.38	4.82	0.78	0.18	-5.71

Table 4.24. Comparison of Linear Measurements (mm) in Resting and Smiling Faces in 56 Controls Cases

backwards when smiling was consistent with the findings of increases in the nasolabial angle and the midface prominence angle between upper lip, nasal root and nasal base. The nasal tip angle reduction by 2.89% in function was statistically significant.

The total lower lip height shortened by 4.91% which achieved statistical significance but the vermillion lip height shortened by 5.71% and the p value of the comparison between expressions was 0.177. The coefficient of variance for the lower lip vermillion height was high, between 0.16 and 0.27. This explains the lack of statistical significance attributed to the larger percent displacement.

Three-dimensional analysis was necessary to describe the movement of individual landmarks in function.

4.3.2. Three-Dimensional Analysis in Control Subjects

Tables 4.25. and 4.26. show X, Y, Z and 3D displacements for enR, enL, sn, ls, chR and chL, key ocular and perioral landmarks. In this analysis the axes are not altered by the method of superimposition so the X and Y axes are horizontal and vertical in the frontal plane and the Z axis is horizontal in the sagittal plane. The minimum, median and maximum displacements in each direction are listed. The p value stated refers to the statistical significance of the difference between the displacement recorded and zero displacement.

Table 4.25 shows that while the assumption was made that the endocanthion and subnasale landmarks were not displaced in function there was a statistically significant displacement of these points along the Y and Z axes. As the cumulative displacement in absolute terms was less than 1mm for each of the three landmarks, the assumption that these landmarks were stable for analytical purposes was justified.

		Rest (Lips Closed) to Smile			
		X	Y	Z	3D
Cheilion Left	Minimum Displacement	2.1	-1.1	-14.7	4.7
Cheilion Left	Median Displacement	5.8	4.3	-6.4	12.3
Cheilion Left	Maximum Displacement	9.6	8.9	-1.3	19.2
Cheilion Left	Wilcoxon p-Value	0.19	<0.001	<0.001	
Cheilion Right	Minimum Displacement	2.4	-2.5	-13.6	3.8
Cheilion Right	Median Displacement	5.8	3.8	-6	11
Cheilion Right	Maximum Displacement	10.5	8	-1.9	16.8
Cheilion Right	Wilcoxon p-Value	0.53	<0.001	<0.001	
Labrale Superiorus	Minimum Displacement	-1.7	0.3	-7.2	1.4
Labrale Superiorus	Median Displacement	-0.1	3.4	-2.5	4.9
Labrale Superiorus	Maximum Displacement	1.7	5.9	-0.7	8.8
Labrale Superiorus	Wilcoxon p-Value	0.44	<0.001	<0.001	
Endocanthion Right	Minimum Displacement	-2	-1.4	-0.4	0.1
Endocanthion Right	Median Displacement	0.1	-0.4	0.3	0.8
Endocanthion Right	Maximum Displacement	1.9	0.7	0.9	2.4
Endocanthion Right	Wilcoxon p-Value	0.19	<0.001	<0.001	
Endocanthion Left	Minimum Displacement	-1.7	-1.1	-0.2	0.1
Endocanthion Left	Median Displacement	-0.2	-0.4	0.3	0.6
Endocanthion Left	Maximum Displacement	1.9	0.3	0.8	2.4
Endocanthion Left	Wilcoxon p-Value	0.52	<0.001	<0.001	
Subnasale	Minimum Displacement	-0.8	-0.2	-1.4	0.1
Subnasale	Median Displacement	0	0.7	-0.5	0.9
Subnasale	Maximum Displacement	0.6	2.1	0.1	2.6
Subnasale	Wilcoxon p-Value	0.44	<0.001	<0.001	

Table 4.25. Displacement (mm) of Ocular and Perioral Landmarks in Smile in Control Cases (with lips closed at rest)

	Rest (Lips Apart) to Smile				
		X	Y	Z	3D
Cheilion Left	Minimum Displacement	3.8	2.3	-14	10
Cheilion Left	Median Displacement	6	5.3	-7.7	11
Cheilion Left	Maximum Displacement	6.6	7.1	-4.8	15.3
Cheilion Left	Wilcoxon p-Value	0.007	0.007	0.007	
Cheilion Right	Minimum Displacement	4.8	3	-9.1	6.8
Cheilion Right	Median Displacement	5.3	4.4	-6.3	10.5
Cheilion Right	Maximum Displacement	8.8	8	-3.8	12.7
Cheilion Right	Wilcoxon p-Value	0.007	0.007	0.007	
Labrale Superiorus	Minimum Displacement	-0.8	-0.5	-4.9	2.3
Labrale Superiorus	Median Displacement	0.5	2.9	-2.9	4.6
Labrale Superiorus	Maximum Displacement	0.9	4.5	-1.9	5.3
Labrale Superiorus	Wilcoxon p-Value	0.74	0.02	0.007	
Endocanthion Right	Minimum Displacement	-0.5	-0.7	0	0.2
Endocanthion Right	Median Displacement	0	-0.4	0.2	0.6
Endocanthion Right	Maximum Displacement	0.6	-0.1	0.5	1
Endocanthion Right	Wilcoxon p-Value	0.84	0.008	0.008	
Endocanthion Left	Minimum Displacement	-0.6	-0.8	0.1	0.5
Endocanthion Left	Median Displacement	-0.1	-0.6	0.4	0.7
Endocanthion Left	Maximum Displacement	0.6	-0.1	0.5	1
Endocanthion Left	Wilcoxon p-Value	0.31	0.008	0.008	
Subnasale	Minimum Displacement	-0.1	0.2	-1	0.2
Subnasale	Median Displacement	0	0.9	-0.6	1.1
Subnasale	Maximum Displacement	0.2	1.3	-0.1	1.6
Subnasale	Wilcoxon p-Value	0.31	0.008	0.008	

Table 4.26. Displacement (mm) of Ocular and Perioral Landmarks in Smile in Control Cases (with lips apart at rest)

The changes with expression will be described for the subjects who were photographed at rest with a closed lip pose.

The commissures moved outwards, upwards and backwards relative to their 3D location at rest, the median displacement was 11.0 mm on the right and 12.3 mm on the left. The posterior displacement in the Z axis was the largest component of the movement vector, this was 6.0 mm on the right and 6.4 mm on the left. The movements of the right and left commissures mirrored each other.

The main component of the displacement vector of the midline of the upper vermillion border was along the Y axis, there was 3.4 mm upward displacement. There was 2.5 mm posterior displacement along the Y axis. This finding confirmed the posterior displacement of the upper lip observed clinically, with the lip pulled towards the maxillary teeth. In the analysis of the linear measurements an increase was noted in the nasolabial angle and the angle between upper lip, nasal root and nasal base. This could be explained by the alteration in the upper lip border location. The subnasale point was shown to remain relatively stable and the soft tissue nasion point is closely related to underlying bone and stable for anatomical reasons.

The findings in analysis of facial expression must be considered with regard to the stated assumptions of stable landmarks for superimposition and homology of landmarks in the resting and smiling face.

4.3.3. Assessment of Facial Function (Cleft Subjects vs. Control Subjects)

4.3.3.1. Interlandmark Distance Analysis

The differences between linear measurements at rest and in function were calculated for each subject and the averaged differences compared. All subjects were analysed together regardless of initial resting lip pose as the change with function was investigated, not the absolute values of the linear measurements. In some children's images the landmarks at the ear (otobasion inferiorus) could not be identified due to coverage of the area by hair. Distances between the ears and other landmarks have not been included in this analysis as only 5 of the cleft subjects who performed an acceptable smile had uncovered ears, despite the researcher's efforts during image capture.

The results of comparisons between the three subject types are presented in Tables 4.27. and 4.28. The results of pairwise comparison are reported in Table 4.29.

The patterns of change in soft tissue morphology with function in the cleft subjects were similar to the control subjects' patterns. The degree of deformity or deviation from normal, noted in the cleft subjects at rest did not change when the subjects smiled with the exception of the left nostril floor and upper lip in the area of the cheiloplasty.

The intercanthal distance, columellar width and length and lower lip changed very little with changing expression in the cleft subjects as in the control subjects. In function the upper face height reduced; the nose, nasal base and both nostrils widened; the mouth width increased; and the cutaneous upper lip and upper lip vermillion height shortened to the same extent as in the control sample. The nostril flare was significantly greater in the UCL and UCLP cases than in the control cases on the cleft affected side of the face. The isolated unilateral additional nostril flaring would suggest nasal base asymmetry increased in function. The increases in nasal base width, soft tissue and anatomical nose width in function did not differ between subjects.

			UCL (N=11)	UCLP (N=15)	Control (N=65)
			Mean	Std Dev	p Value
Intercanthal width:	enR - enL	UCL	-0.48	1.42	
		UCLP	-0.27	1.09	
		Control	-0.21	1.20	0.79
Upper face height:	n - sto	UCL	-3.87	2.47	
		UCLP	-3.26	3.49	
		Control	-4.45	2.54	0.29
Midface prominence	sl - n - sn	UCL	-1.38	2.42	
		UCLP	0.43	2.93	
		Control	-0.45	2.29	0.17
Nose height:	n - sn	UCL	-0.59	1.96	
		UCLP	-0.34	2.70	
		Control	-0.53	1.68	0.93
Nose bridge length:	n - prn	UCL	-0.60	2.17	
		UCLP	-0.24	2.80	
		Control	0.34	1.47	0.21
Nasal protrusion	sn - prn	UCL	0.65	1.68	
		UCLP	-0.02	1.74	
		Control	-0.60	0.95	0.01
Nasolabial angle:	prn - sn - ls	UCL	6.65	5.84	
		UCLP	7.51	7.99	
		Control	6.83	5.67	0.92
Nasal tip angle	n-prn-sn	UCL	-2.81	5.11	
		UCLP	-0.78	5.18	
		Control	-3.41	3.08	0.06
Anatomical width of nose:	acR - acL	UCL	2.21	1.78	
		UCLP	2.54	1.55	
		Control	2.54	1.35	0.78
Nasal Base Width	sbalR-sbalL	UCL	4.26	2.39	
		UCLP	3.73	1.71	
		Control	3.05	1.55	0.06
Width of right nostril floor:	sbalR - sn	UCL	1.44	1.42	
		UCLP	0.60	2.29	
		Control	1.54	0.82	0.03
Width of left nostril floor:	sbal L to sn	UCL	2.86	2.26	
		UCLP	3.38	2.44	
		Control	1.62	0.97	<0.001
Width of the columella:	sn'R - sn'L	UCL	-0.24	0.55	
		UCLP	-0.19	0.51	
		Control	0.02	0.58	0.21
Length of columella (right):	cR - sn	UCL	-0.27	1.02	
		UCLP	0.26	1.03	
		Control	-0.04	0.81	0.30
Length of columella (left):	cL - sn	UCL	-0.05	0.98	
		UCLP	-0.27	0.98	
		Control	-0.24	0.83	0.79

Table 4.27. Comparison of Differences between Upper Face Linear Measurements (mm) at Rest and Smiling in 11 UCL, 15 UCLP and 65 Control Cases

			UCL (N=11)	UCLP (N=15)	Control (N=65)
			Mean	Std Dev	p Value
Soft tissue nose width	alL - alR	UCL	1.23	2.01	
		UCLP	1.05	1.66	
		Control	1.74	1.40	0.22
Length of right ala:	acR - prn	UCL	0.38	1.64	
		UCLP	-1.19	1.84	
		Control	0.82	0.91	<0.001
Length of left ala:	acL - prn	UCL	1.75	2.11	
		UCLP	2.88	2.34	
		Control	0.89	0.74	<0.001
Soft tissue nose:mouth width:	alR-alL:chR-chL	UCL	-0.15	0.07	
		UCLP	-0.18	0.06	
		Control	-0.13	0.05	0.01
Anatomical nose:mouth width:	acR-acL:chR-chL	UCL	-0.15	0.07	
		UCLP	-0.16	0.07	
		Control	-0.13	0.06	0.12
Mouth width:	chR - chL	UCL	13.21	3.87	
		UCLP	12.67	4.11	
		Control	11.43	3.45	0.21
Upper lip height:	sn - sto(s)	UCL	-2.99	1.62	
		UCLP	-2.15	2.90	
		Control	-3.57	1.75	0.04
Upper cutaneous lip height:	sn - ls	UCL	-1.72	1.14	
		UCLP	-0.51	1.78	
		Control	-2.29	1.39	<0.001
Upper vermillion lip height:	ls-sto(s)	UCL	-1.63	1.84	
		UCLP	-1.63	2.17	
		Control	-1.74	1.67	0.96
Lower lip height:	sto - sl	UCL	-0.43	1.90	
		UCLP	0.01	1.96	
		Control	-0.57	1.35	0.43
Lower cutaneous lip height:	li - sl	UCL	-0.27	1.16	
		UCLP	0.87	2.02	
		Control	0.09	1.45	0.13
Lower vermillion height:	sto - li	UCL	-0.20	1.46	
		UCLP	-0.86	1.60	
		Control	-0.16	1.24	0.19
Midline - High Lip Point	ls - lhp	UCL	7.47	1.25	
		UCLP	6.77	2.25	
		Control	0.29	0.41	<0.001
Maximum Vermillion Height	lhp -llp	UCL	8.20	3.48	
		UCLP	6.31	2.29	
		Control	4.00	1.08	<0.001

Table 4.28. Comparison of Differences between Lower Face Linear Measurements (mm) at Rest and Smiling in 11 UCL, 15 UCLP and 65 Control Cases

		UCL-Control		UCLP-Control		UCLP-UCL	
Nasal protrusion	sn - prn	-2.19	-0.31	-1.41	0.25	-0.47	1.81
Length of right ala:	acR - prn	-0.49	1.37	1.19	2.83	0.43	2.70
Length of left ala:	acL - prn	-1.89	0.16	-2.90	-1.10	-2.38	0.11
Width of right nostril floor:	sbalR - sn	-0.86	1.07	0.09	1.79	-0.34	2.01
Width of left nostril floor:	sbal L to sn	-2.39	-0.08	-2.78	-0.75	-1.93	0.88
Soft tissue nose:mouth width:	alR-alL:chR-chL	-0.02	0.07	0.01	0.09	-0.03	0.08
Upper lip height:	sn - sto(s)	-2.11	0.95	-2.75	-0.07	-2.69	1.03
Upper cutaneous lip height:	sn - ls	-1.69	0.55	-2.75	-0.79	-2.56	0.16
Midline - High Lip Point	ls - lhp	-8.01	-6.36	-7.20	-5.75	-0.30	1.71
Maximum Vermillion Height	lhp -llp	-5.55	-2.85	-3.50	-1.12	0.24	3.54

Blue Ink indicates Significant Result

Table 4.29. Pairwise Comparison of Differences between Linear Measurements (mm) at Rest and Smiling in 11 UCL, 15 UCLP and 65 Control Cases

The most obvious added deformity when smiling was in the upper lip area. In the control cases, the upper lip high and low points were in the midline as was seen from the negligible distance between the labrale superiorus and lip high point, i.e. these points were co-incident. The distance between the high and low lip points was the same as the upper vermillion lip height measured in the midline in the control cases.

In the children with either type of cleft, the high point of the lip was significantly removed from the midline and located on the side of the cleft. The distance between the eccentric high lip point and the lip low point was greater than the vermillion lip height measured in the midline in the cleft cases. This would suggest there was abnormal function of the repaired lip with drooping of the mucocutaneous junction on the affected side of the upper lip. At the approximate site of the cheiloplasty, there was marked asymmetry of the upper lip line during an maximal smile.

4.3.3.2. Three-Dimensional Analysis

The groups were analysed according to resting lip pose and presence or absence of cleft lip, to examine the effect of cheiloplasty on the lip outline in function. Tables 4.30. and 4.31. summarise the findings.

The 3D displacement of key landmarks in cleft subjects with alteration in expression from rest to maximal smile was similar to that demonstrated in control subjects. The cleft cases also showed very minor sub-millimetre displacement of endocanthion and subnasale landmarks, which had been assumed to be stable for simplification of 3D analysis.

Intercanthal Area

There was a statistically significant difference in left endocanthion displacement between cleft and control samples in cases of initially closed lip pose. A greater proportion of the more numerous control subjects produced the

	Rest (Closed Lips) to Smile				
		X	Y	Z	3D
Cheilion Left	Median Displacement in Cleft Sample	5.2	5.5	-3.9	11.3
Cheilion Left	Median Displacement in Control Sample	5.8	4.3	-6.4	12.3
Cheilion Left	Wilcoxon p-Value	0.88	0.75	0.07	0.95
Cheilion Right	Median Displacement in Cleft Sample	5.8	4.4	-6.1	12.9
Cheilion Right	Median Displacement in Control Sample	5.8	3.8	-6	11
Cheilion Right	Wilcoxon p-Value	0.71	0.84	0.81	0.31
Labrale Superiorus	Median Displacement in Cleft Sample	-0.2	3.6	-0.6	5
Labrale Superiorus	Median Displacement in Control Sample	-0.1	3.4	-2.5	4.9
Labrale Superiorus	Wilcoxon p-Value	0.38	0.76	0.06	0.82
Endocanthion Right	Median Displacement in Cleft Sample	0.3	-0.4	0.3	0.7
Endocanthion Right	Median Displacement in Control Sample	0.1	-0.4	0.3	0.8
Endocanthion Right	Wilcoxon p-Value	0.13	0.64	0.58	0.93
Endocanthion Left	Median Displacement in Cleft Sample	-0.3	0	0	0.8
Endocanthion Left	Median Displacement in Control Sample	-0.2	-0.4	0.3	0.6
Endocanthion Left	Wilcoxon p-Value	0.49	0.02	0.01	0.2
Subnasale	Median Displacement in Cleft Sample	0	0.4	-0.3	0.7
Subnasale	Median Displacement in Control Sample	0	0.7	-0.5	0.9
Subnasale	Wilcoxon p-Value	0.3	0.11	0.11	0.5

Table 4.30. Displacement (mm) of Ocular and Perioral Landmarks in Smile in Control Cases and Cleft Cases (with lips closed at rest)

Rest (Lips Apart) to Smile		X	Y	Z	3D
Cheilion Left	Median Displacement in Cleft Sample	6.4	10.9	-5.3	14.8
Cheilion Left	Median Displacement in Control Sample	6	5.3	-7.7	11
Cheilion Left	Wilcoxon p-Value	0.27	0.005	0.11	0.03
Cheilion Right	Median Displacement in Cleft Sample	7.3	10.8	-3.7	15.9
Cheilion Right	Median Displacement in Control Sample	5.3	4.4	-6.3	10.5
Cheilion Right	Wilcoxon p-Value	0.2	0.01	0.13	0.01
Labrale Superiorus	Median Displacement in Cleft Sample	0.5	2.4	-2.3	4.3
Labrale Superiorus	Median Displacement in Control Sample	0.5	2.9	-2.9	4.6
Labrale Superiorus	Wilcoxon p-Value	0.84	0.84	0.18	0.31
Endocanthion Right	Median Displacement in Cleft Sample	-0.2	-0.4	0.2	0.7
Endocanthion Right	Median Displacement in Control Sample	0	-0.4	0.2	0.6
Endocanthion Right	Wilcoxon p-Value	0.18	0.78	0.84	0.27
Endocanthion Left	Median Displacement in Cleft Sample	0.2	-0.4	0.3	0.7
Endocanthion Left	Median Displacement in Control Sample	-0.1	-0.6	0.4	0.7
Endocanthion Left	Wilcoxon p-Value	0.35	0.97	0.31	1
Subnasale	Median Displacement in Cleft Sample	0	0.8	-0.5	1
Subnasale	Median Displacement in Control Sample	0	0.9	-0.6	1.1
Subnasale	Wilcoxon p-Value	0.84	0.6	0.31	0.6

Table 4.31. Displacement (mm) of Ocular and Perioral Landmarks in Smile in Control Cases and Cleft Cases (with lips apart at rest)

desired closed lip pose so this statistically significant difference could be attributed in part to the differing sample sizes. The statistically significant differences were less than 1mm. This shows the sensitivity of 3D shape analysis. This finding of sensitivity was replicated when the shape of the dental arch was examined.

Commissures

The displacement vectors for the commissures were similar for the cleft sample and the control sample in the groups which were photographed with the lips closed at rest. In moving from a lips-apart resting pose to a maximal smile, there was statistically significant greater displacement of the commissures in cleft cases than in control cases. The median displacement of the right and left commissures was 14.8 mm and 15.9 mm in the cleft cases. In the control cases, the commissure displacements were 11.0 mm on the left and 10.5 mm on the right.

The differences appeared to be due to the vertical component of displacement. In the cleft cases there was over 10 mm of upward displacement bilaterally compared with less than 5.5 mm of upward displacement in the control cases.

In both cases, the movement was away from the midline and away from the affected side of the face consistent with anatomical movement, suggesting there was no synkinetic movement of the unaffected commissure.

Upper Lip Border

There was no significant difference in the displacement of the midline of the upper vermillion border between the cleft and control samples, the Ls landmark.

As was reported in the control samples, there was less than 1 mm horizontal displacement along the X axis. The principal component of the 3D displacement vector was upward displacement along the Y axis. There was greater upward displacement of the midline landmark in moving from a resting pose with lips-closed to smile than in moving from a lips-apart resting pose to smile, in both cleft and control samples.

There was less posterior displacement of the upper vermillion midline in the cleft sample than in the control sample when moving from a lips-closed resting expression to maximal smile, 0.6 mm rather than 2.5 mm. This difference was not statistically significant. Comparison of the samples photographed at rest with the lips apart did not replicate this finding, posterior displacement was over 2.4 mm in the cleft sample and 2.9 mm in the control sample on maximal smile.

Chapter 5

Results of Dental Arch Analysis

5.5. Dental Arch Analysis

5.5.1. Dental Profile of Study Samples

5.5.1.1. Incisor Classification

The profile of the recruited subjects is shown in Table 5.1. Most of the UCL subjects had a Class I incisor relationship with 1 having a Class III relationship and only 2 exhibiting a Class II Division I incisor relationship. Five of the 16 UCLP subjects had a Class III incisor relationship but most were Class I. None of the control subjects had a Class III incisor relationship. The most commonly observed incisor relationship in the control subjects was Class I with 47.4% of the sample in this category. A Class II Division I relationship was seen in another significant percentage, 39.7%. No mandibular displacement on closure was detected in any subject.

5.5.1.2. Crossbite

Crossbite was buccal except where a lingual crossbite is stated (Table 5.2.). A buccal crossbite exists when the buccal cusps of the mandibular molars occlude laterally to the buccal cusps of the maxillary molars. A lingual crossbite is described by the lingual cusps of the mandibular molars occluding lingually to the palatal cusps of the maxillary molars. The crossbites were anterior, with posterior extension in cases of larger number of affected maxillary teeth. There were no isolated posterior buccal crossbites in any subjects. No crossbites had an associated mandibular displacement on closure. Some of the UCL and UCLP subjects had more than one type of crossbite; thus, the sum of percentages exceeds 100.

Over 50% of the UCL subjects had no teeth in crossbite. A total of 36.4% had between 1 and 3 teeth in crossbite and 1 subject had a combination of 1 tooth in anterior crossbite and 2 teeth in posterior lingual crossbite.

	UCL		UCLP		Control	
	N=11	%	N=16	%	N=78	%
Class I	8	72.7	7	43.7	37	47.4
Class II Div. I	2	18.2	0		31	39.7
Class II Div. II	0		1	6.3	6	7.7
Class III	1	9.1	5	31.3	0	
No lower impression	0		3	18.7	4	5.2

Table 5.1. Incisor Classification in 11 UCL, 16 UCLP and 78 Control Cases

	UCL		UCLP		Control	
Number of Teeth Affected	N=11	%	N=16	%	N=78	%
0	6	54.5	1	6.3	66	84.6
1	1	9	4	25.2	1	1.3
2	2	18	3	18.9	1	1.3
3	1	9	1	6.3	4	5.2
4	0		1	6.3	0	
5	0		1	6.3	0	
7	0		1	6.3	1	1.3
9	0		1	6.3	0	
Lingual Crossbite 1	0		0		0	
Lingual Crossbite 2	2	18	1	6.3	0	
Longual Crossbite 3	0		0		0	
Lingual Crossbite 4	0		0		0	
No lower impression	0		3		4	5.2

Table 5.2. Crossbite Profile of 11 UCL, 16 UCLP and 78 Control Cases

Only 6.3% of the UCLP subjects had no buccal crossbites. One tooth was in crossbite in 25.2 % of cases, 2 teeth in another 18.9% of cases but 1 case had 7 teeth affected and another case had 9 affected teeth. One case had 1 tooth in anterior crossbite and 2 teeth in posterior lingual crossbite.

