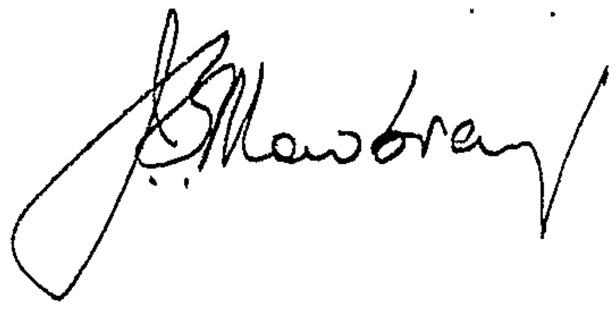
THE UNIVERSITY OF
SYDNEY

A STUDY OF FACIAL GROWTH IN CHILDREN

WITH TREATED UNILATERAL CLEFT LIP AND PALATE

This thesis, embodying original work, is submitted as a partial fulfillment of the requirements for the Degree of Master of Dental Science in the Faculty of Dentistry, University of Sydney.



John B. Mowbray, B.D.Sc. (Hons.) U. of Q.
Department of Preventive Dentistry,
November 1973.

CONTENTS

INTRODUCTION

Cleft Lip and Palate conditions have been recorded since earliest civilizations. Smith and Dawson (1924) noted a case of cleft palate in their study of Egyptian mummies and more recently, Skoog (1969) has demonstrated a cleft lip condition in a miniature pottery head found in excavations in ancient Greece.

Today at least one birth in every thousand has a cleft condition of the lip or palate, or a combination of these abnormalities. Our recording methods are replacing the qualitative assessments with the quantitative and hopefully, we are moving closer to a fuller comprehension of these conditions.

Most adults could readily describe the appearance of a repaired cleft lip; they would just as readily associate a nasality of speech with cleft palate. It is interesting, perhaps merely conjecture on my part, but one might just as readily expect the comment that one seems to be less aware of such cases nowadays.

Facial aesthetics are judged in terms related to normal growth. Each of the various types of cleft conditions has its own degree of growth potential which is dependent on the extent of the initial insult. This growth potential can be enhanced by rehabilitative techniques, but this requires the successful integration of the involved specialities to achieve the most favourable results.

From observations at the Cleft Palate Clinic at the Royal Alexandra Hospital for Children in Sydney, there is a high degree of multidisciplinary involvement and integration. The Plastic Surgeon,

the E.N.T. Surgeon, the Speech Therapist, and the Orthodontist all provide a combined effort to marshall what growth potential there is and redirect it in the most desirable direction possible.

The 'Unilateral Cleft Lip and Palate' group has been singled out as one that appears to have a generally favourable growth prognosis. In view of the combined efforts of this Cleft Palate Clinic there may well be some justification in saying how much better the results seem today. It is the intention of this investigation to examine a group of such patients and assess their growth status in the early mature dentition stage.

The following presentation is in two parts. Part One is a review of the relevant literature and forms two sections. The first section is concerned with the development, aetiology, and epidemiology of cleft lip and palate. The treatment of the unilateral cleft lip and cleft palate, and the significance of growth in this condition is discussed in the second section.

Details of the investigation are presented in Part Two; this is followed by a discussion of the results, the conclusions, and a summary.

PART ONE

Section One

1. EMBRYOGENESIS

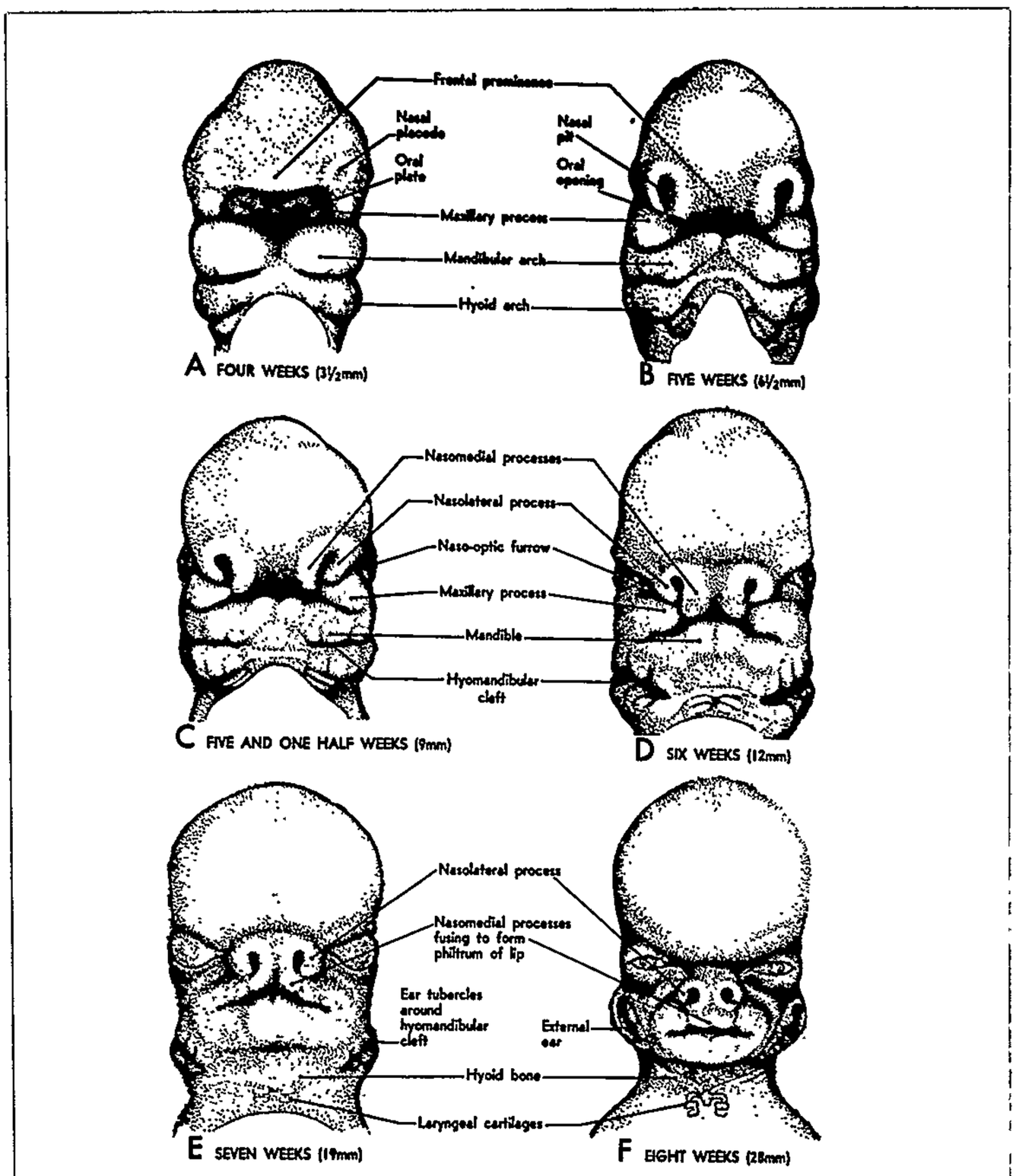
(a) Development of the Primary Palate

It is not until the fifth week of intra-uterine life (6.5mm. C.R.) that the developing embryo exhibits organisation of the para-stomodeal structures. The maxillary, medial-nasal and lateral-nasal processes extend to form the natural superior limits of the primitive mouth opening.

During the next fourteen days this upper 'lip' becomes more defined; the organisation of these processes is completed by eight weeks (28mm. C.R.). (Patten, 1964, 1971). This is clearly seen in Fig. 1.

Embryologists differ in their concepts of primary palate formation. The classical concept outlined by Dursey, and by His, has been referred to often in the literature. (Stark, 1954). Fusion of the projecting processes of ectodermal covered mesoderm was thought to complete the upper lip; the nasal pits were conceived as cul-de-sacs which joined with the oral cavity by disintergration of the bucco-nasal membrane. (Scott and Symons, 1958).

Pohlman's thesis and the later work of Veau, referred to by Ross and Johnston (1972), outline the concept of mesenchymal penetration. This gained impetus with Streeter's publication (1948). Thus, the idea that mesenchyme provides the basis of facial development has become accepted.



Drawings showing, in frontal aspect, some of the important steps in the formation of the face. (After William Patten, from B. M. Patten: *Human Embryology*, 3d ed., 1968, courtesy of the Blakiston Division of McGraw-Hill Book Company.)

FIGURE 1

The absence of discrete clefts and processes in the central third of the normally developing face is an important feature of this concept. The mesodermal contribution to the 'intermaxillary segment' (Patten, 1971), is generally thought to arise from the maxillary and medial-nasal areas. Tondury (1958, 1964), and Fraser (1968), include lateral-nasal mesoderm as well. The general view is one of mesodermal penetration and coalescence.

Recent work of Trasler (1968) indicates an invagination process of the nasal pit. Olfactory epithelium migrates until direct communication occurs between the nasal and oral cavities. This reinforces the earlier findings of Streeter (op. cit.). By such invagination, mesoderm automatically becomes integrated within this primary palate area; migration of mesoderm may not necessarily occur.

Thus, primary palate formation appears to be one of epithelial invagination in the region of the nasal placodes, together with a concomitant process of epithelial fusion, dissolution and penetration by mesoderm.