The control cases were unaffected by teeth in crossbite in 85.9% of cases. Between 1 and 3 teeth were affected by crossbite in 5.8% of cases and only 1 case had a crossbite affecting 7 teeth which was as severe as the second most severely affected UCLP case. One case had 4 teeth in lingual crossbite, the right and left maxillary primary molars.

5.5.1.3. Erupted Supernumerary Teeth

Only the UCL sample had a significant incidence of erupted supernumerary teeth (Table 5.3.). In each case, the side of the arch carrying the additional teeth and the side of the face affected by the cleft lip co-incided.

5.5.1.4. Decayed, Missing and Filled Teeth Score

The decayed, missing and filled teeth (DMF) score profile shows the reasons for a score of more than zero (Table 5.4.). None of the children had untreated caries.

Only 1 case of UCL had an absent incisor related to the cleft. Unerupted molars accounted for the other 2 cases of UCL with a DMF score of greater than zero.

One or more incisors were absent adjacent to the cleft in 75.6% of the UCLP cases. Treated caries was seen in 25.2% of the UCLP cases, the only children in the study who had restorations placed.

The control population was caries free on visual inspection. The only reason for a DMF score of greater than zero was uneruption of second primary molars.

	UCL		UCLP		Control	
Number of additional teeth	**N=11**	**%**	**N=16**	**%**	**N=78**	**%**
0	9	81.8	15	93.8	78	100
1	2	18.2	1	6.2	0	
>1	0		0		0	

Table 5.3. Erupted Supernumerary Teeth in 11 UCL, 16 UCLP and 78 Control Cases

	UCL		UCLP		Control	
	N=11	**%**	**N=16**	**%**	**N=78**	**%**
0	8	72.3			73	93.4
1 u/e	1	9.1	0		1	1.3
2 u/e	0		1	5.9	3	4
3 u/e	0		0		0	
4 u/e	1	9.1	1	5.9	1	1.3
1 absent	1	9.1	10	59	0	
2 absent	0		2	11.8	0	
3 absent	0		0		0	
2 restored	0		1	5.9	0	
3 restored	0		2	11.8	0	
4 restored	0		1	5.9	0	

Table 5.4. Decayed, Missing and Filled Teeth Profile of 11 UCL, 16 UCLP and 78 Control Cases

5.5.2. Analysis of Linear Dental Arch Dimensions

5.5.2.1. Maxillary Arch

5.5.2.1.1.Comparisons of Arch Dimensions between Subject Types

Except for the second intermolar width, the linear maxillary arch dimensions in cleft cases were significantly different from the control group (Table 5.5).

Pairwise comparison is shown in Table 5.6. This demonstrated that the UCL cases did not differ significantly from the control cases. Significant differences existed between the UCL cases and the UCLP cases with all dimensions, except second intermolar width, greater in the UCL cases. The figures in blue print indicate the range in millimetres by which the control dimensions exceeded the same dimensions in the UCLP arches

The largest difference was between the left (affected) anterior quadrant lengths. The difference in millimetres between UCL and UCLP, as a proportion of mean lengths in UCL and UCLP, was the greatest for this dimension.

In UCLP cases, the arch circumference was shorter, the arch depth was shallower, the left anterior quadrant length was shorter and the intercanine width was also narrower than in control and UCL cases. All of the differences listed achieved statistical significance (p value <0.02). Pairwise comparison between UCLP and control cases demonstrated a large difference between left anterior quadrant lengths. For this dimension, the difference between UCLP and control cases was less than the difference between UCLP and UCL as the mean left anterior quadrant length was greatest in the UCL cases.

	UCL	(N=11)	UCLP	(N=16)	Control	(N=76)	
	Mean	Std Dev	Mean	Std Dev	Mean	Std Dev	p value
Arch Circumference	70.06	4.19	64.65	4.41	70.58	3.22	<0.001
Arch Depth	27.36	2.51	23.99	2.85	27.82	1.74	<0.001
Intercanine Width	28.74	3.19	24.35	2.44	27.83	1.82	<0.001
First Intermolar width	31.97	2.78	28.90	2.92	31.02	1.87	0.00
Second Intermolar width	37.87	2.98	36.09	3.01	36.72	1.95	0.16
Left Anterior Quadrant length	20.07	3.17	14.91	2.44	19.05	1.10	<0.001
Right Anterior Quadrant Length	18.72	1.26	17.20	1.28	18.82	0.97	<0.001

Table 5.5. Comparison of Maxillary Arch Dimensions (mm) in 11 UCL, 16 UCLP and 76 Controls

	UCL vs. Control		UCLP vs. Control		UCLP vs. UCL	
Arch Circumference	-2.42	3.47	3.50	8.36	1.84	8.97
Arch Depth	-1.16	2.07	2.43	5.23	1.39	5.36
Intercanine Width	-2.52	0.69	2.11	4.84	2.44	6.34
First Intermolar width	-2.61	0.70	0.70	3.52	1.06	5.08
Second Intermolar width	-2.95	0.66	-0.89	2.15	-0.41	3.96
Left Anterior Quadrant length	-2.31	0.26	3.04	5.24	3.60	6.72
Right Anterior Quadrant Length	-0.71	0.91	0.93	2.31	0.54	2.50

Blue ink indicates significant result

Table 5.6. Pairwise Comparisons of Maxillary Arch Dimensions in 11 UCL, 16 UCLP and 76 Controls

5.5.2.2. Mandibular Arch

5.5.2.2.1. Comparisons of Arch Dimensions between Subject Types

There were no significant differences in size between mandibular arch linear dimensions in any of the subject types (Table 5.7.). The cleft-related maxillary arch deformity in size did not appear to have any related mandibular arch deformity.

5.5.2.3. Maxillary and Mandibular Arches

The ratios between the maxillary and mandibular arch circumferences and arch depths were also significantly different for the subject types (Table 5.8.). Pairwise comparison between UCL and control cases showed no difference between the ratios of the longer maxillary arch circumferences to the shorter mandibular arch circumferences (Table 5.9.). Figures in blue print indicate significantly larger ratios in the control or UCL cases relative to the UCLP cases. The ratios between the greater maxillary arch depths and lesser mandibular arch depths showed no difference, between UCL and control cases, on pairwise comparison. In both of the latter groups, the maxillary to mandibular arch depth ratio was greater than the maxillary to mandibular arch circumference ratio.

Comparison of UCL and UCLP cases showed significant differences in maxillary to mandibular arch ratios. The inter arch ratios in the UCLP cases were smaller, approaching 1:1 for both circumference and depth.

Comparison of UCLP and control cases found the same significant differences, with smaller upper to lower arch ratios in the UCLP cases than in the control cases.

	UCL	(N=10)	UCLP	(N=12)	Control	(N=69)	
	Mean	Std Dev	Mean	Std Dev	Mean	Std Dev	p value
Arch Circumference	63.51	4.28	62.74	4.14	64.21	3.05	0.34
Arch Depth	24.12	1.96	22.85	2.09	23.92	1.72	0.16
Intercanine Width	22.85	1.61	22.09	1.88	22.54	1.59	0.51
First Intermolar width	26.85	2.33	26.11	1.99	26.35	1.86	0.63
Second Intermolar width	33.98	2.40	33.24	1.92	32.96	1.81	0.27
Left Anterior Quadrant length	14.84	1.04	14.29	1.13	14.50	0.92	0.39
Right Anterior Quadrant Length	14.35	1.11	13.90	1.12	14.36	0.98	0.31

Table 5.7. Comparison of Mandibular Arch Dimensions (mm) in 10 UCL, 12 UCLP and 69 Controls

	UCL	(N=10)	UCLP	(N=11)	Control	(N=68)	
	Mean	Std Dev	Mean	Std Dev	Mean	Std Dev	p value
Arch Circumference U:L	1.10	0.06	1.02	0.04	1.10	0.04	< 0.001
Arch Depth U:L	1.13	0.06	1.02	0.06	1.17	0.06	< 0.001

Table 5.8 Comparison of Maxillary : Mandibular Arch ratios for 11 UCL, 10 UCLP and 68 Control cases

	UCL vs Control		UCLP vs Control		UCLP vs UCL	
Arch Circumference U:L	-0.035	0.035	0.048	0.115	0.036	0.127
Arch Depth U:L	-0.019	0.082	0.095	0.193	0.047	0.178
Blue ink indicates significant result						

Table 5.9. Pairwise Comparison of Maxillary : Mandibular Arch Ratios for 11 UCL, 10 UCLP and 68 Control Cases

5.5.2.4. Summary of Two Dimensional Arch Analysis

Compared to the control group, deformity in size was present in the maxillary arches of subjects with UCLP. The dental arches of subjects with UCL demonstrated no significant deformity of size compared to the control sample. The deformity of the maxillary arch in UCLP was closely allied with the incidences of Class III incisor classification and buccal crossbite rates in this group (Table 5.1).

Two-dimensional analysis detected apparent foreshortening of the anterior quadrant on the cleft affected side of the arch which clearly shows the inability of linear distances to describe form. Visual inspection of the maxillary arch in the UCLP subjects showed notching of the arch and of the occlusal plane in the region of the repaired cleft and the possible bony defect. This explained the shorter straight line distance measured between the distal of the canine and mesial of the central incisor. Linear dimensional analysis failed to demonstrate the quality and quantity of the clinically significant deviation from the control arch form seen in the UCLP maxillary arch.

The left anterior quadrant length appeared to be the arch dimension with the greatest powers of discrimination between dental arch size in the 3 subject types.

5.5.3. Three Dimensional Dental Arch Analysis

The differences between arch forms for subject types are summarised in Tables 5.10. and 5.11. Differences in shape between subject types were found where no differences in size had been demonstrated. Arch form cannot be summarised as a single number and must be presented graphically. The differences between average arch forms can be described in terms of individual tooth positions relative to the control arch forms which were the reference arches for 3D superimposition.

	All Arch Landmarks p values	Canine to Canine Arch Landmarks p values
UCL vs. Control	<0.001	<0.001
UCLP vs. Control	<0.001	<0.001
UCL vs. UCLP	0.993	0.687

Figure 5.10. Comparison of 3D Maxillary Arch Forms for 10 UCL, 10 UCLP and 61 Controls

	All Arch Landmarks p values	Canine to Canine Arch Landmarks p values
UCL vs. Control	0.050	0.076
UCLP vs. Control	0.006	0.002
UCLP vs. UCL	0.997	0.992

Table 5.11. Comparison of 3D Mandibular Arch Forms for 10 UCL, 10 UCLP and 61 Controls

5.5.3.1. Maxillary Arch

5.5.3.1.1. Comparison of UCL and Control Arch Forms

X-Y comparison - arch form viewed from occlusal - Figure 5.1 - In the UCL arch the mesial aspects of the left and right central incisors were rotated palatally and labially respectively, opening a wider space between the contact points. The right central and lateral incisors were both rotated distopalatally. The canine tips are very close to coincident in both arches.

X-Z comparison - arch form viewed from posterior - Figure 5.2 - In the UCL arch, there was distal tilting of the right lateral incisor.

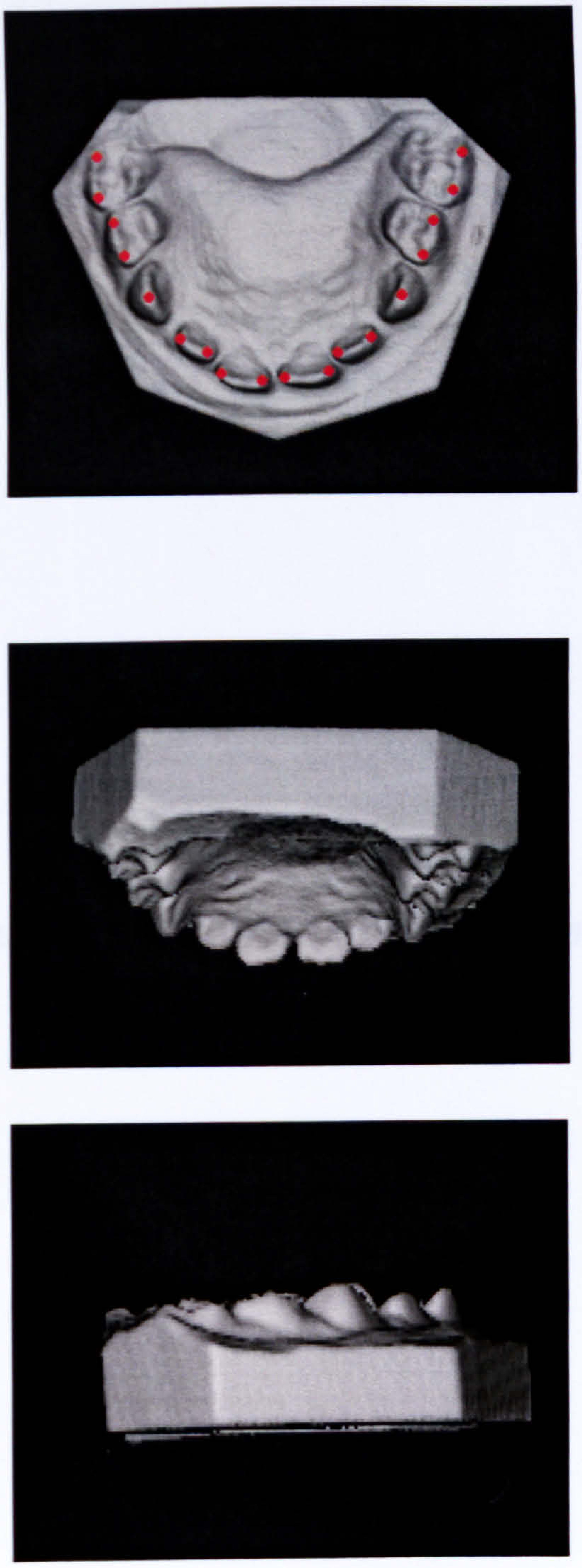
Y-Z comparison - arch form viewed from lateral - Figure 5.3 - The distal tilting of the right lateral incisor is confirmed in this view.

5.5.3.1.2. Comparison of UCLP and Control Arch Forms

X-Y comparison - arch form viewed from occlusal - Figure 5.4 - There is a shorter space between canine tip and left central incisor seen with distolabial rotation of the left central incisor, creating a median diastema. The left UCLP canine is mesiopalatally located. There is distopalatal rotation of both right incisors but the right canine tip in the UCLP arch is located labial to the control canine tip.

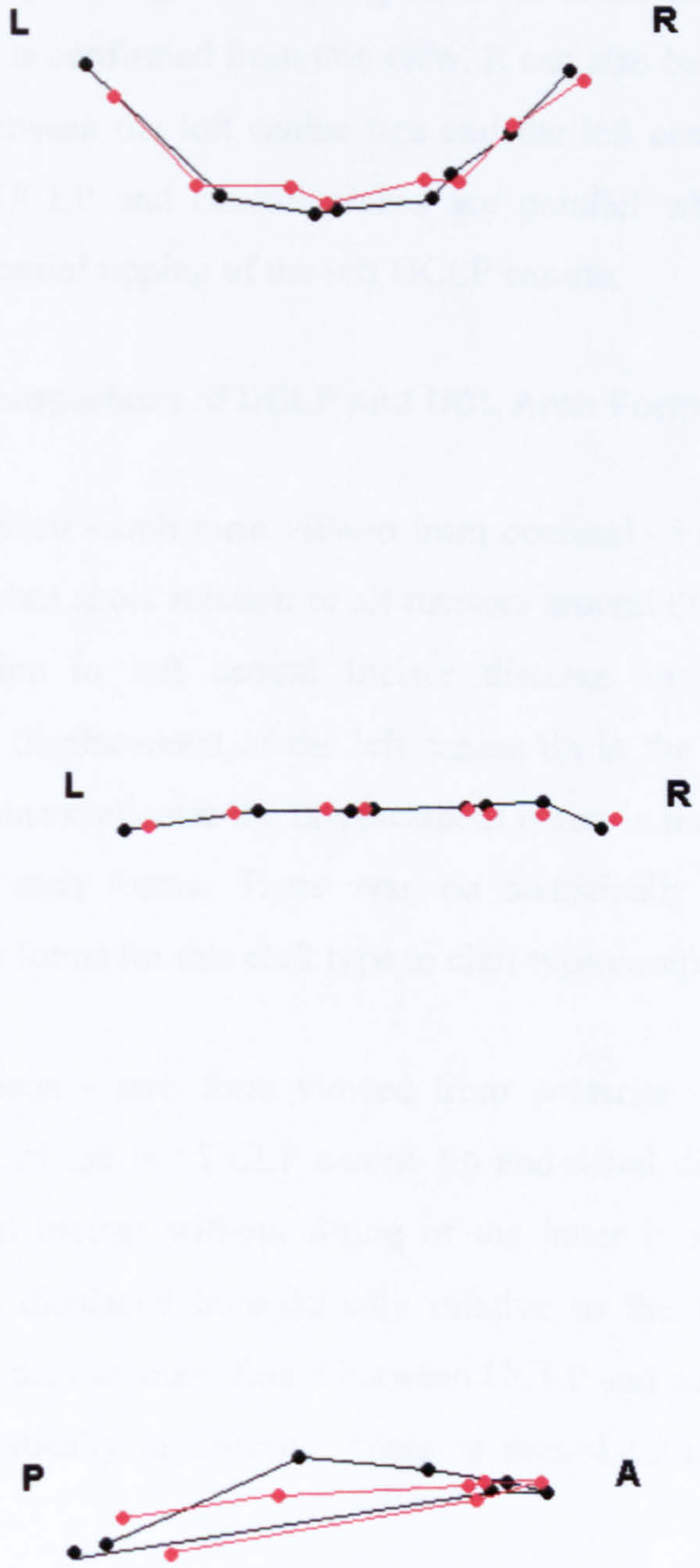
X-Z comparison - arch form viewed from posterior - Figure 5.5 - There is no distal tilting of the left central incisor seen. The left canine tips are at the same level on the Z axis. The displacement of the left canine in the UCLP arch is confirmed.

On the right there is distal tilting of the right lateral incisor in the UCLP arch and the right canine tip in the UCLP arch is located incisolabially to the canine



Figures 5.1. to 5.18 - Reference Views

Figures 5.1. to 5.3. Maxillary Arch Form Comparison - UCL (Blue) with Control Cases (Black)



Figures 5.4. to 5.6. - Maxillary Arch Form Comparison - UCLP (Red) with Control Cases (Black)

tip in the control arch.

Y-Z comparison - arch form viewed from lateral - Figure 5.6 - The right lateral incisor tipping and right canine displacement in the UCLP arch relative to the control arch is confirmed from this view. It can also be seen that the Euclidean distances between the left canine tips and the left central incisor distoincisal corners in UCLP and control arches are parallel which might suggest the absence of mesial tipping of the left UCLP canine.

5.5.3.1.3. Comparison of UCLP and UCL Arch Forms

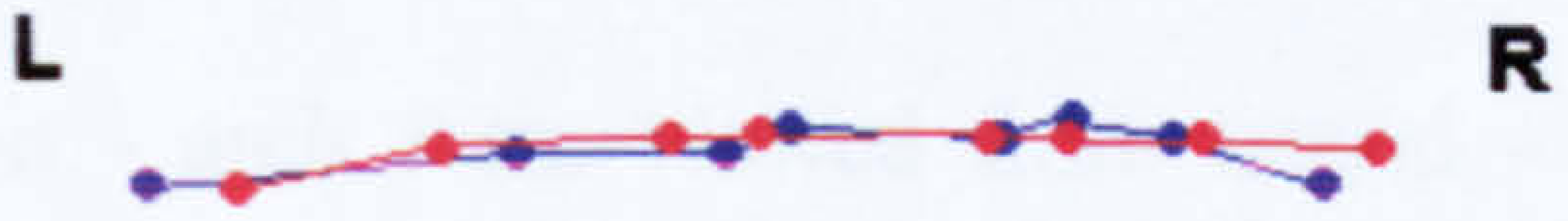
X-Y comparison - arch form viewed from occlusal - Figure 5.7. - both UCLP and UCL arches show rotation of all incisors around the same axes. There is a smaller canine to left central incisor distance in the UCLP arch. The mesiopalatal displacement of the left canine tip in the UCLP arch relative to the UCL canine replicates the displacement noted in the comparison of UCLP and control arch forms. There was no statistically significant difference between arch forms for this cleft type to cleft type comparison.

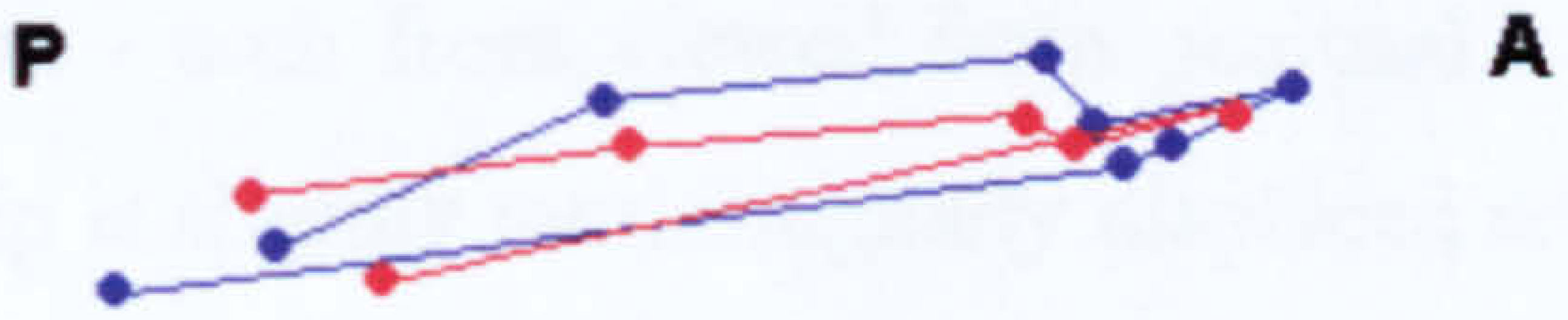
X-Z comparison - arch form viewed from posterior - Figure 5.8. - Palatal displacement of the left UCLP canine tip and distal displacement of the left UCLP central incisor without tilting of the latter is seen. The right UCLP canine tip is displaced incisolabially relative to the UCL canine tip. This replicates the displacement found between UCLP and control right canines but was not statistically significant. There is mesial tilting of the right UCLP lateral incisor .

Y-Z comparison - arch form viewed from lateral - Figure 5.9 - The mesial tilting of the right UCLP lateral incisor and the incisal displacement of the right UCLP canine in confirmed.

5.5.3.2. Mandibular Arch

The sensitivity of methods of 3D analysis to differences in form was





Figures 5.7. to 5.9. - Maxillary Arch Form Comparison - UCLP (Red) with UCL Cases (Blue)

demonstrated in the comparison of mandibular arch forms. Subtle differences, of negligible clinical significance, were of marked statistical significance Table 5.11.

5.5.3.2.1. Comparison of UCL and Control Arch Forms

There were no statistically significant differences detected between the average arch forms, from canine to canine, of the UCL and control cases.

X-Y comparison - arch from viewed from occlusal - Figure 5.10 - There were no apparent differences.

X-Z comparison - arch form viewed from posterior - Figure 5.11 - There were no apparent differences.

Y-Z comparison - arch form viewed from lateral - Figure 5.12 - There is relative flattening of the curve of the arch from canine to central incisors seen in the UCL arch.

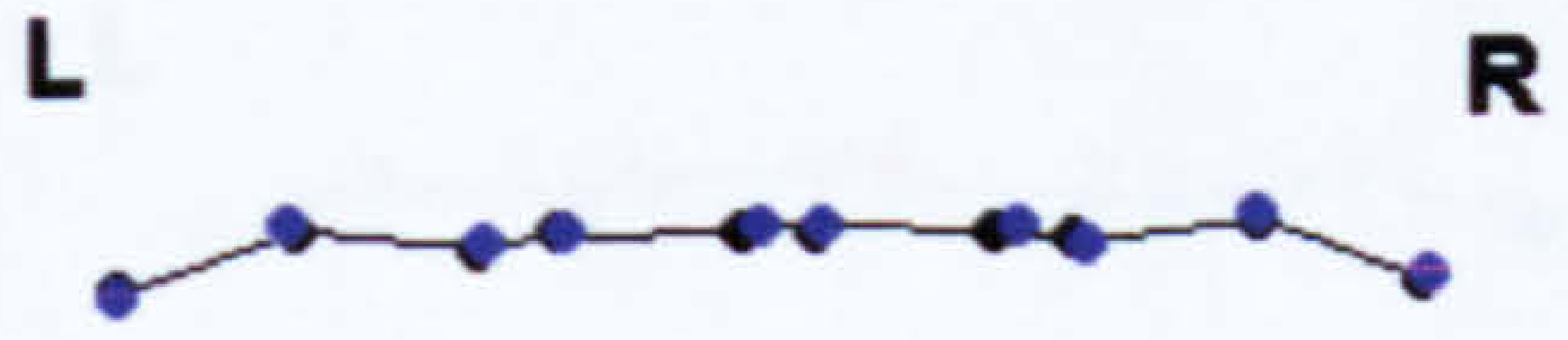
5.5.3.2.2. Comparison of UCLP and Control Arch Forms

There were statistically significant differences between the average arch forms of UCLP and control cases.

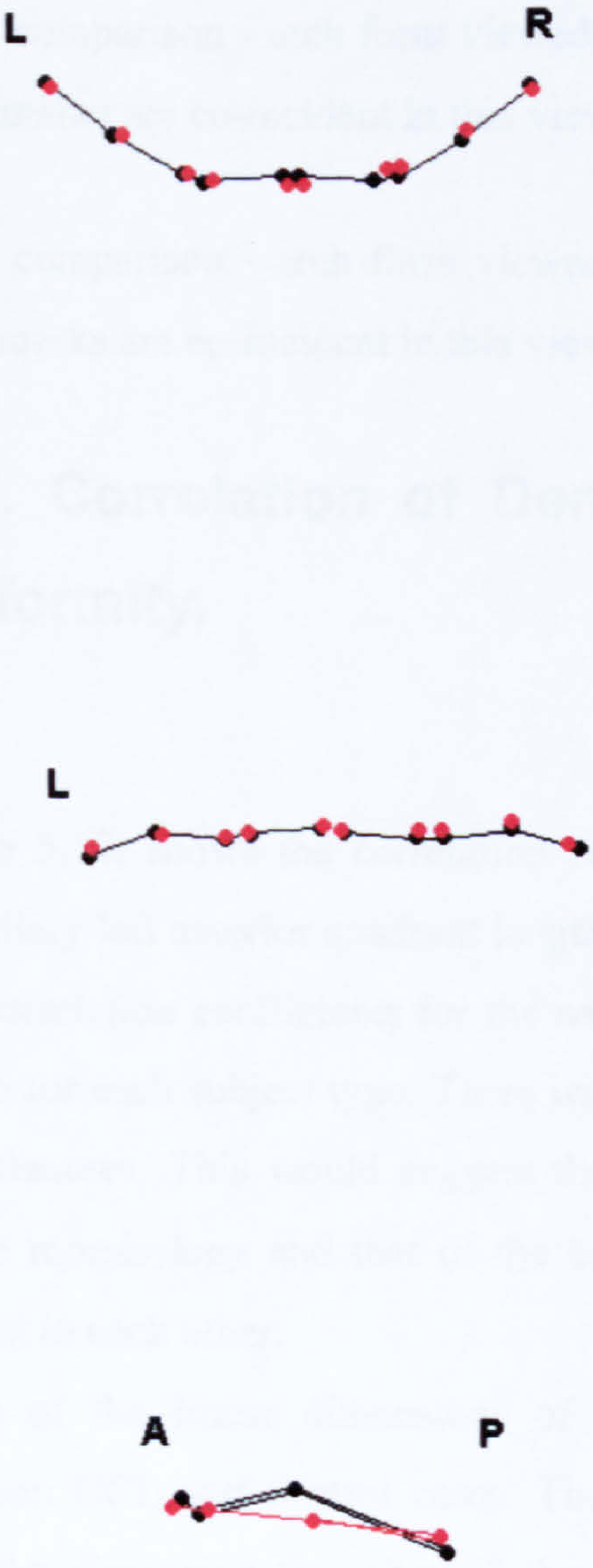
X-Y comparison - arch from viewed from occlusal - Figure 5.13 - The left UCLP canine tip is slightly mesiolingually displaced relative to the left control canine in the arch form.

X-Z comparison - arch form viewed from posterior - Figure 5.14 - There were no apparent differences.

Y-Z comparison - arch form viewed from lateral - Figure 5.15 - There is relative flattening of the curve of the arch from canine to central incisors seen in the UCLP arch.



Figures 5.10. to 5.12. Mandibular Arch Form Comparison - UCL (Blue) with Control Cases (Black)



Figures 5.13. to 5.15. Mandibular Arch Form Comparison - UCLP (Red) with Control Cases (Black)

5.5.3.2.3. Comparison of UCLP and UCL Arch Forms

X-Y comparison - arch from viewed from occlusal - Figure 5.16 - There were no apparent differences.

X-Z comparison - arch form viewed from posterior - Figure 5.17 - The dental landmarks are co-incident in this view

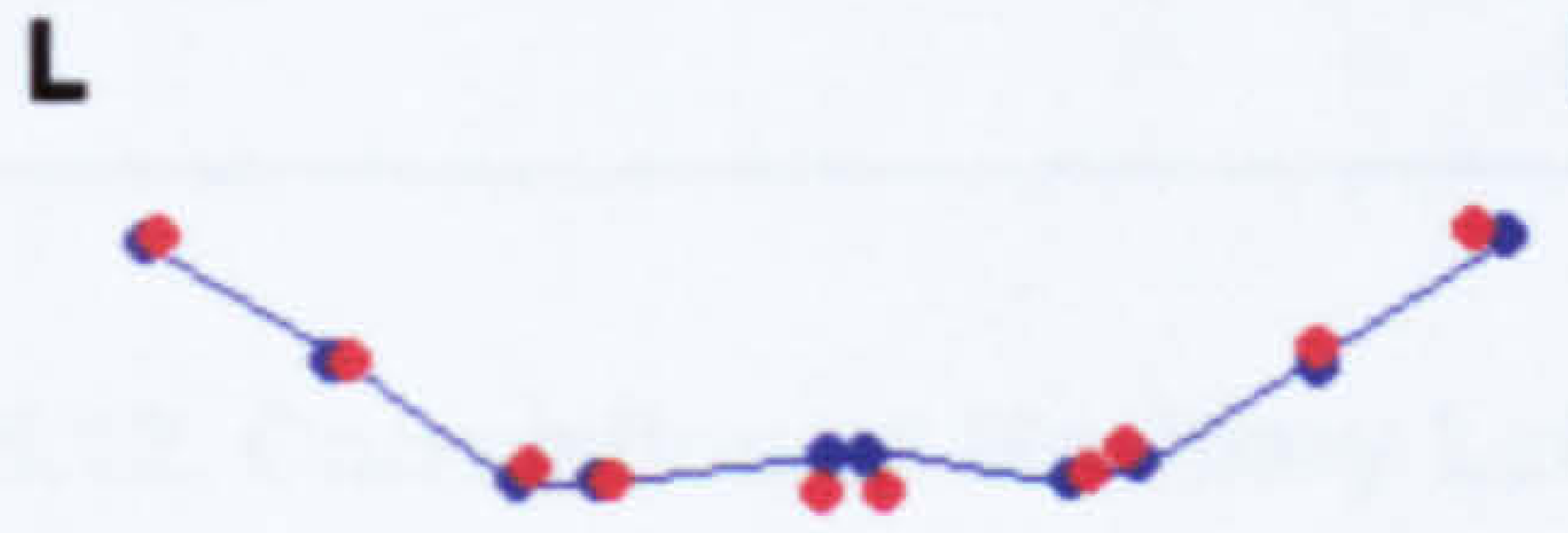
Y-Z comparison - arch form viewed from lateral - Figure 5.18 - The dental landmarks are co-incident in this view.

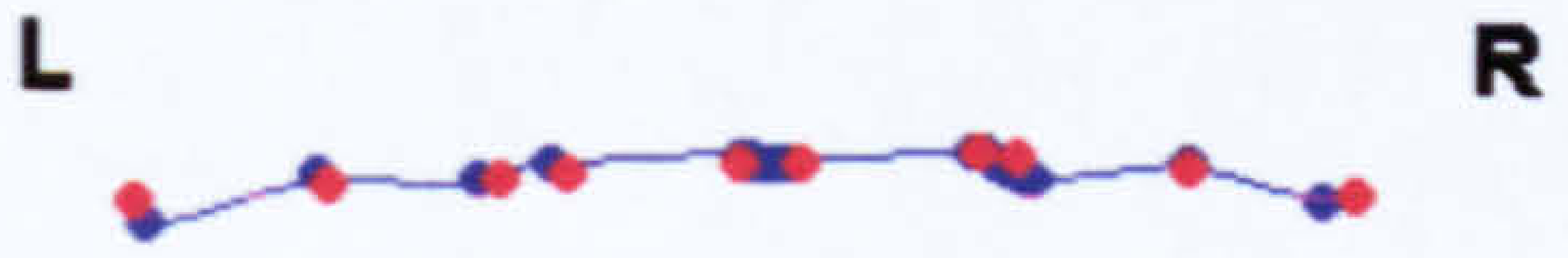
5.5. Correlation of Dental Arch and Soft Tissue Deformity.

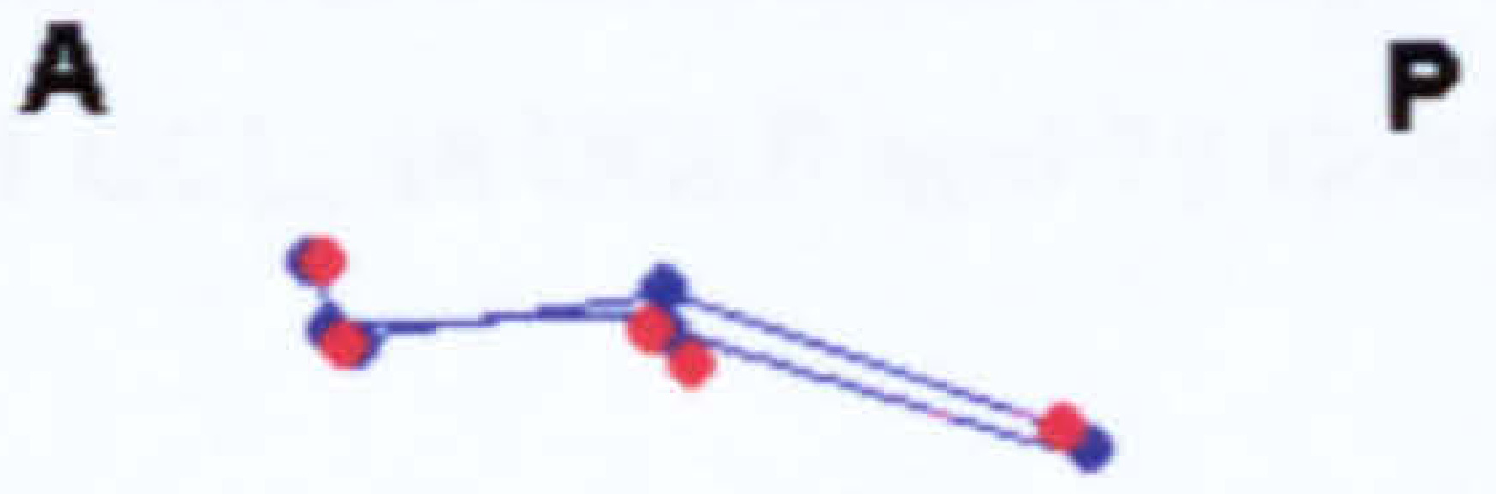
Table 5.12. shows the correlation coefficients for the nasal base width and maxillary left anterior quadrant length for each subject type. Table 5.13. shows the correlation coefficients for the nasal base width and maxillary intercanine width for each subject type. There was no evidence of correlation between the two features. This would suggest that the cleft-related deformity of the soft tissue morphology and that of the bone based dental arches are not directly related to each other.

None of the linear dimensions of the arches were significantly different between UCL and control cases. The maxillary left anterior quadrant length was the dimension that showed the largest proportional difference between UCLP and control cases and between UCLP and UCL cases so this was chosen as the feature with the greatest power of discrimination between subject types in 2D analysis.

As the 3D location of the anthropometric landmarks and the 3D dental arch configurations are different entities, it was not possible to test the correlation of the two shapes. Each could be defined as a single point in Kendall's shape space but the distance between the two scatters in shape space would far







Figures 5.16. to 5.18. Mandibular Arch Form Comparison - UCLP (Red) with UCL Cases (Blue)

	Correlation Coefficient	p value
UCL	-0.021	0.952
UCLP	0.4	0.139
Controls	0.117	0.325

Table 5.12. Correlation of Maxillary Left Quadrant Length and Nasal Base Width in 11 UCL, 16 UCLP and 78 Control Cases

	Correlation Coefficient	p value
UCL	0.159	0.64
UCLP	0.276	0.319
Controls	0.197	0.095

Table 5.13. Correlation of Maxillary Intercanine Width and Nasal Base Width in11 UCL, 16 UCLP and 78 Control Cases

outweigh the variation within each scatter. Kendall's shape space is a geometric construction underlying geometric morphometrics. In shape space the shape of a figure is represented by a single point. Therefore, scatters of points correspond to scatters of entire landmark configurations, not merely scatters of single landmarks (Dryden & Mardia, 1998).

Chapter 6

Discussion

6.1. Imaging Techniques and Co-ordinate Extraction for Facial Soft Tissues and Dental Study Models

The initial review of the literature in the field of direct and indirect anthropometry concluded that appropriateness and accuracy were the key features to be considered when choosing a method of acquiring 3D landmark co-ordinates or assessing interlandmark distances. Computerised stereophotogrammetry and CT scanning were the chosen imaging techniques, in this study, for recording and evaluating the surface detail of the facial soft tissue in young children and that of small, stone dental study models. The outcome from both imaging modalities was the same; sets of 3D co-ordinates corresponding to named landmarks. These landmark sets were manually identified on screen in both cases.

The advantages of computerised stereophotogrammetry and CT scanning of dental casts utilised in this study and landmark digitisation will now be discussed.

6.1.1. Computerised Stereophotogrammetry

The major advantage of this technique is the speed of data collection, which permitted the application of the imaging technique to the paediatric population. This allowed the recording of images of 124 3-year-old children at rest and while each performed a maximal smile.

A similar system based on a liquid crystal shutter, called a liquid crystal range finder, has also been employed in the investigation of children as young as 3 months (Yamada et al 2002a). This form of non-invasive imaging would appear to be the most promising technique for overcoming the lack of co-operation likely in small children. The liquid crystal range finder requires two successive images to be captured, at differing angulations of the head, to record detail in the nasal base area. Each image takes 1 second to capture but there must be an additional time requirement to reposition either the equipment or the subject which must extend the capture time beyond 2 seconds. This question was not discussed by Yamada et al (2002).

The data collected by the liquid crystal range finder have been shown to be independent of head position, up to extremes of 45 degrees away from the natural resting head position (Yamada et al 1999). The computerised stereophotogrammetry system employed in this study was shown to collect reproducible configurations of landmarks over a range of 60 degrees of head tilt in an inanimate object. This, however, does not allow one to conclude that the position of the head is unimportant. The whole facial soft tissue may alter in shape as the tissue of the lower face is stretched or compressed with extension and flexion of the cervical spine, even if a small number of anthropometric landmarks remain fixed relative to each other.

The assumption that head position should be standardised has been made previously. Studies, to validate methods of head fixation, have been performed which did not attempt to assess the significance of head position or to examine the effect of head position on the soft tissue surface (Soncul & Bamber 2000). In the study reported here, head position was not fixed rigidly as children would not have tolerated this, but efforts were made to align the Frankfort plane approximately parallel with the floor and to have the child looking forward for image recording.

The reported findings demonstrate the advantage of stereophotogrammetry over standard photogrammetry. On a standard photographic image of a subject photographed with the head tilted backwards, measurement of the upper face is invalid. The upper face would appear foreshortened due to the camera angle and vertical upper face measurements on the image would be shorter than direct facial measurement. Using stereophotogrammetry, with backward head tilt of 30 degrees, or less, from natural head position, the true upper facial surface would be recorded and the upper facial dimensions would be accurate.

This availability of coloured photorealistic images produced by vision-based surface imaging systems, was utilised, in the study reported here, when locating upper lip landmarks such as the most superior point on the white roll during maximal smile. The blanching of the tissue in the line of the cheiloplasty and alteration of the apparent outline of the white roll was shown to contribute to the asymmetry of the upper lip line in smiling in the cleft subjects.

The accuracy of computerised stereophotogrammetry in the present study, when applied to infant facial casts, was 0.79 mm (SD 0.57). The operator error, or inconsistency, of 0.23mm (SD 0.35) increased to a maximum of 0.5 mm when the landmark digitisation was performed on images of live paediatric subjects. Published error estimations for other methods of soft tissue surface imaging have ranged from 0.5mm, for the liquid crystal range finder and laser scanning, to 3.8mm for an early version of computerised stereophotogrammetry, using a large voxel size of 1.27mm^3 (Coombes et al 1991); (Prasad et al 2000);(Yamada et al. 2002b).

Other authors have published accuracy as a percentage of length (the variation in recorded length between two landmarks on repeated measurement) but this form of error estimation is difficult to compare directly with other systems (Burke & Beard 1967).

No published estimates of the stability of the resting expression in children have been located to compare with the stability assessment carried out in the present study. It was shown that the landmark configurations recorded in subjectively similar resting expressions vary by 1.04mm (SD 0.47). In the study reported here, the resting expression was taken to be stable, in common with other authors who did not actually measure this variability. Manual and automated landmark extraction will be discussed later in this section, in combination with a consideration of landmark extraction in dental study models.

The computerised stereophotogrammetry equipment was bulky and sensitive to rough handling of the frame holding the camera pods. The combination of bulk and sensitivity is a disadvantage of this imaging modality compared to fixed rigid imaging equipment such as laser scanning or portable equipment such as handheld video cameras. The data files generated were not physically bulky, like dental study models, but at 120 megabytes per image-set per child, 0.5 gigabytes of storage media were consumed for the archiving of raw and edited data, including back-up files, per child.

6.1.2. CT Scanning of Dental Casts

Spiral or 3D CT scanning was chosen to record the fine surface detail and homogenous appearance, of dental study models. Batch scanning allowed 98 paired sets of study models and 7 upper arch casts to be scanned in 210 minutes. This compares favourably with laser scanning of dental casts which has been reported to require 40 minutes per cast (Kuroda et al 1996). A more recent report of ultra high-speed laser imaging has described capture times of 0.6 seconds per scan with 4 scans of each cast required to capture data in undercut areas (Sohmura et al 2000).

In the present study, spiral CT scanning of dental casts was shown to have a precision, or reproducibility, of 0.38mm (SD 0.18). Precision is the closeness of repeated measurements to each other. The effects of repeated scanning and operator inconsistency in landmark identification were quantified separately. The combined effect of these two factors is the reported precision.

Accuracy is the closeness of a measurement, or estimate, to its true value. The result of direct caliper measurement was taken to be the true value. When indirect measurement of the intercanine width of the scanned casts on screen was compared with direct caliper measurement, indirect measurement was found to exceed direct measurement by 0.35 mm (SD 0.13). This could be stated as 1.4% of the intercanine distance. Directly located 3D landmark co-ordinates were not available for comparison as an alternative to interlandmark distances.

This accuracy was comparable to that reported for laser imaging which was 0.3mm (Hirogaki et al. 2001). The accuracy of CT scanning in this thesis was better than the error of 0.2 -1.0 mm reported for direct digitisation with a Reflex Microscope (Derijcke et al. 1994). The slowest imaging technique, laser scanning, had a reported accuracy of 0.05 mm, but this would have required 120 hours of image collection to scan all the study models collected in this study. (Kuroda et al 1996).

6.1.3. Landmark Digitisation

As landmark co-ordinates remain the common output of indirect anthropometric methods and the basis of advanced shape analysis, the type of landmarks and the method of landmark identification will now be discussed.

Anthropometric facial landmarks were utilised in the present study; the clearly defined anatomical basis for such landmarks is the strongest evidence available for the claimed homology from case to case. While the basis is anatomical, however, the landmarks themselves may not be anatomical. For example, pronasale is the most prominent anterior point on the nasal profile but it is not the anatomical tip of the nose in all subjects. For the cleft subjects in the study reported here, the anatomical tip of the nose, the dome of the crura, was often flattened and located to the cleft affected side of the pronasale point. Pronasale is an example of a type II landmark as the homology from case to case is supported by geometric evidence.

In other cases, the detailed anatomical description of the landmark did not aid the operator's digitisation of the landmark. The subalare point is the insertion of the nasal alar wing into the upper lip but the mean alar thickness in the control subjects in this study was 3.13 mm. The operator still had to choose an appropriate site, within the 3.13 mm wide alar insertion, to represent the subalare point. In this study, examination of the operator inconsistency in landmark digitisation, ranked according to landmark, showed which landmark definitions lacked precision. Clear tissue boundaries such as the acute skin fold at the inner canthus, endocanthion, (en), were identified with greater consistency than landmarks such as otobasion inferiorus (obi) which is the inferior insertion of the fleshy pinna of the ear into the skin of the face. The first named landmark (en) could be digitised with 0.35 mm deviation between the 3D location of repeatedly placed landmarks while the second named landmark (obi) had a greater deviation of 0.60mm within repeated digitisations. Imaging techniques which utilise placed skin markers report greater precision in landmark digitisation but larger errors of 2-3 mm in repeated placement of the skin markers (Ferrario et al 1996) (Trotman et al. 1998b).

There were no type III, constructed or pseudo landmarks utilised in the study reported in this thesis.

Automated forms of landmark identification are objective but can identify only type II or type III landmarks, as digitisation of type I landmarks requires the operator's clinical judgement. Automation can take several forms as shown in a recent study of normal paediatric facial morphology (Yamada et al 2002a). Facial landmarks were extracted based on the use of the following; maximum distance anterior to a constructed frontal plane; maximum degrees of convexity or concavity in manually identified 3D curvatures; discriminant analysis of boundaries of colour change such as around the vermillion border. Techniques which harness clinical judgement to guide semi-automated landmark extraction render the process less tedious and time consuming; they retain the extraction of type I landmarks but do not significantly reduce operator error (Frantz et al. 1999). A promising application for automated landmark extraction is the combination of automatic identification with tracking of facial movement (Wachtman et al 2001).

It has not yet been established that anthropometric landmarks identified in resting and animated images are co-incident, although researchers have been forced to make this assumption to render statistical analysis of facial animation feasible. Identification of landmarks on the skin, without the need for added markers, and tracking of the same landmarks in function would give a robust account of vectors of movement.

The landmarks located on the CT scanned dental study models or directly with caliper tips in the present study were also anatomical, type I, landmarks. The relative ease of landmark identification on the detailed occlusal surface of the primary tooth was reflected in the lower level of operator error when digitising dental casts. The mean operator error associated with the former was 0.24 mm (SD 0.10). Anthropometric landmark digitisation on images of the living subject had a mean operator error of up to 0.5mm, depending on the landmarks being digitised.

The higher proportion of clearly defined landmarks over small areas in the dental arch has led to the availability of numerous schemata of dental landmarks and interlandmark distances, unlike in facial anthropometry. Landmarks defined by Farkas et al (1992) have been adopted widely in the latter field aiding inter-study comparisons. It must be concluded that the relative paucity of reliable facial anthropometric landmarks has forced the development of consensus and the universal adoption of Farkas-defined anthropometric landmarks.

The various definitions of arch dimensions were examined in the earlier review of the literature. Dental landmarks will now be considered. The inanimate nature of dental study models has allowed direct digitisation of landmarks and 3D co-ordinate extraction to develop to a level not replicated in facial anthropometry. In the latter area, direct facial digitisation featured briefly in the literature before surface scanning and other techniques of facial surface data collection were developed. The availability of high resolution imaging and automated landmark extraction may diminish the importance of direct digitisation and lead to consensus on landmark data sets, as the number of landmarks that can be reliably identified automatically is smaller than the range of landmarks suitable for direct measurement.

The varying suitability of certain landmarks to indirect anthropometry was demonstrated in this study. Direct measurement was performed between landmarks suggested by Moorrees (1969) with one modification. The first and second molar arch widths were measured between the mesial fossae on both the maxillary and mandibular arches. Moorrees' scheme of landmarks assumed normal posterior occlusion and used the tips of the disto-buccal cusps of the mandibular molars to define the posterior mandibular arch widths. Cusp tips were more clearly visualised than fossae on the images of CT scanned dental arches so, in this study, for indirect anthropometry, a set of cusp tip landmarks was used as an alternative to the Moorrees' scheme of landmarks. When photographed study casts have been studied, the cusp tips have also been found to be more readily identified (McAlarney & Chiu 1997).

6.2. Soft Tissue Facial Morphology at Rest

The results of facial anthropometry of the control sample at rest, comparison of cleft and control cases and the relationship between soft tissue facial morphology at rest and the facial skeleton will be considered now.

6.2.1. Shape Analysis in Control Cases

Normative data for soft tissue facial morphology in 3-year-old children are not available for many populations (Farkas 1994). In the control sample in the study reported here, the application of stereophotogrammetry produced values for linear and 3D facial dimensions that, at present, cannot be compared to the work of other researchers. There are no published reference values for this age group and ethnic group in this region of the world. Facial asymmetry is excluded from this statement and is discussed separately in section 6.5. The findings in the control sample are the basis for estimation of cleft-related soft tissue facial deformity. The effect of lip pose on data collected and the stability of the resting paediatric face also lack published counterparts.

6.2.1.1. The Effect of Lip Pose on Face Shape

The resting face with the lips lightly approximated is the documented basis for facial anthropometry. In older subjects, this can be achieved with ease. Data from subjects with lips both approximated and apart have been analysed together, dividing the sample into groups based on cleft type (Duffy et al 2000). If the lips were closed, a single landmark was digitised at the junction of the upper and lower lip in the midline. If the lips were apart, two landmarks representing the high and low points of the mucocutaneous border were digitised.

Comparison of linear dimensions, for lips open and lips closed poses, showed that the vertical lip dimensions were affected by alteration in lip pose. Horizontal lip dimensions were not affected. Three-dimensional analysis using

Procrustes superimposition confirmed that the lower area of the face was affected by alteration in lip pose. The sensitivity of methods of 3D analysis and the variability of the facial shape means that resting expression must be standardised to avoid the incorporation of additional variability. The results of the 3D analysis of laser scanned images performed by Duffy et al (2000) must be interpreted with caution, since this additional variability was not accounted for. Study methodology using imaging modalities that incorporate the results of successive images, such as the use of a liquid crystal range finder, should be modified to reflect the possibility of subtle changes in facial expression affecting the quality of the data collected (Yamada et al 2002a).

In the study reported here, the variability of linear dimensions and of face shape, within the group with lips apart at rest, was not quantified. In the control sample, 15 of the subjects had lips apart at rest. Subdivision of this group of 15 children into categories of lip separation would have produced sample numbers too small to permit meaningful statistical analysis. The findings in the face at rest with the lips apart, therefore, should be interpreted with caution.

6.2.1.2. The Stability of the Resting Face

The variability of subjectively similar resting facial expressions measured in this study quantifies the safety of the assumption that the resting face is stable. This is not an assumption that has previously been objectively tested, although it has been alluded to as a confounding variable in direct anthropometry (Farkas 1996). For practical purposes, this assumption will remain a constant feature of facial imaging and the researcher's efforts should be directed at excluding subjective differences when collecting successive data.

6.2.2. Cleft Related Soft Tissue Deformity

In the study reported here, the difference between soft tissue morphology in cleft and control cases has been termed deformity. It is considered within the context of the research work of others, even though much of this work does not refer to control data and is not analysed using 3D shape analytical methods, even when 3D data was collected.

Significant cleft-related stigmata were identified in the soft tissue morphology of Scottish UCL and UCLP samples at 3 years of age.

6.2.2.1. Upper Face

In the study described in this thesis, a wider intercanthal distance was detected in the UCLP cases than in the UCL and control cases, with the affected inner canthus displaced laterally. This co-incided with the finding of wider intercanthal width in UCLP cases than in controls on linear dimensional analysis of liquid crystal range data from Japanese 4- and 18-month-old subjects (Yamada et al. 2002b). Intercanthal width in Japanese UCLP subjects was within normal limits at 4 years of age. As this was a mixed cross-sectional study, it cannot be concluded that telecanthus had resolved at 4 years. Whether there was unilateral or bilateral displacement of the inner canthi in UCLP was not established by the authors. A study of older British subjects using laser scanning found reduced intercanthal width in both UCLP and BCLP subjects relative to control subjects (Duffy et al 2000).

There are two possible explanations for the discrepancy between the Japanese and British studies. Different samples could have produced different results. Alternatively, the growth rates in the ocular area in UCLP subjects are lower than in control subjects leading to normalisation of neonatal telecanthus. The results of cephalometric analysis of UCLP, BCLP and CPO cases at 3 months, 6 months and annually from 1 to 6 years would support the latter explanation (Ishiguro et al 1976). Nasal and maxillary widths were wider in the UCLP and BCLP cases at 1 year but grew more slowly than in the CPO cases. However, a more recent study found no difference between inner interorbital distances in UCL and UCLP cases at 22 months (Hermann et al 2000). Another, relatively recent, cephalometric analysis found that the wide upper face width in UCLP reduced after surgery but persisted at 8 years of age (Han et al 1995). Follow-up of the Scottish subjects in the present study would demonstrate whether the soft tissue intercanthal width would develop as the upper facial skeleton has been shown to develop.

6.2.2.2. Mid Face

This was the site of the most marked deformity in the Scottish cleft subjects. The soft tissue morphology in the UCL and UCLP groups was similar. The morphology of the cleft sample was different to the control sample. The nasal tip and nasolabial angles were both more obtuse, the soft tissue and anatomical nose widths were greater and the columella was wider in both UCL and UCLP than in controls. The nasal base widths differed significantly between the three subject types with the nasal base width greatest in UCLP cases, least in control cases and between both extremes in UCL cases. Nostril floor widths were enlarged in the cleft cases, on the affected side of the face, but in UCLP cases the nostril floor was also wider on the unaffected side of the face.

Some of these findings were also reported in comparable studies. In laser scanned images of older children the nasal base was wider in cleft cases, but wider in UCL than in UCLP subjects (Duffy et al 2000). Perhaps reflecting the wider age range examined; there was a larger co-efficient of variance than that recorded in the study in this thesis. As a result, some findings were not statistically significant, even though there were differences between subject types that mirrored the findings reported here. Soft tissue and anatomical tissue nasal widths were not examined in the laser-scanned images.

Wider nasal bases were reported at 4 and 18 months in UCLP cases, which were within normal limits at 4 years in a mixed cross-sectional study (Yamada et al 2002b). Increased nasal tip angle was also detected in the Japanese subjects and it was concluded the nasal tip was retropositioned. The distance between the base of the columella and the pronasale point was reduced, which was not found in the Scottish cleft subjects. The relative 3D position of the nasal tip was not identified, so there was no evidence offered in support of the claim of nasal tip displacement.

Increased nostril width on the affected side was present at 18 months and persisted at 4 years in the Japanese UCLP cases, which does not concur with the finding of wider nostrils on the unaffected side of the face in the Scottish UCLP cases. The racial differences in Oriental and Caucasian nose shapes mean that the absolute values for children of similar age cannot be compared between the Japanese and Scottish studies (Aung et al 2000).

It was postulated that reconstruction of the orbicularis oris may have normalised functional facial forces and accounted for the diminution of differences between UCLP and control cases at 4 years (Yamada et al 2002b). The soft tissue and anatomical nasal widths that are located away from the orbicularis oris muscle had not been measured. Evidence from other authors about the unchanging nature of nasal deformity, even following rhinoplasty, would suggest that, had these linear dimensions been measured they would have remained significantly different to normal at 4 years (Timoney et al 2001).

In the study reported here, the lateral displacement of the left alar insertion could also explain the observed shift of the nasal prominence away from the midline. The distance from the columellar base to the nasal prominence was within normal limits for the cleft cases, suggesting the prominence of the nasal septum was undiminished in these cases. Displacement of the left alar insertion could create unbalanced force vectors on the nasal tip, with the observed flattened left ala on a wider nostril floor opposing the more curved right ala. Therefore, there would be a tendency for the most prominent part of the nose to be shifted away from the affected side of the face with subsequent shortening of the columella on the left side, as was noted in this thesis.

6.2.2.3. Lower Face

In the Scottish samples, a broader than normal philtrum was seen in both UCL and UCLP cases. The upper left lip length was also greater in the UCL cases. The ratio between the philtrum and the upper lip (christa philtrae to chelion) is constructed at the time of surgery. Perhaps the constructed ratios should have been compared between the two cleft groups in the present study. Philtral width was not measured in the older laser scanned cleft and control subjects (Duffy et al 2000). Neither did Yamada et al (2002b) measure the overall philtral width in Japanese children. The hemiphiltral width on the unaffected side was wider than on the cleft affected side and had normalised by 4 years of age (Yamada et al 2002b). The upper lip in these cases had been reconstructed with a triangular flap rather than the rotation advancement flap, which is performed in Scotland. This may explain the difference in the surgical outcome of the upper lip. Yamada et al (2002b) again commented on 3D location of landmarks with no evidence offered to support the result. The vermillion protrusion was reported to be deficient in Japanese UCLP cases, whereas in the study reported here, the relative 3D location of the midpoint of the upper white roll did not differ between subject types at rest.

Larger mouth width in UCL than in UCLP and control cases was reported in this thesis. Duffy et al (2000) reported reduced mouth widths in the cleft cases relative to control cases. When pairwise comparison for cleft types was performed, no significant difference was detected between mouth widths in any pairing. Yamada et al (2002b) reported greater mouth width in UCLP cases at 18 months that had normalised by 4 years. Larger co-efficients of variance were apparent suggesting greater variability of mouth width measurements in the Japanese sample than in the Scottish samples.

6.2.2.4. Mid Face Position

Comparison of the location of the subnasale (sn) landmark in 3D space in the three subject types showed no significant differences between types in the relationship of the soft tissue overlying the anterior nasal spine to the nasal bridge and the submental fold. This demonstrates that, in the present study, the soft tissue mid face was not in a retruded position in cleft subjects at 3 years of age. The soft tissue face height was also normal in the cleft cases.

Dental arch analysis, however, detected reduction in the upper anteroposterior arch dimensions in UCLP cases. As the upper teeth are borne by the alveolar process of the maxilla, one reason for this anteroposterior arch deficiency could be a retruded nasomaxillary complex. Dental arch relationships have been considered synonymous with the skeletal midface previously (Friede et al. 1991); (Atack et al 1997). Radiographic analysis of the facial skeleton was not performed in the current study so nasomaxillary growth retardation, or otherwise, cannot be verified.

Animal models with created clefts have suggested that the maxillary base is more significantly influenced by palatal disruption than the dental arch (Voss & Freng 1983). The final dimension of the dental arch in domestic cats on a hypoplastic maxillary base seemed to be more responsive to compensatory factors.

6.2.3. Soft Tissue to Skeletal Relationship

The reported normal soft tissue midface position in Scottish cleft cases with soft tissue deformity limited to the area involved in primary surgery, at 3 years of age, is clinically significant. This could be an appropriate point, after scar maturation, at which to examine the results of the primary surgery, before possible skeletal growth abnormalities become manifest.

The variable relationship of the facial soft tissue with the underlying facial skeleton has been known for over 40 years, confirmed in 3D and explained more fully latterly (McCance et al 1997b;Subtelny & Rochester 1959). These findings would explain the discrepancy between soft tissue and possible skeletal midface developmental delay reported in this thesis.

The soft tissue to bone relationship in clefts can be considered with reference to the Functional Matrix Hypothesis of craniofacial growth and development (Moss 1997a); (Moss 1997c;Moss 1997b). This well accepted theory postulates that the developmental origin, growth and maintenance of cranial skeletal elements are responses to the demands of the related cephalic non-skeletal cells, tissues and operational volumes. It is proposed that changes in the capsular matrices in the head, such as the neurocranial capsule, or the periosteal matrices, such as muscles and tendons, are responsible for bone translation and local bone remodelling respectively.

According to this theory, the nasomaxillary complex, would grow partly in response to greater functional demands made on the nasal cavity, to expand and increase the diameter of the upper respiratory tract as the rest of the body grows. Another growth theory, affecting the nasomaxillary complex, is the notion of the nasal septal cartilage as determinant of craniofacial growth with the nasal septum acting as a pacemaker for maxillary growth (Proffit & Fields 2000). The normal skeletal growth seen in unoperated cleft cases demonstrates that palatoplasty disrupts normal nasomaxillary complex growth (McCance et al 1990). This would suggest that the effect of palatal scarring may negate the positive influence of the functional matrices and the nasal septal cartilage. The point at which the impaired skeletal development becomes manifest in the facial soft tissues is yet to be determined. At 3 years of age in the study reported in this thesis, the soft tissue midface position is within normal limits. It has been shown that at 5 years of age, differences between soft tissue facial profile as seen on lateral cephalograms can be detected between UCLP subjects treated in different European centres (Mackay et al 1994). This was not a case-control study so the degree of deformity relative to the normal population was not quantified.

6.3. Paediatric Facial Function

The discussion of facial function in this study can be considered under three broad headings: the acquisition of an image of a maximal expression, the interpretation of the data from control subjects and the relationship between the data from control and cleft subjects.

6.3.1. Maximal Smile Image Acquisition

Maximal smile was chosen as the maximal expression to be performed in this study for three reasons. It has been shown to be a reproducible expression in older subjects. It is an expression that young children could be instructed to perform with minimal explanation and it would test the apparent function of the repaired peri-oral muscle slings, and the orbicularis oris muscle in particular, by involving several of the action units (AU) described by Ekman & Friesen (1976). A maximal smile exercises risorius, to stretch the lip, depressor labialis, to part the lips, orbicularis oris, to raise the cheeks, zygomaticus major, to pull out the corner of the lip and relax orbicularis oris, and mentalis, to part the lips, levator labii superiorus and alaeque nasi to wrinkle the nose and buccinator to dimple the cheeks. This comprises 6 AUs in total.

A simple anecdotal classification of smile types, based on outline shape, attributed alterations in shape to dominant activity of one of the above AUs (Rubin 1974). In a study of 100 adults, 67% had what was described as a "Mona Lisa" smile due to active zygomaticus major. A "canine" smile with levator labii superiorus kinking the upper lip to reveal the canines was seen in 31% and 2% had a "full denture" or rectangular smile due to equal dominance of the lower lip muscles. The smiles produced by the children in this study were not classified into sub groups.

The pilot study showed that multiple (up to five) subjectively similar resting and smiling images could be acquired in most 3-year-old children without coaching. Furthermore, these expressions could be captured using computerised stereophotogrammetry in the short length of time the children would maintain the maximal expressions. The pilot study had demonstrated that the resting expressions were reproducible. There was an average within-landmark standard deviation of 0.77 over 1 or 30 minute delays, which was equivalent to an average distance of 1.09 mm. Using multiple smiling images recorded from a subset of children in the study group, the reproducibility of a maximal smile was shown to be similar, with 1.03 mm being the average displacement of landmarks on repetition of maximal smile. Following an hour-long data collection process in each adult subject, a reproducibility of "within 1mm" for maximal smile was reported by Frey et al (1994). Coaching prior to image recording was used also with adult subjects by Trotman et al (1997) and Peck et al (1992) but the level of reproducibility was 1.5mm in the former group's work and was not stated explicitly by the latter pair of researchers.

6.3.2. Analysis of Control Data

The results of analysis of the data from the Scottish control group agreed with the work of other researchers in facial function in adults. Commissure excursion in the horizontal and lateral directions was exceeded by displacement of the commissures in the anteroposterior direction. This finding was in keeping with Gross et al (1996), who showed that measurement of 2D amplitude was an inadequate assessment of facial motion. Three-dimensional displacement could be 33% greater than apparent 2D movement. This finding of movement in 3D would agree with, and extend the work of Paletz et al (1994). The need for reconstructive surgery to create lateral and vertical movement was discussed by Paletz et al (1994). In the study reported here, posterior displacement was seen to be a significant component of the displacement vector of the commissures. This displacement could not be detected by the 2D-measurement technique used by Paletz et al (1994). The inadequacy of measurement of lateral commissure excursion as a justification for the segmental gracilis muscle transplantation, in reconstructive surgery for young children with Mobius syndrome, has been confirmed by the findings in similarly aged Scottish children (Zuker et al 2000).

It has been claimed that the tragii and mid dorsum of the nose are static points on the face during maximal expression (Frey et al 1994). The notion of static points in facial function must be questioned by the finding that even landmarks closely anchored to bone, such as the inner canthi, moved during smiling in this study. This lack of stasis was seen in both distance analysis and 3D analysis. The lowest percent displacements (less than 0.8%) were seen in the width of the columella and the periocular distances involving the endocanthion landmarks. Analysis of movement vectors of the endocanthion landmarks showed up to 1mm displacement of individual landmarks between rest and smiling.

Rather than absolute stability, relative stability of landmarks is a valid construct and has also been adopted in facial movement tracking by Trotman et al (1998b). Trotman et al (1998a)have also employed the same method of superimposition used in the present study, Procrustes analysis, to analyse the perioral vectors of displacement. Rather than using markers attached to a headband or the teeth as anchor points for superimposition, as this group had done previously, anthropometric landmarks were utilised (Trotman et al. 1998c). Although 3D analysis of facial surface change in function was conducted, the results reported by these authors have been limited to distance analysis and asymmetry scoring. The question of homology of landmarks in function was not addressed by these authors.

6.3.3. Cleft Related Soft Tissue Facial Deformity in Function

The comparison of changes in facial surface with smiling in the cleft and the control subjects, in this study, showed differences that other authors had not reported and did not demonstrate other differences that other researchers had detected. The synkinetic movement of the unaffected commissure towards the midline in function noted by Offerman et al (1964) was not found in this study as commissure excursion was equal bilaterally in cleft and control groups. Increased asymmetry was noted in function but it was related to the nostril base width in subjects in this study and not to the upper lip lengths noted by Offerman et al (1964). The greater upper cutaneous lip and total upper lip height shortening by 0.1 to 2.75 mm seen in the UCLP cases disagreed with Offerman's findings of no differences in vertical dimensions between clefts and controls. Shorter upper lip heights in CLP subjects than in controls were found by Susami et al (1993). The movement being examined was maximum mouth opening, not maximal smile. A weighted extension device was placed between the lips, as the aim had been to evaluate elasticity as well as shape following cheiloplasty, so the results are not directly comparable. There was also disagreement between the results of this study and the work of Trotman et al (1999) which found no difference between UCLP and controls in maximal smile.

The subjects in the studies of Offerman et al (1964), Susami et al (1993) and Trotman et al (1999) were all older than the 3-year-olds in this study. Some of the discrepancies may be explained, therefore, by the effect of growth although at 3 years the upper lip was noted to have achieved full maturity in females by Farkas et al (1992). The study reported by Offerman et al (1964) was based on cine camera images so the documented discrepancy between 2D and 3D interlandmark measurement may account for the disagreement between findings.

One counterpart for the additional high and low upper lip points introduced in this study has been found in a study of unilateral and bilateral facial paresis by Frey et al (1994). Points in addition to standard anthropometric landmarks were digitised on the upper lip in an effort to give a more explicit description of the deformation of the lip contour during smiling. Other than the use of placed skin markers, additional anatomical landmarks do not appear to have been utilised in studies of cleft deformity at any age. This study had shown that, although the vermillion lip height in function remained within normal limits, there was asymmetry of the upper lip shape and drooping at the mucocutaneous border on the affected side.

This finding of drooping of the mucocutaneous junction in the upper lip could perhaps be compared with the results of a study by Carvajal et al (1994). The upper lip EMG profiles of UCLP subjects, aged 11 years, were examined before and after treatment with an appliance intended to eliminate the restrictive effect of the orbicularis oris muscles on the sagittal maxillary and dentoalveolar positions. An improvement in the dentoalveolar position correlated with a reduction in muscle electrical activity. The finding of a restrictive upper lip in UCLP was not replicated in the study reported here, which found no differences in horizontal lip measurements or midline lip border displacements in function between cleft subjects and controls. A consensus on the effect of function on the soft tissue morphology in cleft subjects is lacking.

When the results of integrated electromyography (iEMG) and skin surface measurement during mimetic expression in normal adults were correlated, a logarithmic relationship between muscle electrical activity and skin motion was identified (Burres 1985). This author criticised surface techniques as inadequate for assessment of motor function. It could be postulated, however, that the findings of cleft-related lip drooping and the absence of cleft / control horizontal upper lip discrepancies would indicate less muscle electrical activity in the area of the cleft.

The effect of this laxity of the upper lip on the underlying hard tissue can be considered with reference to the Functional Matrix Hypothesis (Moss 1997a). A disproportionate number of the children with clefts had anterior crossbites in the area of the repaired lip. There was less evidence for the restrictive effect of upper lip scar tissue, with the upper lip position within normal limits at the anthropometric landmarks and remaining so in function. If the upper lip was not forcing the teeth backwards into a position of neutral force perhaps this dentoalveolar deformity could be explained, in part, by possible reduced muscle activity in the overlying soft tissue and altered muscular function.

Scarring in another site, on the palate, would also influence the displacement of anterior maxillary teeth. In older children, up to 3mm palatal displacement of anterior teeth was reported adjacent to unilateral palatal scaring while displacement of posterior teeth was not detected (Ishikawa et al 1999).

The limitations of end points of maximal mimetic expressions, as indicators of facial function, are acknowledged. Static images cannot describe a dynamic process. The challenge is to create a dimensionally accurate, 4D imaging method to reduce the reliance on displacement vectors. Four-dimensional data will present even greater analytical difficulties than 3D data.

6.4. Symmetry

The following will be discussed in this section: various methods of measuring asymmetry; results of studies of facial asymmetry and the connection between attractiveness and symmetry

6.4.1. Measurement of Asymmetry

As predicted in the review of the literature, the form of 3D-shape analysis for which the results were most easily illustrated was asymmetry scoring of the subjects. Boxplots of the asymmetry scores for the different subject types and different regions of the face allowed rapid subjective visualisation of differences for regions or subject types.

A drawback of the asymmetry analysis utilised was the need to report the results in terms of unit size, the size to which the facial landmarks configuration were scaled to allow comparison, rather than millimetres. The merits of analysis of symmetry independent of an arbitrary symmetry plane are discussed later in this section. These benefits outweigh the disadvantage of reporting in unit size and not millimetres.

Showing an example of the facial appearance of a subject from either end of the spectrum of asymmetry can attach some sense of clinical significance to the calculated values. Researchers employing this mode of symmetry analysis have yet to publish images representing degrees of asymmetry, which would add an intuitive sense of value to the reader's interpretation of results in numerical form (Maull et al 1999).

By utilising superimposition of mirror images on the source image, this study did not make any assumption about a midline plane of symmetry. Examination of the ranking of individual landmark asymmetry demonstrated that the landmarks that were by definition midline landmarks, nasion (n), subnasale (sn), labrale superiorus (ls), stomion (sto), labrale inferiorus (li) and sublabialis (sl) were not co-incident when superimposed on their mirror images. This implies that the facial midline does not lie on a single sagittal plane when running cranio-caudally. This finding was in agreement with the results of a study employing opto-electronic landmark location that concluded that the axis of symmetry was not the midline points (Ferrario et al. 1994). Yet the same author, 7 years later, published an account of asymmetry, related to gender and age, based on an arbitrary symmetry plane bisecting both endocanthions (Ferrario et al 2001).

The results presented here would suggest that reflection of images across a plane, constructed from midline landmarks or between paired landmarks, is an oversimplified approach to the assessment of asymmetry (Ferrario et al 2001); (Ras et al 1995). Studies measuring distances from assumed midline symmetry planes would be affected by the same erroneous assumption (Kyrkanides et al. 1995). This assumption depends on the acceptance of two conflicting concepts. The concept that a fixed midline entirely divorces the facial halves from each other disagrees with the concept that growth centres, which may be centrally located in the face, grow at different rates.

6.4.2. Results of Asymmetry Assessment

In the present study, assessment of asymmetry, using paired linear measurements, failed to detect differences between the right and left sides of the face in control subjects. This disagreed with the finding of 3mm, or 3% difference, as the upper limit of normal reported by Farkas & Cheung (1981) using the same analytical technique. That study was conducted in older children with larger linear measurements. Others would suggest the degree of asymmetry does not vary with time and that while the 3mm difference may have varied, the 3% difference would be retained (Skvarilova 1993). In 720 control subjects, aged 6-18 years, greater asymmetry of 4 - 5 mm, which was unchanged with age, was detected using direct anthropometry and comparison of paired measurements (Skvarilova 1993).

Comparison of 3D facial landmark configurations, in control subjects in this study, was found to be more sensitive and demonstrated evidence of asymmetry related to individual landmarks. This lack of a predictable relationship between measurements and landmarks had been noted previously (Ras et al 1995); (Shaner et al 2000).

Ras et al (1995) also reported asymmetry, concentrated in the immediate area of the cleft, which was also found in the Scottish sample. The asymmetry of the periphery of the nose was similar in the three subject types. The perioral asymmetry was significantly higher in the cleft subjects than in control subjects. The asymmetry scores for the whole face were also greater than normal in cleft cases with the cleft types resembling each other. The upper face and nasal prominence, which were closer to normal in the cleft cases, did not counteract the disproportionate effect of the nasal base and upper lip deformity on the whole face asymmetry.

The mean paired interlandmark distances were compared in this study. There may have been individual cases of asymmetry of interlandmark distances within the control sample with dominance of one side of the face. As facial asymmetry is fluctuating, rather than directional, other individuals could have masked these putative cases with dominance of the opposite side of the face. The concept of dominance of one side of the face is also tied to the somewhat simplistic notion that the facial halves can be divided along a straight midline symmetry plane.

Asymmetry in the study reported here was shown to be less marked in the upper face, even in controls, which is not in agreement with other studies. In direct anthropometry of normal children, it has been concluded that the greatest asymmetry was in the upper third of the face (Farkas & Cheung 1981). More authors have suggested that asymmetry increases in a caudal direction (Peck et al. 1991); (Skvarilova 1994). Skvarilova (1994) disagreed with a simple increase in asymmetry from upper face downwards. The greatest asymmetry was detected in the cranial vault, the least in the upper face and the asymmetry in the mandibular area was mid way between the other two sides. This was, however, a radiographic study rather than a study of soft tissue asymmetry.

6.4.3. Attractiveness and Symmetry

As this study did not consider the attractiveness of the cleft subjects when examining deviation from normative values, it was not possible to comment on the relationship between the degree of asymmetry measured and perceived attractiveness in these subjects. Asymmetric smiles have been shown to be less attractive in normal subjects (Grammer & Thornhill 1994). The cleft subjects in this study had greater than normal asymmetry, accentuated in function around the nostril floor and upper lip contour. However, attractiveness and facial deformity in cleft subjects have been demonstrated to be unrelated (Laitung et al. 1993). Therefore, the greater asymmetry measured in the cleft subjects at rest which can be considered features of facial deformity, cannot lead to the conclusion that the cleft subjects were less attractive.

6.5. Dental Arch Analysis

The findings of this study will be discussed as follows: dental arch relationship; linear dimensional arch analysis; three-dimensional analysis of arch form and the dental arch in relation to the soft tissue morphology in 3-year-old children.

The work on dental arch analysis presented here addresses a deficiency in the literature that exists between investigation of the neonatal maxillary dental arch before, and at the time of, reconstructive surgery and investigation of the maxillary arch at 5 years of age.

6.5.1. Dental Arch Relationships

The antero-posterior inter-arch relationship is the feature of the dental occlusion that is considered critical to determining the grading according to the 5-Year-Olds' Index and the Goslon yardstick (Mars et al 1987); (Atack et al 1997). Incisor classification alone does not correspond with the categories in the 5-Year-Olds' Index and the Goslon yardstick but the evidence of emerging antero-posterior growth discrepancy in this study's UCLP sample suggests that the features determining outcome are already manifest at 3 years of age. A Class III incisor relationship was found in 31.3% of the UCLP subjects. None of the control sample and 9.1% of the UCL sample exhibited this incisor relationship.

The significant differences detected between UCLP cases, and both UCL and control cases, for arch depth and circumference ratios is further evidence of abnormal maxillary arch development manifest at an early age. The largest incremental increases in arch length and widths have occurred by the age of 2 years, even though growth continues in the maxillary arch until 8 years and in the mandibular arch until 13 years (Bishara et al 1997); (Bishara et al 1998). Therefore, at 3 years, when children can be persuaded to co-operate with impression taking without difficulty, it can be postulated that significant change of the dental arch from its neonatal form may have occurred. The predictive power of the 5-Year-Olds' Index was shown to be poor, and especially poor on an individual basis (Atack et al 1997). Abnormalities of dental occlusion at 3 years are likely to have even lower predictive power.

If dental arch development is examined as an outcome of primary surgery, rather than as a predictor of eventual outcome, the results of arch analysis in this study could be regarded as evidence in support of earlier assessment of primary surgery outcome. This could be 2 years earlier than the currently agreed assessment stage at 5 years of age.

6.5.2. Linear Dimensional Analysis

The finding of normality of the mandibular arch in UCL and UCLP subjects in the present study agrees with the findings of other studies in older subjects (Derijcke et al 1994). The smaller anterior arch dimensions reported show that the deforming effect of surgery, as described by Derijcke et al (1994), is of early onset. Cheiloplasty is generally agreed to affect anterior arch width while palatoplasty has adverse effects of varying severity on the transverse and anteroposterior arch growth. The similarities reported here, between UCL and control maxillary arch dimensions partially agree with the findings of two other studies in older Japanese children (Ohishi et al 1992); (Honda et al 1995). These authors postulated that intrinsic factors in the different types of OFC led to a variable response to the same surgery. Honda et al (1995) reported shallower maxillary arch depth in UCLP than in UCL cases which was found also in the Scottish samples.

Differences in maxillary posterior arch widths and not in anterior arch widths were found between cleft types in the Japanese 4-year-olds which disagrees with the findings reported here (Honda et al 1995). Palatoplasty performed on Japanese children seems to exert a greater adverse effect on transverse palatal growth than in Scotland, where posterior widths were found to be within normal range but intercanine width was reduced in UCLP cases. However, in the Japanese children, push-back palatoplasty had been performed which is not carried out in Scotland.

In a study of Finnish 3 year olds, the three cleft types UCLP, BCLP and CPO were found by Nystrom and Ranta (1990) to have similar maxillary arch dimensions following palatoplasty, which did not replicate the findings of Ohishi et al (1992). There were no control subjects in the Finnish study, so deviation of the dental arch from normal was not quantified. The three cleft types investigated all had palatal clefting so the findings cannot be compared with the findings in the Scottish samples.

Nystrom and Ranta (1990) found that mandibular arch intercanine width and depth were significantly shorter than normal in UCLP cases. In the present study, both cleft groups had mandibular arch dimensions within normal range. When the Finnish CPO subjects were re-examined at 6 years of age, mandibular arch discrepancies were also detected between the CPO and control subjects (Nystrom & Ranta 1994). The larger number of cleft subjects (180) and the smaller number of control subjects (50) may explain some of the differing results between the Scottish and Finnish centres. Genotypic factors may also account for differing arch dimensions in phenotypically similar subjects.

Opitz and Kratzsch (1997) examined 44 UCLP subjects from birth to the time of palate surgery at 3 years of age. The transverse arch width grew in the UCLP subjects up to the time of surgery. After surgery, the wear of an orthopaedic plate was recommended to avoid maxillary arch collapse. Despite the antero-posterior maxillary arch deficiency noted in the Scottish UCLP sample, the posterior transverse arch width remained within normal limits, 2 years after palatoplasty.

In common with many other researchers, neither the Japanese nor the Finnish studies examined the relative contribution of the affected and unaffected anterior segments to the anterior arch width (Nystrom & Ranta 1990); (Ohishi et al 1992); (Honda et al 1995). The lengths of the cleft-affected and unaffected anterior quadrants were examined separately in the study reported here. This showed that although there were significant differences in both anterior quadrant lengths between UCLP and UCL / control subjects, the left (affected) anterior quadrant in UCLP subjects was 4mm shorter than in UCL and control subjects and just 1.5 mm shorter on the right (unaffected) side. This separate consideration of components of the arch explains some of the contribution of the right and left anterior quadrants to intercanine width differences but linear dimensions still remain crude descriptions of the cleft dental arch.

The results of linear dimensional analysis can be compared within studies more reliably than subjective categorisations of dental occlusion. Such categorisations include the 5-Year-Olds' index or the simple four-grade classification of alveolar arch segmental relationship. The latter has been used to follow evolution of the maxillary arch from birth to 4 years in response to cheiloplasty and palatoplasty (Mazaheri et al. 1993). The classification was composed of four combinations of contact / no contact and overlap / no overlap of the maxillary alveolar segments. Contact of the segments can be determined easily but overlap or otherwise is a more rater-dependent feature.

6.5.3. Three Dimensional Analysis of Arch Form

Three-dimensional analysis of dental arch form, while objective and an explicit description of shape, can be difficult to compare across studies if different mathematical models have been used to analyse the configurations of 3D landmarks collected.

There are few comparable studies of arch form to provide a context in which to discuss the findings reported here. Numerous studies, which collected 3D co-ordinate data using handheld digitisers, analysed the data using only linear dimensional analysis (Derijcke et al 1994); (Heidbuchel & Kuijpers-Jagtman 1997); (Prasad et al 2000); (Stellzig et al 1999).

One study that attempted to exploit the 3D data collected, capitalised on the capacity to create "interlandmarks" between anatomical landmarks and manufactured arch curvatures for the edentulous neonatal arches (Mishima et al 1996). The curvature of the palatal curved surface was approximated to a sphere and the radii and centres of approximated spheres utilised as parameters to evaluate the forms of the alveolar arches. This technique described the major and minor alveolar segments separately and therefore cannot be compared with the results of arch form analysis used in the study. Another study which created mean alveolar arches used the technique of thin plate spline curve generation to represent the arch form (Hermann et al 2001). Rather than an average landmark configuration with the relationship between landmark and average shape retained, as in Procrustes analysis, the curve described the shape of the arch as a single unit. Therefore, the findings of Hermann et al (2001) cannot be compared with the findings reported here, which described the displacement of individual teeth relative to the normal dental arch.

No study, written in, or translated into English has been located that describes cleft arch shape in comparable terms. One paper of note discusses Procrustes superimposition applied to dental arch form compared to linear analysis (McAlarney & Chiu 1997). It concluded that the graphic representation of Procrustes analysis provided an intuitive appreciation of how and where the casts differed and that important descriptions of subtle change may be missed if traditional analytical methods are used. This has been borne out in this study. When the plots of the averaged dental landmark configurations are examined along with the various perspectives of the dental study models, the detail of individual tooth movement could be described.

6.5.4. Soft Tissue Facial Morphology and the Dental Arch

Based on the findings of this study, the use of the single adjective, 'dentofacial,' applied to deformity in cleft subjects should be more correctly termed dental deformity and facial deformity. Abnormalities of dental and soft tissue facial deformity were found to be unrelated at the age of 3 years. The children with the greatest degree of soft tissue deformity were not necessarily the children with the greatest degree of maxillary dental arch dimensional deformity.

The length of the left anterior quadrant, the feature of the dental arch that detected the greatest difference between UCLP and control cases, had no correlation with the width of the nasal base as demonstrated by low correlation co-efficients and high p values. The nasal base width was the soft tissue linear dimension that differentiated between the three subject types and was a key feature of the soft tissue deformity noted in both UCL and UCLP cases.

6.6. Clinical Indices

The literature review described several indices that can be applied to the various clinical sequelae of orofacial clefting. Some indices such as the Goslon Yardstick and the V, L, S classification are specific to clefts (Mars et al 1987); (Assuncao 1992). Others such as the maximal static response assay or facial action code are of universal application to facial pathology (Ekman & Friesen 1976); (Johnson et al 1994). Such indices have not been applied to the data collected in this project but can be discussed with reference to the clinical findings in the paediatric Scottish population. The dental indices have been discussed in section 6.5.

Clinical indices will be discussed as follows; existing indices of soft tissue morphology; proposed indices of soft tissue facial morphology and indices of treatment outcome.

6.6.1. Existing Indices of Soft Tissue Morphology

The main drawback of existing indices of facial deformity is the reliance on standardised photography. The inaccuracy and lack of precision of plain photography is less problematic in subjective grading systems such as the V, L, S classification of secondary lip deformity, as the features are assessed according to the surgeon's opinion and not facial dimensions (Assuncao 1992). The scale applied to nasolabial appearance, by Asher McDade et al (1992) in the oft-quoted six European centre study of cleft outcome, is a similar combination of assessment of plain photography and informed clinical assessment.

However, the significance of the anthropometry-based system devised by Hurwitz (1999) is undermined by the inaccuracy of the measurement technique employed. As has been seen in this project, some of the differences in linear dimensions that constitute facial deformity can be small. The system proposed by Hurwitz (1999) weights certain features according to their deforming effect.

Multiplying these linear dimensions by clinically predicated factors increases the inaccuracy of the assessment model. Nostril outline is one feature that is considered clinically significant. The effect of alteration in camera angle on the tracing of this nostril outline is one clear example of the disadvantage of plain photography.

Applying the same clinically weighted grading system to the data collected in this study would be more robust than basing the method on plain photography.

6.6.2. Proposed Indices of Soft Tissue Facial Morphology

The method of grading facial asymmetry employed in this project has been discussed in section 6.4. The advantages of this approach to facial symmetry have been presented. The fact that the value of the asymmetry score depends on the unit size and cannot be quoted in millimetres is a disadvantage of this asymmetry grading.

One solution to this problem would be to institute a system such as that devised by Tobiasen (1994) and termed a facial impairment scale. This would be for illustrative purposes only and not intended for clinical application. Tobiasen (1994) prepared four sets of images of rated "cleft impairment" and tested the reliability and validity for use of these images, in studies of severity of facial impairment, on panels of non-clinicians. It would be necessary to test whether exposure to sets of images of documented asymmetry could convey perception of the degree of asymmetry.

6.6.3. Indices of Treatment Outcome

A holistic approach to treatment outcome in OFC should include assessment of form and function. Function would include speech, hearing, facial animation and social interaction.

This study demonstrated the lack of correlation between dental and soft tissue facial deformity based on linear dimensions at 3 years of age. The relationship between the anthropometric landmarks in 3D-shape space and the linear dimensions could not be tested as the data were in different forms and the features were different entities.

An adult with good psychological health, near-normal facial appearance and function, normal dental occlusion and normal hearing and speech who was born with OFC can be judged to have a "good" outcome. The difficulty of rendering that judgement objective and applicable in the clinical setting is immense.

The localised soft tissue deformity reported in the Scottish UCL and UCLP cases and the significant maxillary dental arch deformity demonstrated in the UCLP cases at 3 years of age would fall short of ideal treatment outcome at this age.

6.7. Clinical Implications

The results of clinical research must be discussed with reference to clinical management. The question of whether or not evidence has been produced to justify changes in management must be considered. In this study there are several aspects of clinical management that might be affected by the results reported and the method by which those results were generated.

The possible clinical implications of the findings reported here will be discussed according to the following aspects of clinical management: cleft management outcome assessment; outcome assessment data format; surgical technique; health planning and ideal treatment aims.

6.7.1. Cleft Management Outcome Assessment

The present, nationally agreed, assessments at age 5 and 10 years could be augmented by facial imaging and dental arch analysis at age 3 years. Data collection using computerised stereophotogrammetry and recording of occlusal surface impressions is well tolerated at this age, as reported here. In 72% to 93.4% of cases, depending on subject type, the eruption of the primary dentition was complete and the caries rate was low so the dental arch shape and size could be assessed relative to the control sample. The degree of deformity of the facial soft tissue at rest and in function could also be quantified relative to the control sample when skeletal deformity did not appear to be manifest on the facial surface. At 3 years of age, only primary reconstructive surgery has been performed so the study population is what one author has referred to as "clean" (Atack et al. 1998). Confounding variables are minimised

What remains to be established is whether or not the same 3D methods of assessment could detect differences between cleft subjects treated according to different protocols in other centres. The soft tissue profile assessment performed by MacKay et al (1994) and the 5-Year-Olds' Index described by

Atack et al (1997) have been shown to be sensitive enough to apply to multicentre trials.

The main possible benefit of early outcome assessment is early identification of adverse outcome that may require modification of surgical or pre-surgical management. A theoretical benefit of even earlier soft tissue facial imaging is the possibility of surgical simulation. The collection of a sufficiently large body of data from cleft subjects, before and after cheiloplasty, could permit the simulation of a likely postoperative image. It would be necessary to quantify the variability of the neonatal cleft face before the number of subjects, required to validate surgical simulation, could be determined. The well-documented psychological morbidity associated with the birth of a child with facial deformity would further complicate this area of research. The appearance of the child affects the relationship with the mother and the effect of surgical simulation on parental expectations might be unfavourable (Maris et al. 2000).

Conversely, prediction of immediate surgical outcome may help parents to deal with the dramatic change in appearance that occurs with surgery in the early months of life.

6.7.2. Outcome Assessment Data

A common feature of 3D imaging as employed in this project is the limited availability of equipment. Many papers which described the analysis of soft tissue facial morphology have employed experimental equipment available only within research institutions which precludes wider application (Ayoub et al 1996); (Coombes et al 1991); (Kawai et al 1990); (Stevens 1997). Another common feature of such imaging is the data output format, 3D co-ordinates. Validation of such techniques is usually based on direct anthropometry as the gold standard (Aung et al 1995); (Bulstrode et al 1986).

Just as dental study models and cephalometric analysis have been accepted for multicentre trials, perhaps 3D landmark co-ordinate configurations, regardless of the method of data collection, could become the basis for wider investigations. The complexity of the error assessment and the reduction in accuracy when using multiple imaging modalities might render the results invalid. However, comparison of imaging techniques with each other, rather than direct anthropometry, may provide an evidence base for analysis of 3D data from multiple centres.

6.7.3. Surgical Technique

The modified Millard rotation advancement flap cheiloplasty performed on the subjects in this study has been cited as the cause of reduced upper lip height (Assuncao 1992). Reduction in the cutaneous lip height was reported here but the overall lip height was within normal limits. Irregularity of the vermillion border with notching at the base of the scar and broad reconstructed philtra were also detected. Such features are relatively common and not strongly associated with any particular reconstructive surgical technique (Assuncao 1992). The clinical significance of the statistically significant facial deformity and the possible clinical effect of surgical modification have yet to be determined.

As Timoney et al (2001) concluded, when examining the effect of secondary rhinoplasty, the likely benefit of surgery may not justify the intervention. That might transpire to be the case if current surgical protocol in Scotland was modified. Recommendations for modification of surgical procedures should be based on randomised control trials and not on the results of a single study of facial deformity. Although the early outcome at 3 years would suggest that reflection on early surgical management may be appropriate.

An example of evidence-based modification of surgical protocol was seen in one Swedish centre (Friede et al 2000). Seeking to reduce the reported deleterious effects of "push back" palatoplasty, this operation was replaced with a procedure requiring less palatal bony denudation. Only in cases of velar clefting did the subjects benefit to a significant level, with resultant arch width increase. The basic tenet of *primum non nocere* had been respected but long-term follow up is yet to be reported.

6.7.4. Health Care Planning

Resource allocation for medical treatment is an issue that applies to all healthcare provision models, whether state-funded like the British National Health Service or privately funded through health insurance purchased by individuals. The lip deformity that became apparent with maximal smile in the cleft subjects in this study may require future revision of the primary cheiloplasty. Comprehensive objective outcome assessment of cleft treatment either allows budgets to be planned in advance or treatment priorities to be established based on the relative clinical significance of the deformity.

6.7.5. Ideal Treatment Aims

In cleft subjects, where there is tissue deficiency and skeletal disproportion at birth, aiming for dentofacial parameters to be within normal range on discharge may be an impossible ideal but should be maintained as an eventual goal (Subtelny 1966); (Ishiguro et al 1976). The current accepted practice of judging outcome relative to other cleft management centres may be reasonable (Johnson et al 2000b); (Shaw et al. 1992). This practice has evolved in response to a deficiency of normative data. Alternative methods of research and audit could be introduced if normative data were available.

Even though Farkas et al (1992) have published population norms based on as few as 30 subjects, the larger subject numbers examined by Ferarrio et al (1999) are a more robust basis for population norms. Portable, non-invasive imaging, such as the handheld digitiser employed by the latterly named research group, allows collection of large bodies of data from subjects in their normal environment. The handheld digitiser, which has the same disadvantages as direct anthropometry, is unsuitable for the infant subject (Farkas 1996).

The application of 3D imaging modalities to the unaffected subject, as in this study, using computerised stereophotogrammetry, provides an alternative source of reference for the researcher in the field of cleft related deformity. The same methodology, which poses no risk to the subject and costs less time and money than direct anthropometry, could also solve the problem of over reliance on ageing archives (Warren & Bishara 2001). It has been suggested that archives of soft tissue morphology remain valid for only two decades (Farkas 1996).

6.8. Conclusions

Conclusions drawn from the study in relation to each aim are given below:

First Aim: To apply computerised stereophotogrammetry and three-dimensional morphometric assessment to soft tissue facial morphology in 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

Conclusions:

- Computerised stereophotogrammetry is an imaging modality that is suitable for paediatric facial imaging. The accuracy of the technique is acceptable for this purpose.

- Geometric morphometrics is superior to traditional morphometrics in the analysis of three-dimensional landmark data. The former analytical model is more sensitive to differences in shape.

The first null hypothesis stated that: Computerised stereophotogrammetry and three-dimensional morphometric assessment cannot be applied to soft tissue facial morphology in 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

On the basis of the results, this null hypothesis is rejected

Second Aim: To measure the statistically significant differences in soft tissue facial morphology at rest between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

Conclusions:

- There are significant differences in the soft tissue and anatomical nasal widths and in the nasal bases widths between 3-year-old children with unilateral cleft lip or cleft lip and palate and unaffected peers.
- The soft tissue facial deformity in 3-year-old children with unilateral cleft lip or cleft lip and palate is closely related to the site of primary surgery.
- There is significant asymmetry in 3-year-old children with unilateral cleft lip or cleft lip and palate compared to unaffected peers.
- There is telecanthus in 3-year-old children with unilateral cleft lip and palate.
- There is no soft tissue midface retrusion in 3-year-old children with unilateral cleft lip or cleft lip and palate.

The second null hypothesis stated that: There are no statistically significant differences in soft tissue facial morphology at rest between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology.

On the basis of the results, this null hypothesis is rejected

Third Aim: To measure the statistically significant differences in soft tissue facial morphology in maximum smile between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

Conclusions:

- Reproducibility of maximal smile is acceptable in 3-year-old children
- The resting expression in 3-year-old children is acceptably reproducible but the soft tissue morphology differs significantly with mild changes in lip pose.
- In smiling there are additional *facial soft tissue* differences between the affected and unaffected sides of the face in 3-year-old children with unilateral cleft lip or cleft lip and palate at the nasal base and in the upper lip. There is no evidence of compensatory displacement or non-anatomical movement.

The third null hypothesis stated that: There are no statistically significant differences in soft tissue facial morphology in maximum smile between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

On the basis of the results, this null hypothesis is rejected

Fourth Aim: To measure the statistically significant differences in dental arch size and shape between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

Conclusions:

- There is significant deformity of the linear dimensions of maxillary arch in 3-year-old children with unilateral cleft lip and palate. The maxillary arch is short and shallow.
- There is no abnormality of size of the mandibular arch in children with cleft lip or cleft lip and palate at 3 years of age.
- There are significant differences in three-dimensional maxillary arch shape between children with unilateral cleft lip, children with unilateral cleft lip and palate and unaffected children at 3 years of age.
- The deformity of the facial soft tissue and the deformity of the maxillary arch dimensions in 3-year-old children with cleft lip and cleft lip and palate are not directly related.
- Spiral CT scanning of dental study models provides an accurate and detailed description of dental arch form.

The fourth null hypothesis stated that: There are no statistically significant differences in dental arch size and shape between 3-year-old children with repaired unilateral cleft lip or unilateral cleft lip and palate and children with no facial pathology

On the basis of the results, this null hypothesis is rejected

6.9. Recommendations for Further Studies

There were further research questions posed at each stage of this study that have not been considered within this study and require further research. Some recommendations could be carried out using the data already collected for this study with additional analysis. Other recommendations would require repeat examination of the subjects in the study and control samples, perhaps up to 15 years from now.

The development of imaging equipment robust enough to be portable would allow data collection to be carried out away from the hospital and would make larger scale studies feasible. The use of high resolution cameras that could record sufficient detail of the facial surface to render the speckled flash redundant would allow the capture time to be reduced to less than 10 milliseconds and simplify the co-ordination of image downloading from the cameras. Range data could be constructed from colour images.

Homology of anthropometric landmarks between end-expression images was assumed in this study but this remains to be proven. Fewer assumptions about the stability and homology of key landmarks would increase the power of geometric morphometric analysis.

The data from the facial surface between anthropometric landmarks should be analysed to allow less rudimentary average face shapes to be generated.

The perceived attractiveness of images of subjects, at rest and smiling, of known facial asymmetry should be determined and correlated with the documented asymmetry of the subjects' faces.

Asymmetry scores should be categorised into grades between the extreme values recorded in this study and the correlation between shape analysis and informed clinical evaluation examined.

The predictive power of assessment of facial and dental arch deformity at 3 years of age should be tested. This could be done by investigating the same subjects in early adulthood when the eventual outcome of extended multidisciplinary management is known.

The length of time for which facial soft tissue normative data remains valid should be determined.

The relationship between the facial form of cleft-affected children and that of parents and unaffected siblings should be established. Previous work in this area required exposure of unaffected individuals to radiation and this would be avoided by using a non-invasive imaging modality such as stereophotogrammetry. This study has examined the relationship between facial forms in ethnically and geographically similar subjects.

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Erratum

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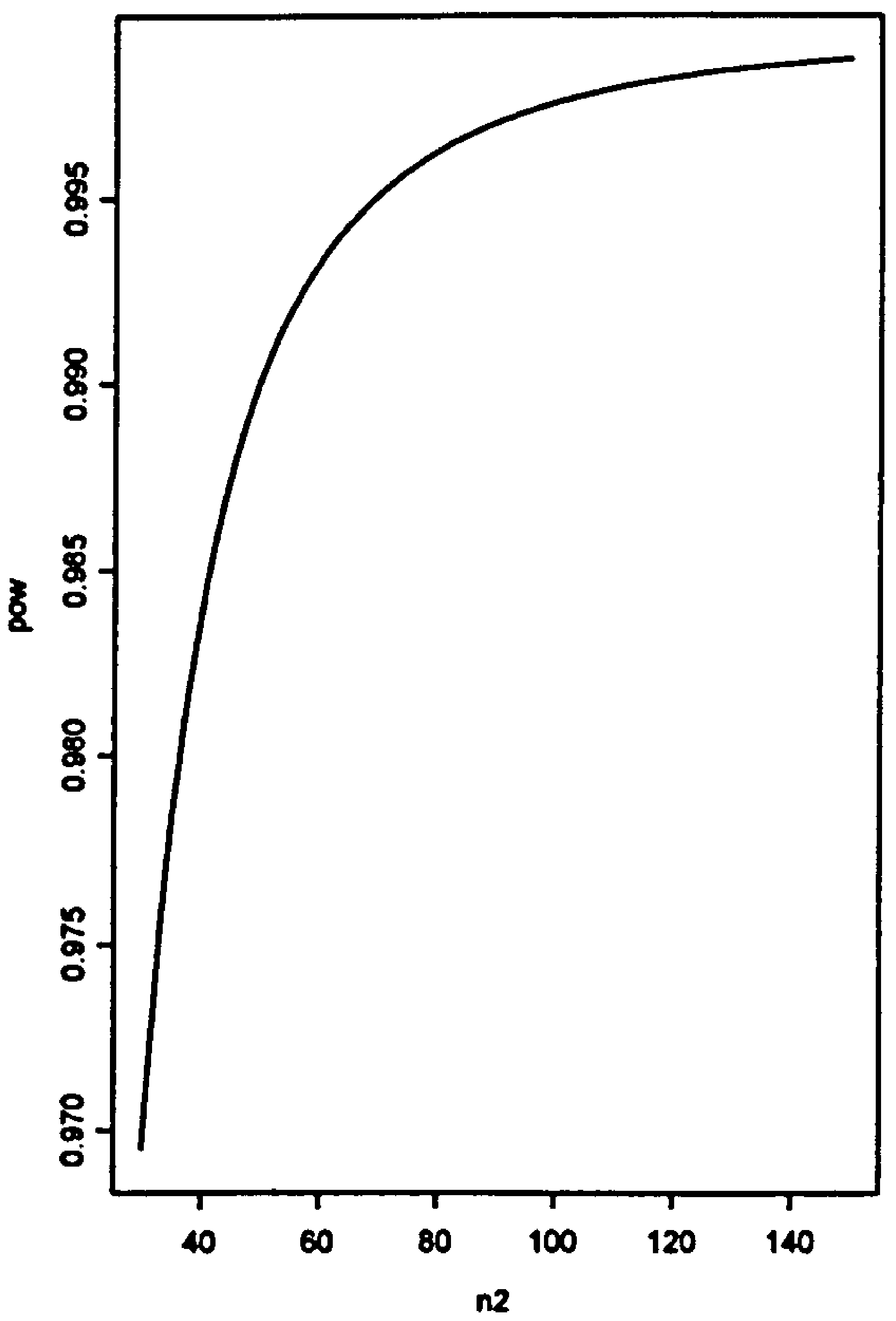
Appendices

Appendix 1 - Sample Size Calculation



It had been projected that 30 children with clefts would be recruited to the study. The above diagrams represent the rationale for a control sample of 90 unaffected children based on a study sample of 30. The diminishing return in additional statistical power with further increases in the control sample is presented

Appendix 2 - Package sent to parents - letter and form to be returned to researcher

Department of Oral Surgery,
Glasgow Dental School,
378 Sauchiehall St,
Glasgow,
G2 3JZ.

Phone number 0141 211 9699
a.garrahy@dental.gla.ac.uk

Dear Ms *Guardian's Surname,*

My name is Ann Garrahy, I am a researcher in Glasgow Dental Hospital. This letter was forwarded to you by *child's name*'s nursery. I am investigating a new method of examining children's faces that uses multiple cameras connected by a computer. The size of facial features can be measured on the picture produced on the computer screen. This camera system is being used to examine the faces of children born with cleft lip and palate. I am contacting children attending nursery schools in Glasgow and the surrounding areas to invite them to take part in the study. It is hoped this camera system will allow measurement of the face and will be able to measure the effects of early surgery in children with cleft lip and palate, without using x-rays.

I would like to photograph *Child's name* as part of the study to compare his measurements with children who have had surgery on their faces as babies. A set of photographs would be taken when *Child's name* is within six weeks of his third birthday. *Child's name* would also be asked to bite into soft putty to take an impression of his teeth. This would happen at the Dental Hospital and the visit should take no more than 45 minutes. Travel expenses will be refunded, if necessary.

I am asking you to consider making a visit to the Dental Hospital close to the time of *Child's name's* birthday. Your contribution to this research will add to the quality of care for children born with cleft lip and / or palate. My research is supported by the Cleft Lip and Palate Association with money from the national Lottery. If you have any questions I can be contacted at the above addresses and phone numbers. I would be grateful if you would return the enclosed form even if you are unable to take part in the study.

Yours sincerely
Dr. Ann Garrahy
Clinical Research Assistant

Cleft Lip and Palate Association / National Lottery Study

Please return this form in the enclosed stamped addressed envelope indicating whether or not you wish your child to take part.

Please tick yes or no

Yes I would like to enter *Child's name* in this research ....

No it will not be possible for *Child's name* to take part ....

If your answer is **Yes** please fill in the following details

Name of parent / guardian ....

Address of parent / guardian ....

....

Post Code of parent / guardian ....

Phone number of parent / guardian ....

Appendix 3 - Consent Form

3 Dimensional Assessment of Facial Shape

Consent Form

	Please initial below
1. I confirm the project has been explained to me and I have had the opportunity to ask questions	______________
2. I understand that my child's participation is voluntary and I am free to withdraw at any time, without giving a reason, without my child's future medical care or legal rights being affected	______________
3. I understand that participation in this study will offer no direct benefit to my child	______________

I, ______________________________ (name of person signing consent form) agree to ______________________________ (name of child) taking part in the above study

Signature ____________________________ Date _______________

Relationship to Child __________________________

Name of Researcher ______________________________

Signature of Researcher _______________________ Date __________

Appendix 4

Section 1

The three potential sources of error in landmark identification were quantified by comparing the co-ordinate configurations recorded using the C3D™ system combined with the Facial Analysis Tool.

Operator error - Discrepancies due to inconsistencies when locating landmarks by hand.

To quantify the amount of error associated with placing landmarks, comparisons were made amongst configurations taken from the same image by the same operator. The average displacement from the average of points around a single landmark (i.e. within-landmark standard deviation of points around their centroids) was used to measure the operator error. The results of these values averaged over the five landmarks are reported in Table A

Image	**Operator**			
	1	2	3	All
1	0.35 (0.31)	0.27 (0.12)	0.58 (0.84)	0.39 (0.50)
2	0.16 (0.14)	0.13 (0.07)	0.13 (0.06)	0.14 (0.09)
3	0.15 (0.03)	0.17 (0.08)	0.20 (0.14)	0.18 (0.09)
4	0.15 (0.05)	0.68 (1.08)	0.12 (0.06)	0.32 (0.64)
5	0.16 (0.06)	0.14 (0.04)	0.15 (0.02)	0.15 (0.04)
6	0.22 (0.13)	0.26 (0.16)	0.45 (0.61)	0.31 (0.36)
7	0.10 (0.06)	0.15 (0.06)	0.14 (0.06)	0.13 (0.05)
Mean	**0.19**	**0.26**	**0.25**	**0.23**
Std. Dev.	**0.15**	**0.42**	**0.40**	**0.35**

Table A: Average displacement (in mm) of repeatedly placed landmarks from their centroid

Imaging System Error - discrepancies due to errors in the underlying model generated by C3D.

To highlight any inherent instability in the imaging system, co-ordinate configurations from different images of the same cast in the same position were compared.

Over all landmarks, casts and operators the average within-landmark standard deviation of points around their centroids was 0.29 mm with a standard deviation of 0.37 mm (0.02 – 2.49mm). As these values were of the same order of magnitude as the operator error summarised in Table A, it was concluded that there is negligible inherent instability in the C3D system when multiple images of the same object are recorded.

Registration error - discrepancies due to differences in the placement of the 'imaged' object.

Images of each cast were recorded with the casts in four different positions relative to the cameras.

Over all 5 landmarks, 7 casts and 3 operators, the average within-landmark standard deviation of points around their centroids was 0.52 mm with a standard deviation of 0.40 mm (0.12 mm - 2.57 mm). The mean value was approximately twice the magnitude of the operator and system error presented in the previous two sections, although the maximum value was comparable. While the range of values was comparable, there was a positive shift in the size of the displacements of identified landmarks around their means. Since the position of the casts affected the confidence of the operators in placing landmarks it was not possible to conclude that the increased size of displacements was due solely to the registration process.

Accuracy

The co-ordinates returned from the C3D™ images were compared with the 'true' landmark co-ordinates available for each facial cast (obtained using an anthropometric devise of documented accuracy). To compare the two sets of measurements the co-ordinate systems of each were standardised, matching the configurations via translation and rotation using a Partial Procrustes Analysis. The response variable of interest was the deviation of the C3D co-ordinates from the true co-ordinates. This was quantified in each direction (X, Y and Z) and by a single (Euclidean) distance measure.

Figure A summarises the distributions of 315 values for each of the 4 displacement measures. The total of 1260 values comes from images of 21 casts each with 5 landmarks, photographed at 4 positions and digitised by 3 operators. The key features of the distributions are summarised in Table B. There was no evidence of a systematic deviation from the true x, y, and z co-ordinate values and the average displacement around the true values was less than 1mm for each. The overall (Euclidean) displacement on average was less than 1mm and the majority of located points were within 3mm of the true value. This degree of accuracy shows that these results, incorporating the three sources of error, are both reliable and accurate. The accuracy or overall system error of C3D™ was taken to be 0.79mm (Std. Dev. 0.57).

Figure A: Boxplots of the distribution of displacements of the landmark co-ordinates around the true values

	Displacements			
	x - direction	y - direction	z - direction	Euclidean
Mean	0.00	0.00	0.00	0.79
St. Dev.	0.60	0.55	0.55	0.57
Median	0.05	-0.05	-0.00	0.68
IQR	0.61	0.62	0.62	0.46
Min.	-4.53	-5.23	-3.53	0.07
Max.	4.87	4.10	2.87	6.21

Table B: Displacements (in mm) from the true co-ordinate values over all landmarks, casts and operators.

Part 2 Head Position

	-40	-30	-20	-10	0	10	20	30	40
					degrees				
Mean Position Error	1.13	0.36	0.28	0.28	NA	0.32	0.4	0.3	0.79
Mean Operator Error	0.44	0.25	0.15	0.18	0.24	0.22	0.24	0.2	0.4
2 SE Position Error	0.64	0.09	0.06	0.07	NA	0.07	0.38	0.08	0.45
2 SE Operator Error	0.23	0.08	0.09	0.09	0.08	0.08	0.06	0.14	0.17

Error (mm) associated with head tilt. Zero degrees was the position where the Frankfort Plane was parallel with the ground

Part 3

Child	Session 1 Average Displacement	Std. dev	Session 2 Average Displacement	Std. dev	Average Average Displacement	Std. dev
1	0.25	0.12	0.28	0.11	0.26	0.12
2	0.21	0.09	0.22	0.10	0.18	0.10
3	0.25	0.10	0.24	0.12	0.25	0.11
4	0.21	0.07	0.23	0.10	0.22	0.08
5	0.26	0.09	0.26	0.10	0.26	0.10
6	0.19	0.12	0.21	0.13	0.20	0.12
7	0.26	0.13	0.21	0.06	0.23	0.10
8	0.19	0.09	0.15	0.09	0.17	0.09
9	0.24	0.06	0.22	0.08	0.23	0.07
10	0.18	0.07	0.21	0.09	0.19	0.08
Average	0.22	0.10	0.22	0.10	0.22	0.10

The operator error is stated in millimetres.

Child	Session 1 Std Deviation	SE	Session 2 Std Deviation	SE	Average Std Deviation	SE
1	0.88	0.44	0.95	0.44	0.92	0.28
2	0.72	0.23	0.68	0.38	0.78	0.17
3	0.77	0.45	0.65	0.32	0.80	0.27
4	0.58	0.40	0.45	0.30	0.60	0.31
5	0.59	0.21	0.65	0.34	0.73	0.19
6	0.88	0.46	0.47	0.28	0.82	0.35
7	0.91	0.66	0.85	0.37	0.87	0.39
8	0.82	0.35	0.43	0.18	0.72	0.19
9	0.77	0.44	0.70	0.32	0.79	0.17
10	0.53	0.25	0.53	0.26	0.58	0.12
Average	0.78	0.44	0.64	0.36	0.78	0.29

The stability of the resting expression is stated in within landmark standard deviation.

Part 4 Smile reproducibility

Inter-Model Averages

Landmark	Clefts	Controls
chL	1.35	1.66
chR	0.76	0.98
cL	1.00	0.70
cR	0.93	0.82
enL	1.35	1.05
enR	1.48	1.07
li	1.19	1.00
ls	0.81	1.06
obiL	1.30	1.27
obiR	1.55	1.68
prn	1.12	0.81
sbalL	0.78	1.25
sbalR	0.88	1.05
sl	1.34	0.96
sn	0.71	0.76
Average	1.05	1.01
Overall	1.03	

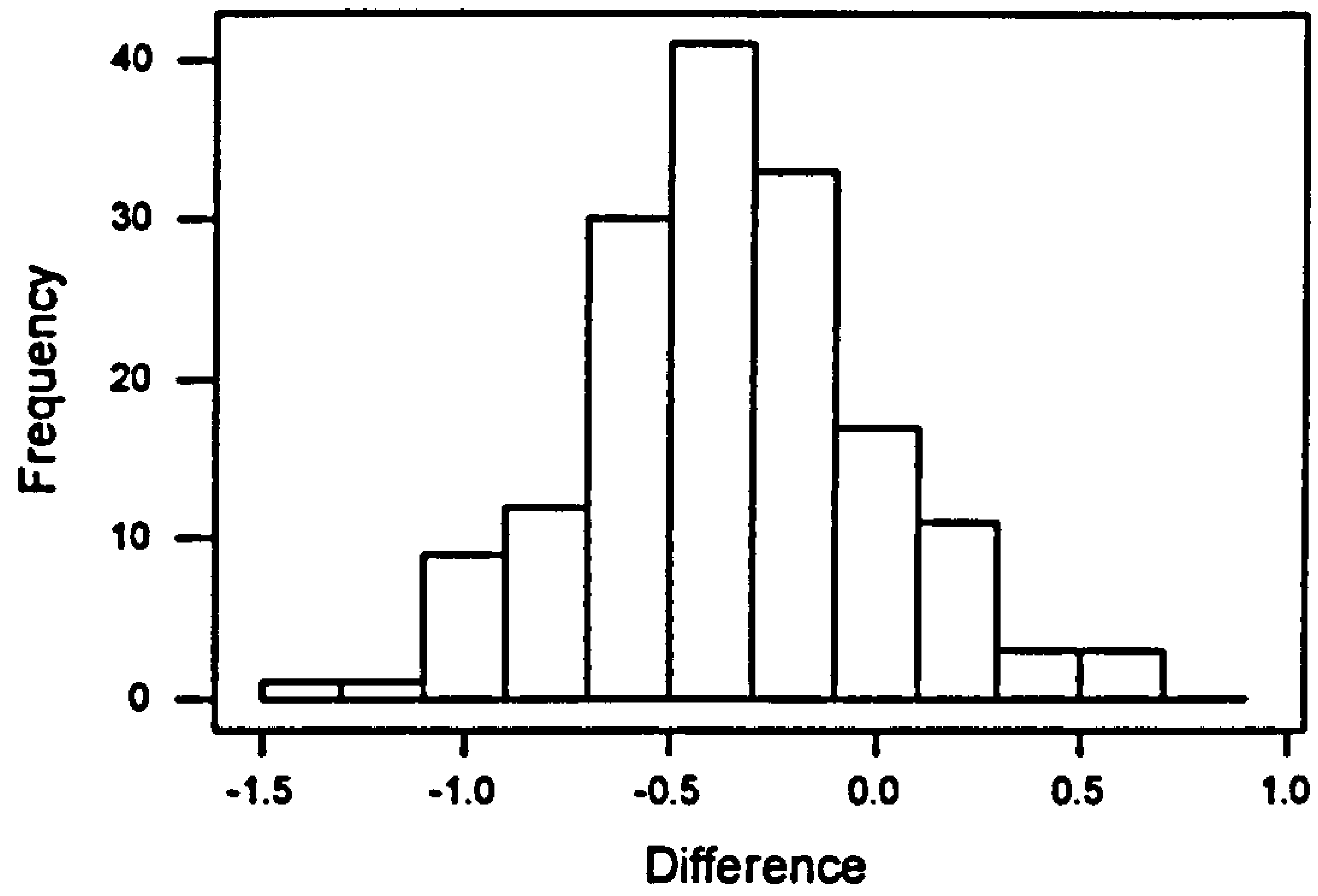
The reproducibility of maximal smile is stated in millimetres

Part 5 CT scanning accuracy and precision

Average displacement	Error type		
Landmark	Inter model	Intraoperator	Grand Total
AI	0.92	0.39	0.65
BI	0.65	0.12	0.37
CI	0.61	0.29	0.44
DI	0.8	0.26	0.52
EI	0.61	0.26	0.43
FI	0.56	0.34	0.44
GI	0.55	0.28	0.41
HI	0.49	0.33	0.4
JI	0.53	0.3	0.41
KI	0.51	0.25	0.38
LI	0.58	0.22	0.39
MI	0.52	0.24	0.38
NI	0.26	0.15	0.21
PI	0.19	0.23	0.21
QI	0.32	0.33	0.32
RI	0.28	0.26	0.27
SI	0.4	0.26	0.34
TI	0.71	0.25	0.46
Au	0.4	0.19	0.3
Bu	0.29	0.22	0.25
Cu	0.33	0.19	0.26
Du	0.37	0.25	0.31
Eu	0.24	0.17	0.21
Fu	0.28	0.24	0.26
Gu	0.28	0.22	0.25
Hu	0.3	0.16	0.23
Ju	0.31	0.19	0.25
Ku	0.39	0.2	0.29
Lu	0.39	0.06	0.22
Mu	0.33	0.15	0.24
Nu	0.66	0.2	0.42
Pu	0.71	0.41	0.55
Qu	0.56	0.29	0.42
Ru	0.83	0.28	0.54
Su	0.21	0.28	0.12
Tu	0.71	0.25	0.46
Total	0.53	0.24	0.38

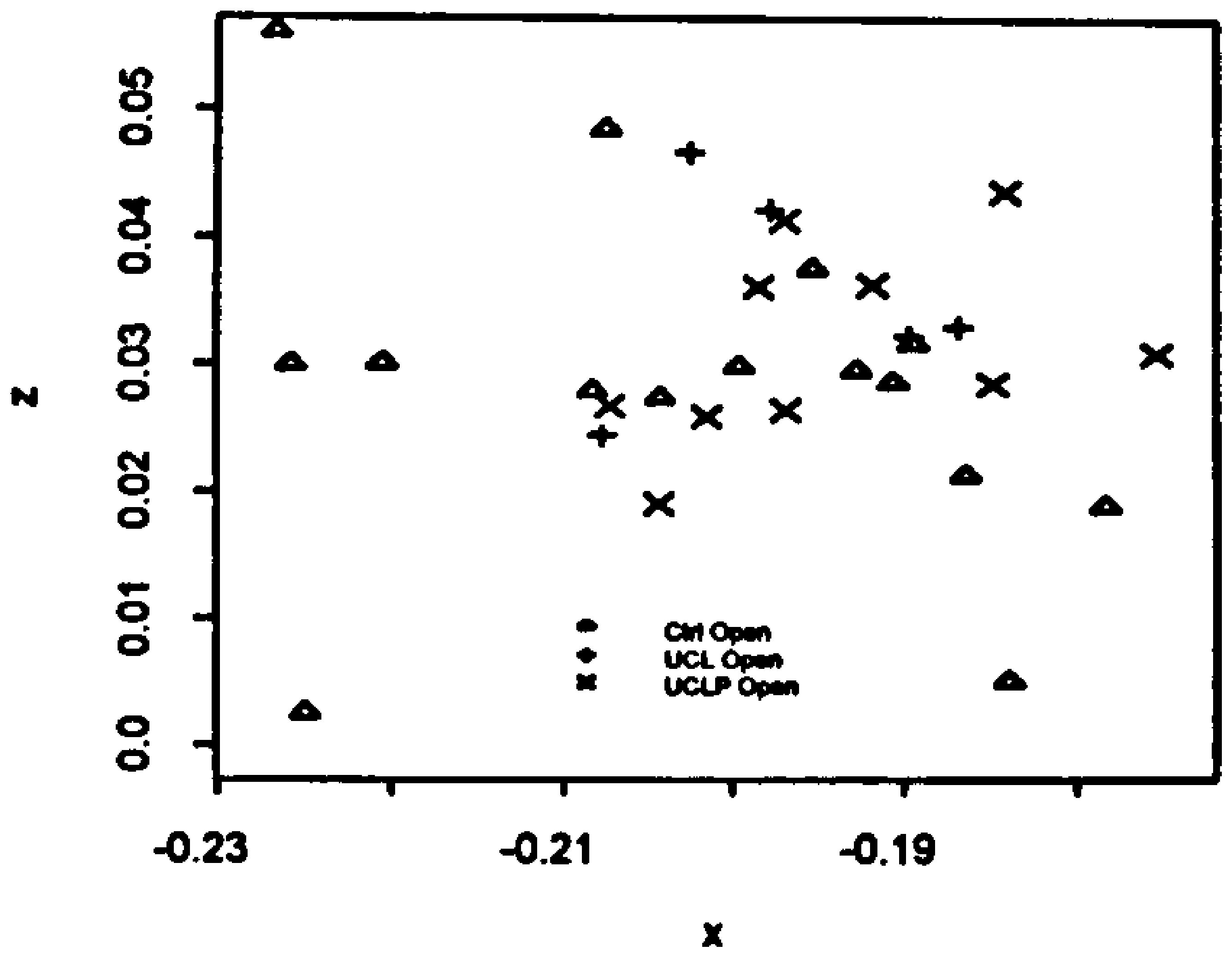
The precision of CT scanning is stated in millimetres

The accuracy of CT scanning is illustrated in this histogram with the intercanine width measured on the CT scanned image consistently larger than directly measured intercanine distance



Appendix 5 a - Relative Position of Right Inner Canthus presented overleaf

Axes as illustrated in Figure 3.24.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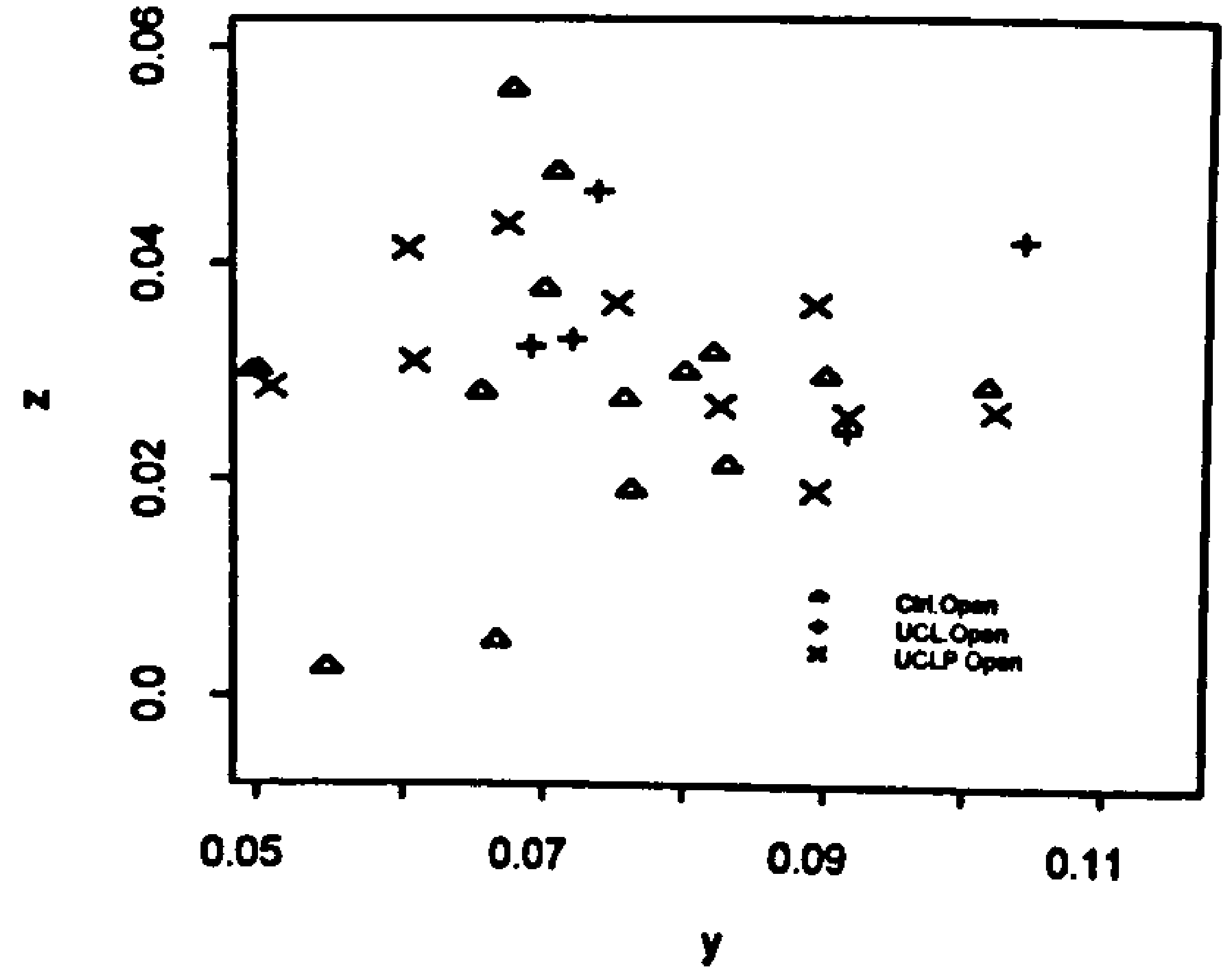
UCLP Open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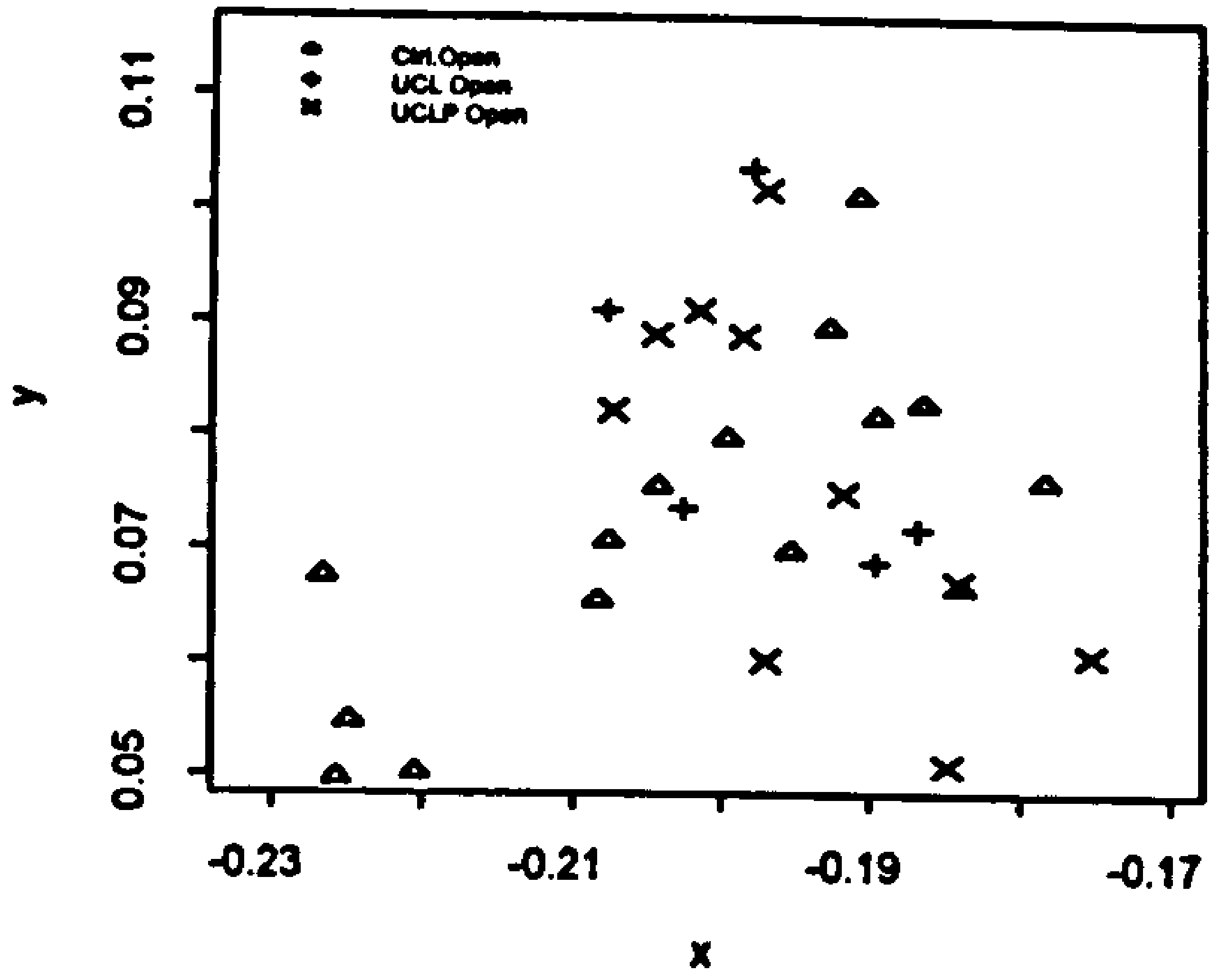
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x
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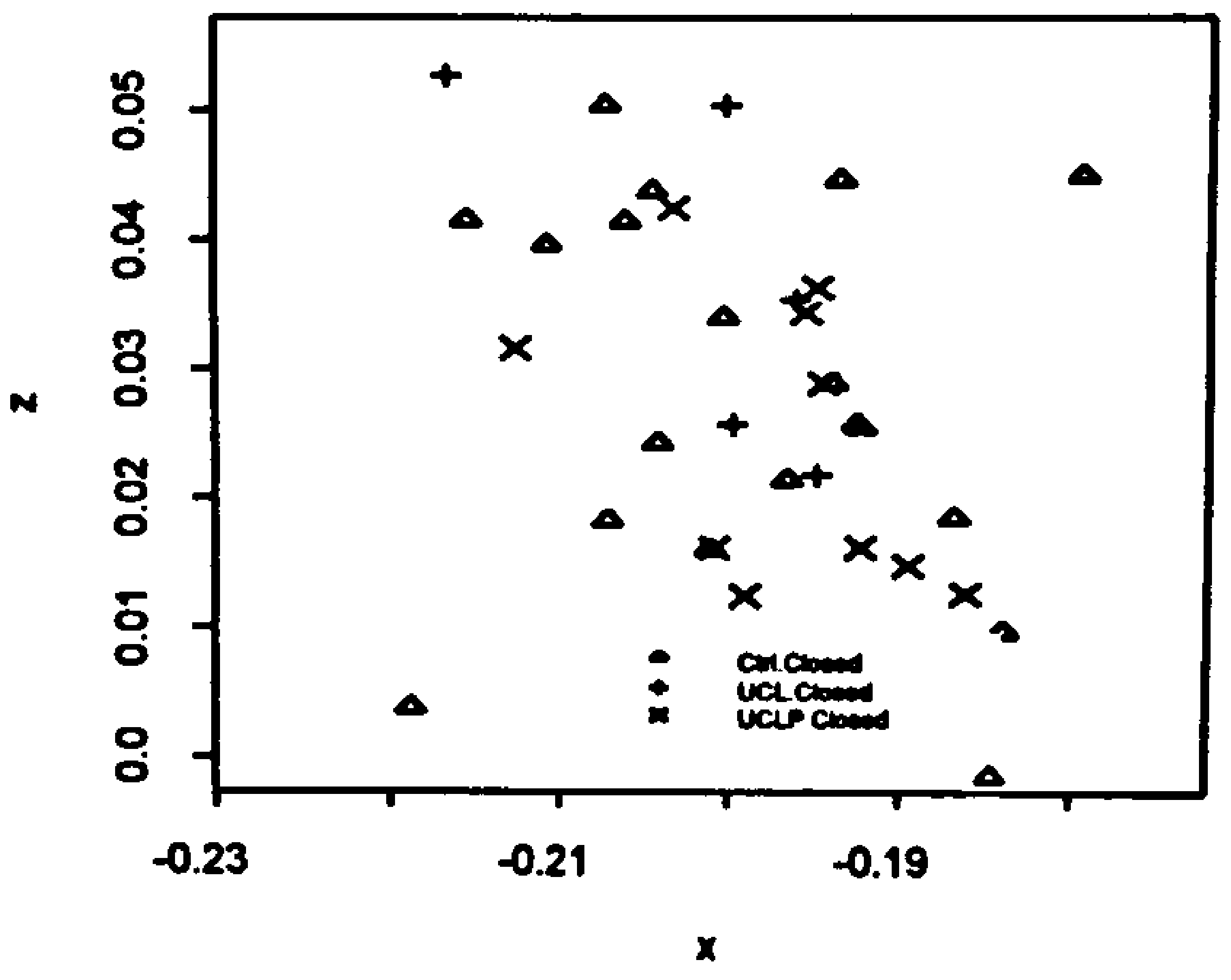
UCLP Open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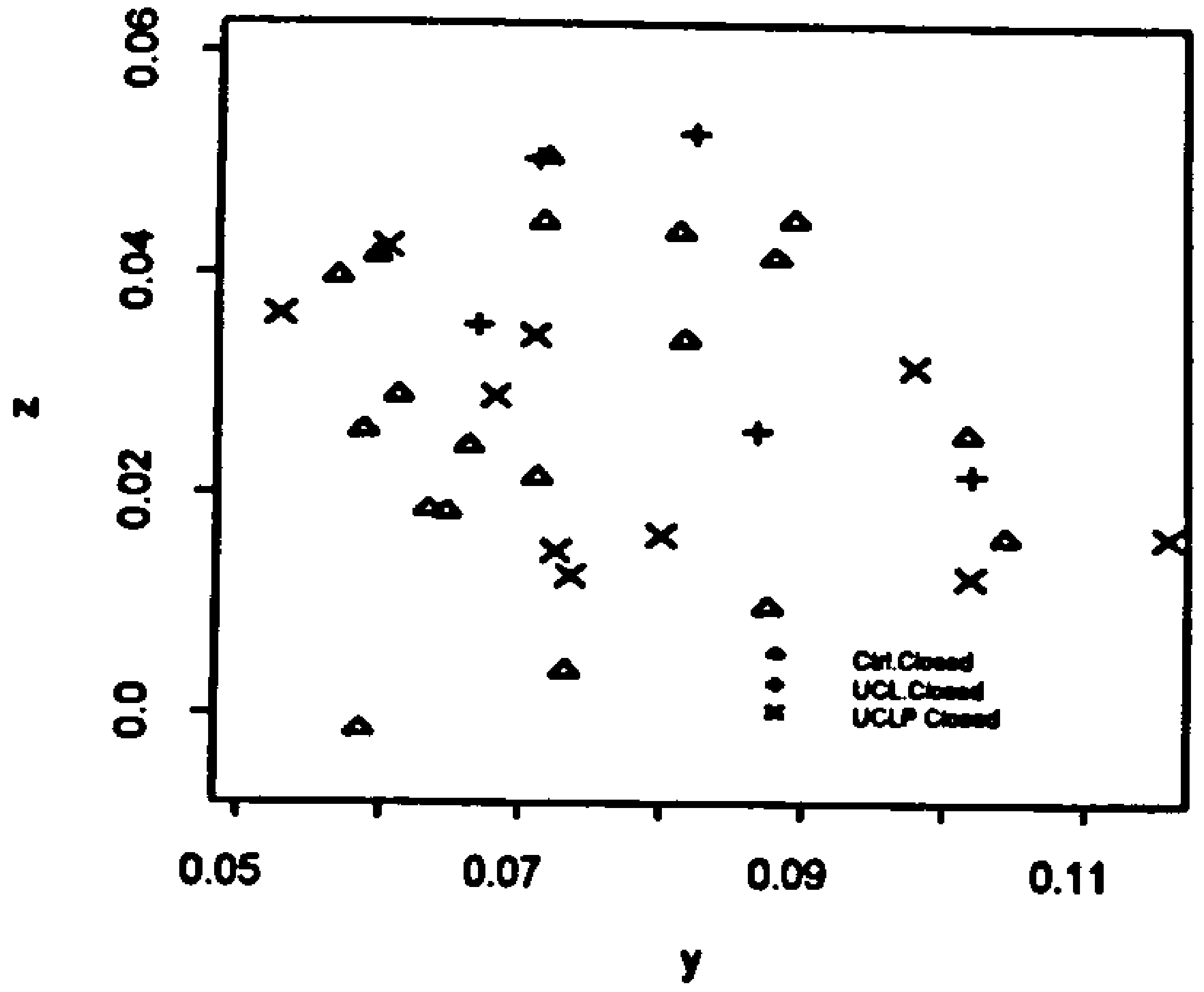
UCLP Closed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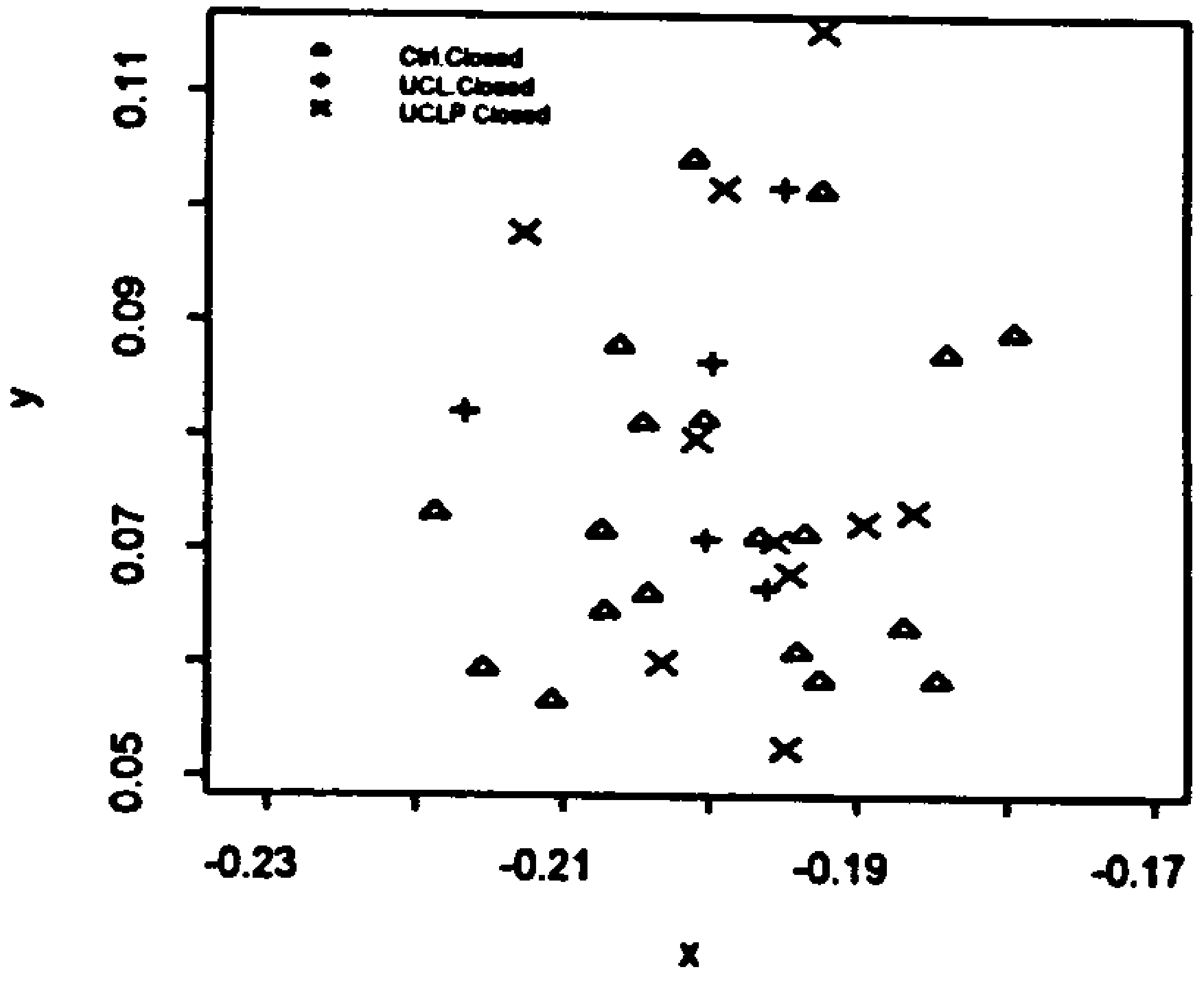
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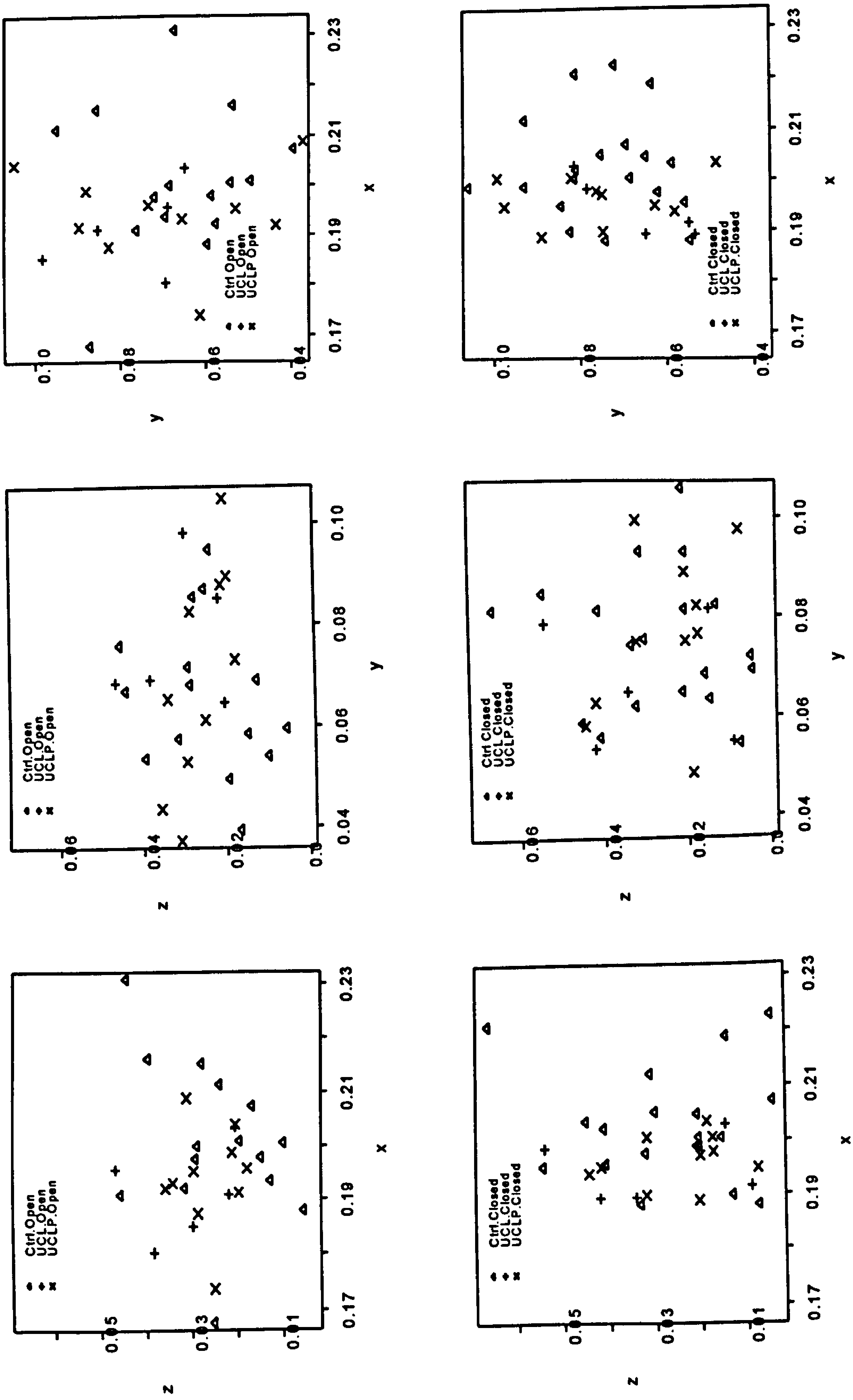
Appendix 5 b - Bookstein Plots of Relative Position of Left Inner Canthus presented overleaf

Axes as illustrated in Figure 3.24.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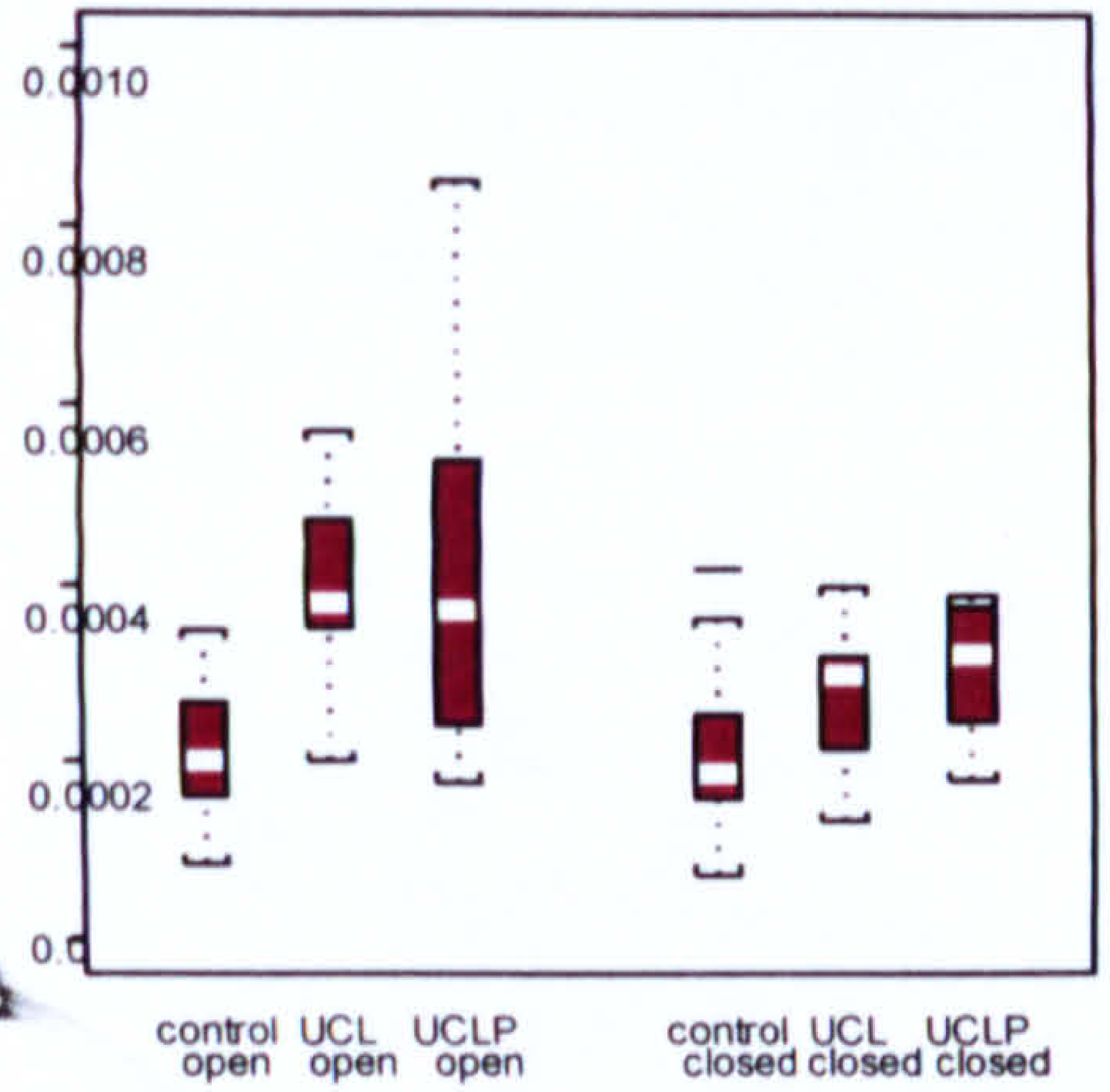
0.06

Appendix 6 - Boxplots of Asymmetry Scores for UCL, UCLP and Control Cases

Image overleaf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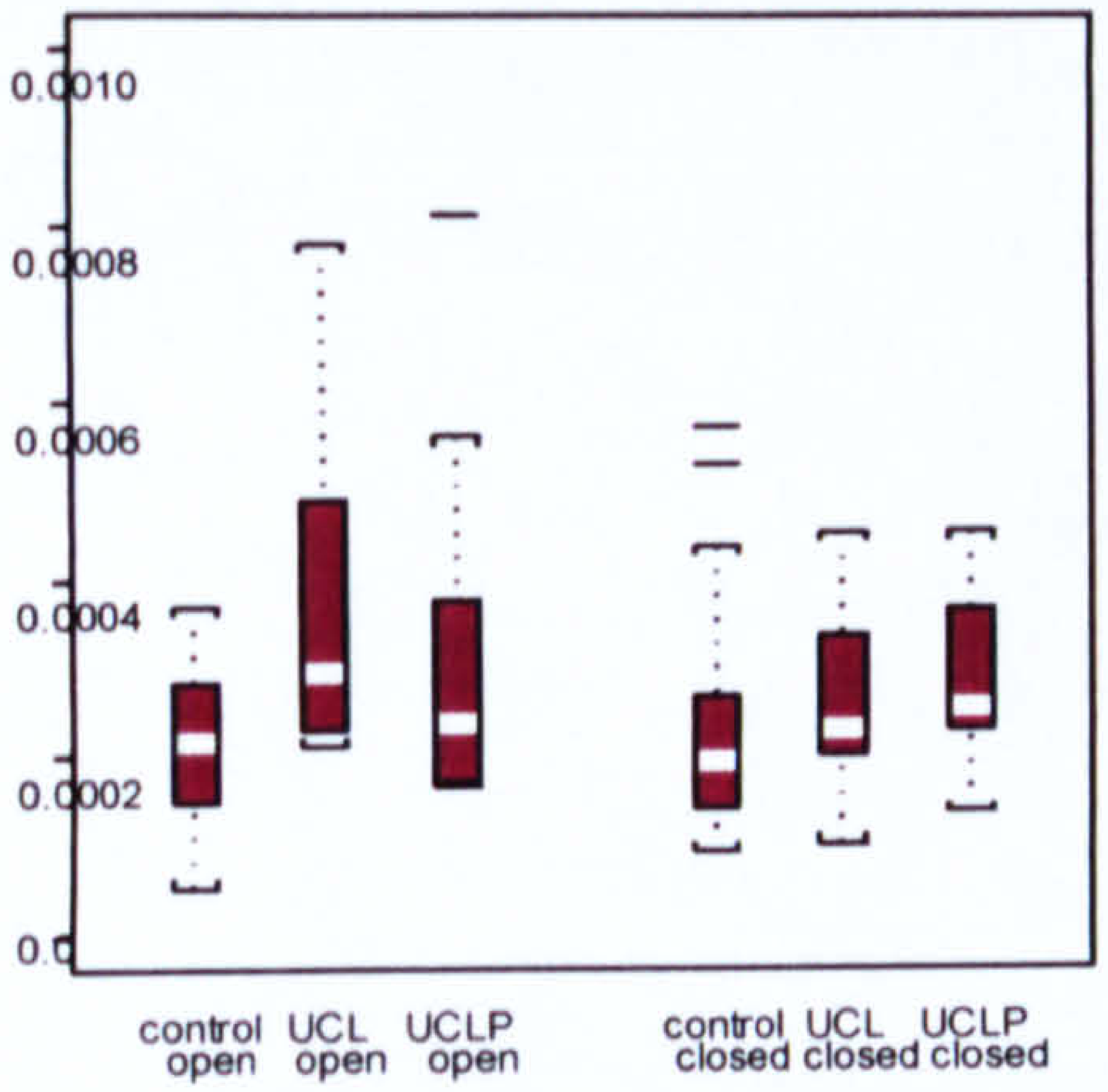
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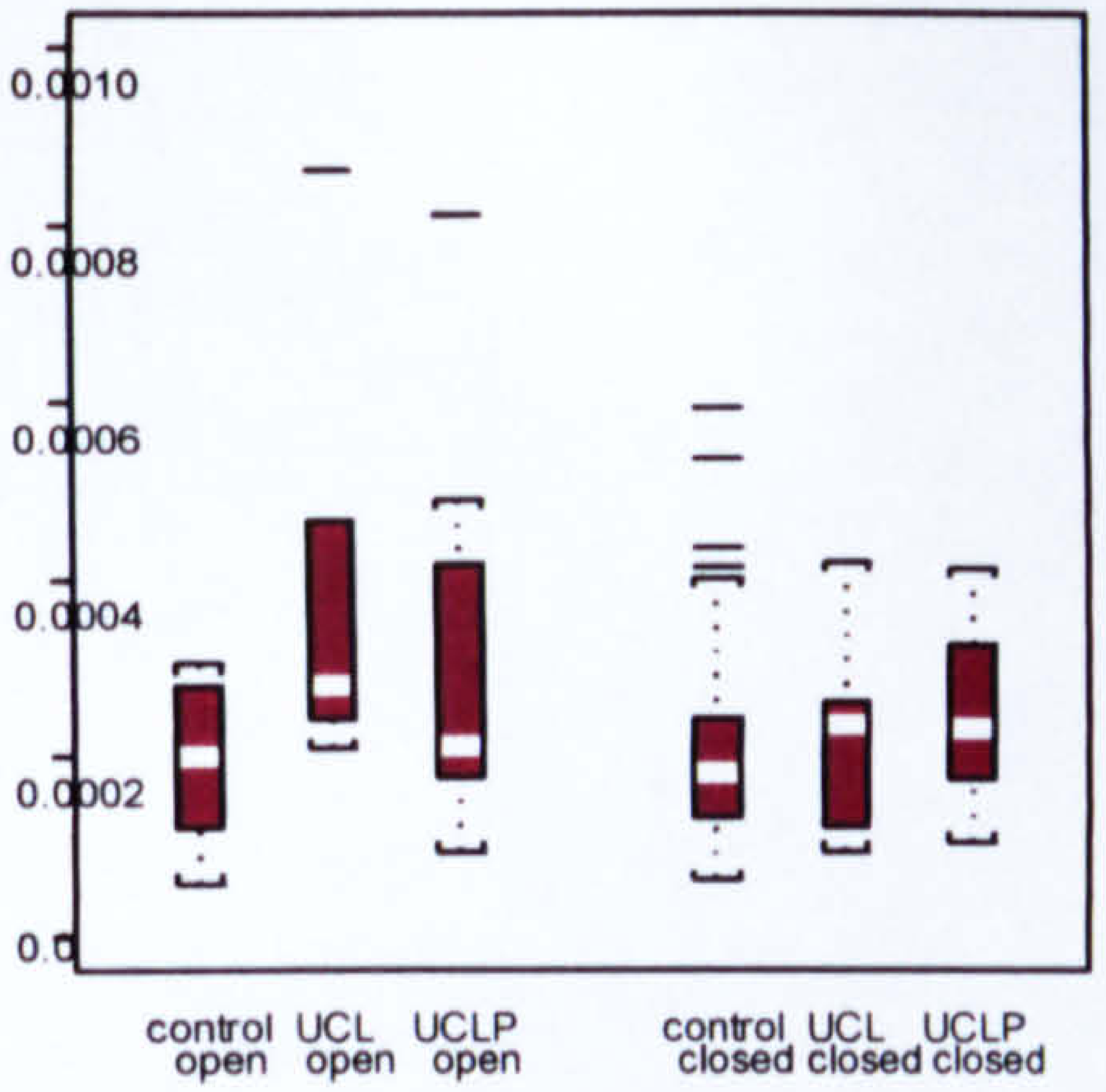
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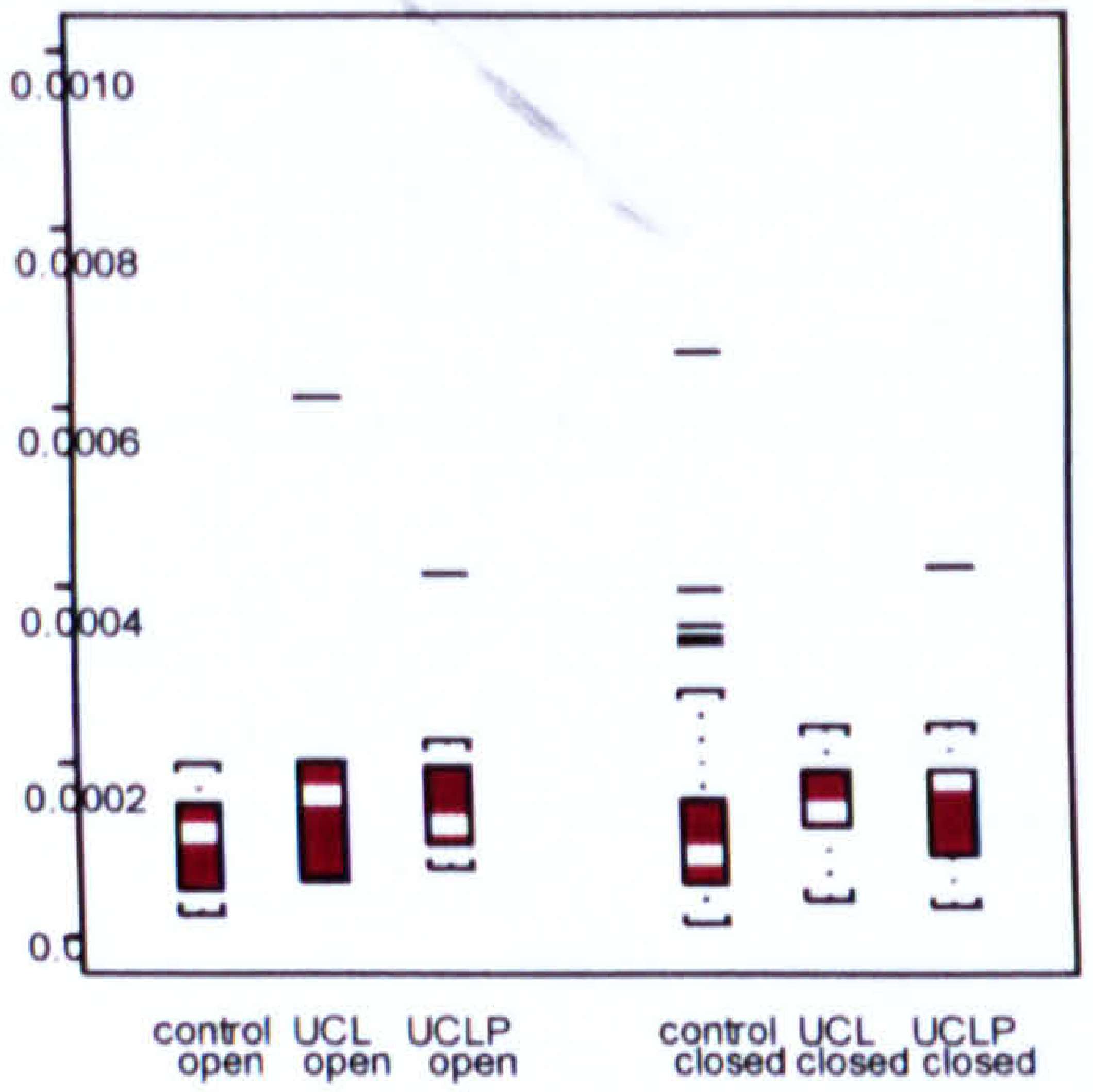
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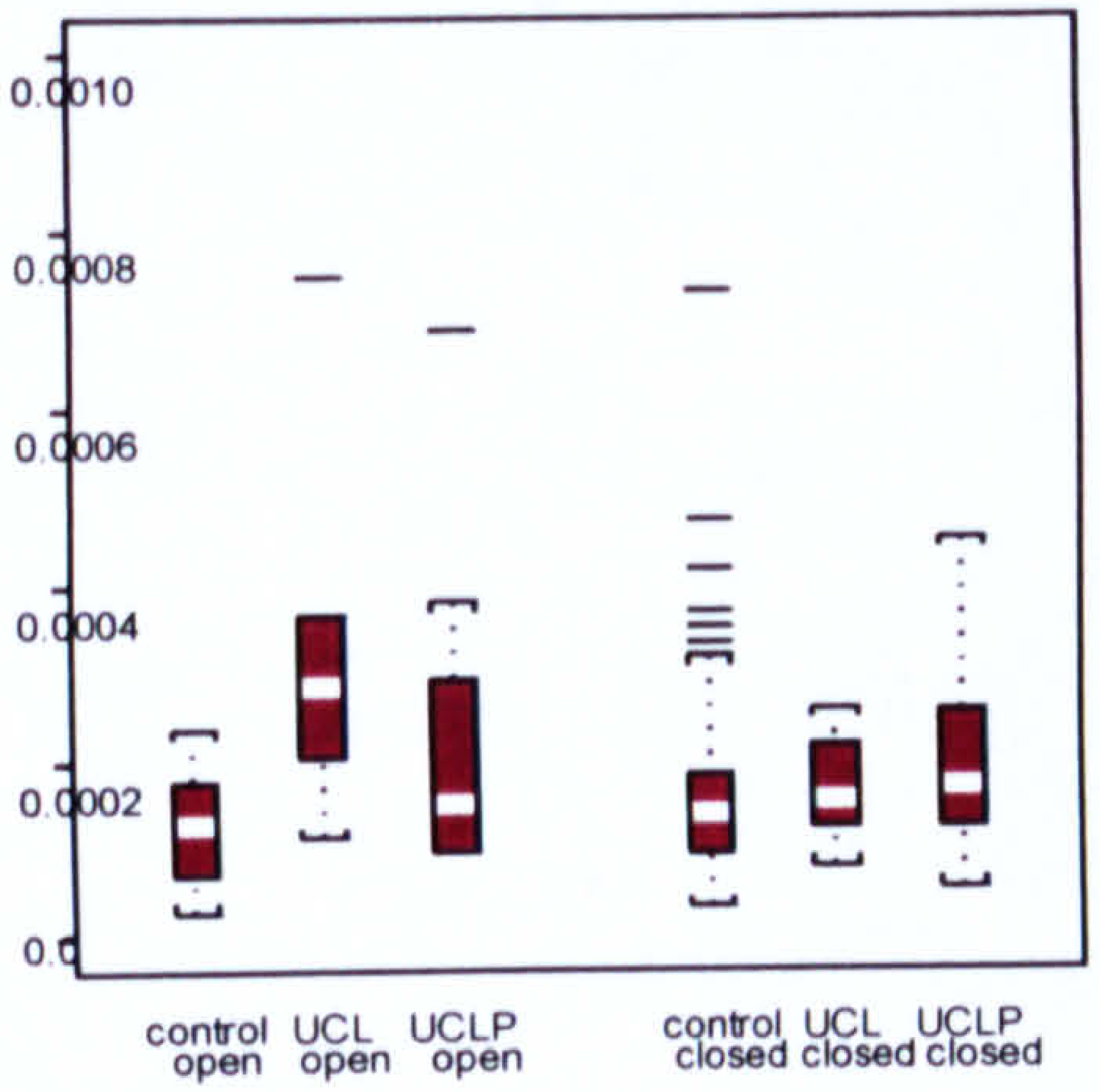
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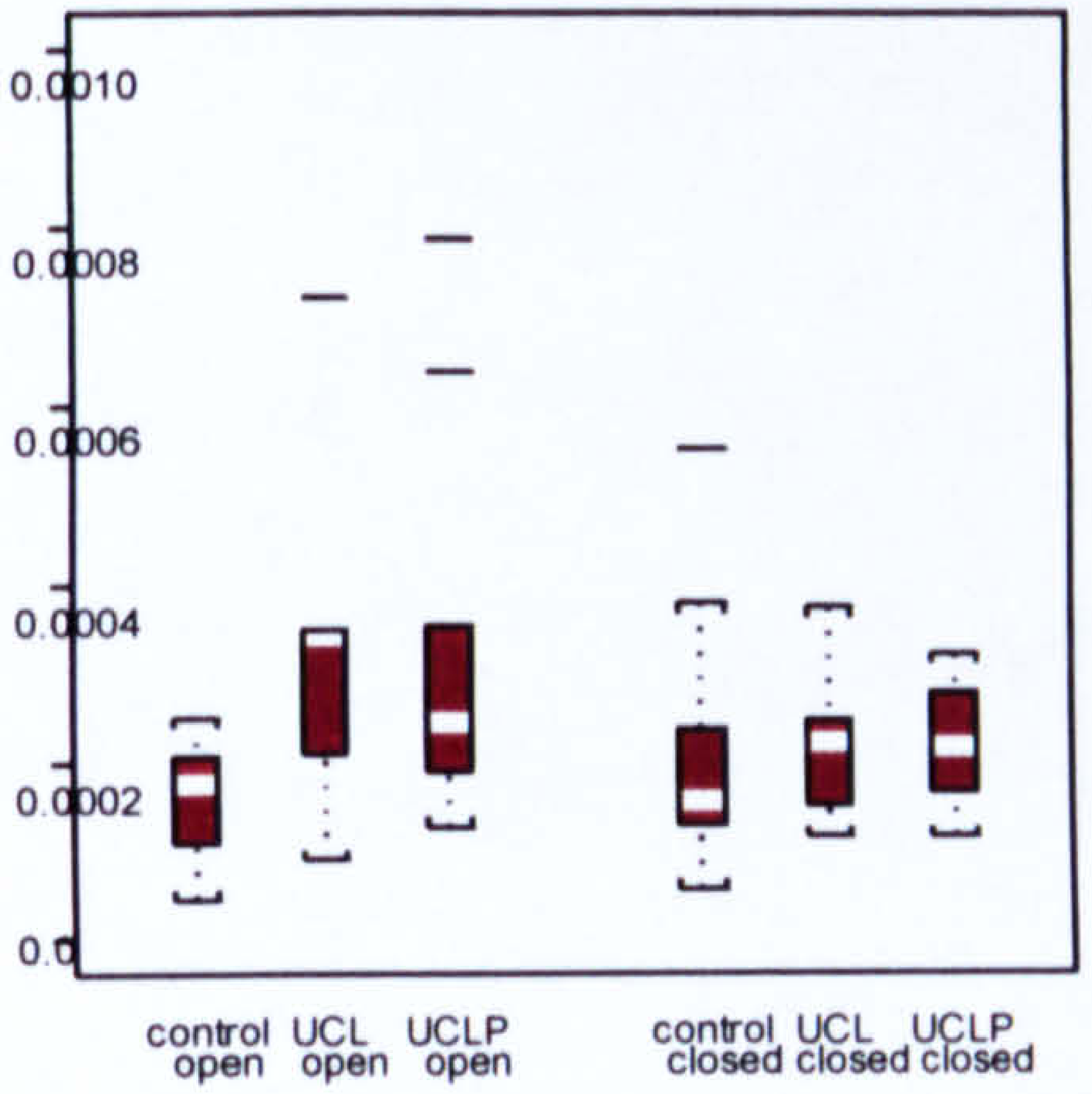
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