(b) <u>Development of the Secondary Palate</u>

There is general agreement on the embryogenesis of the secondary palate. During the seventh week (20mm. C.R.) of intrauterine life, the maxillary processes proliferate medially. Initially the tongue lies between these vertically orientated extensions, then from the posterior, the palatal shelves commence

a horizontal alignment. Zeiler et al. (1964) noted this occurence in rat experiments. Termed 'shelf force', it was thought related to a growth differential between the maxilla and the mandible which in turn allowed the tongue to drop. Fraser (1967) refers to this 'shelf force' in man and its ability to overcome the resistance of the tongue.

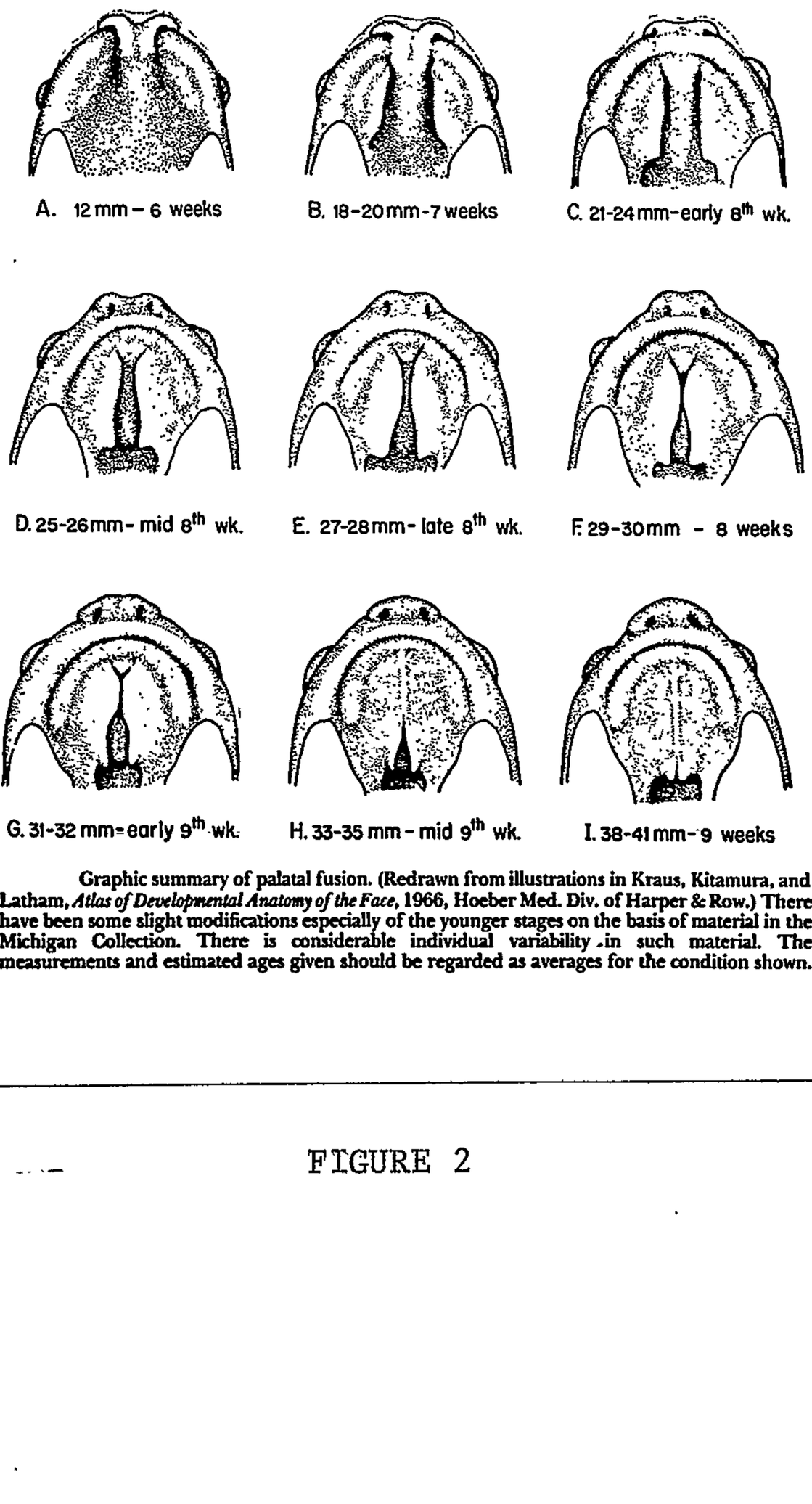
This concept of a force which causes these palatal shelves to change their alignment has been questioned by Ross and Johnston (op cit.). They point out that this phenomenon may well be peculiar to rodents and of doubtful consideration in man.

By the end of eight weeks (28mm. C.R.) the palatal shelves have contacted; usually about one third of the ultimate length from the incisive foramen. The nasal septum descends at this stage and fuses finally with the palatal processes. Palatal closure is completed by nine weeks (40mm. C.R.). Fraser (op cit.) suggests that soft palate formation appears to be one of merging rather than fusion.

Figure 2 and Figure 3 show these activities clearly.

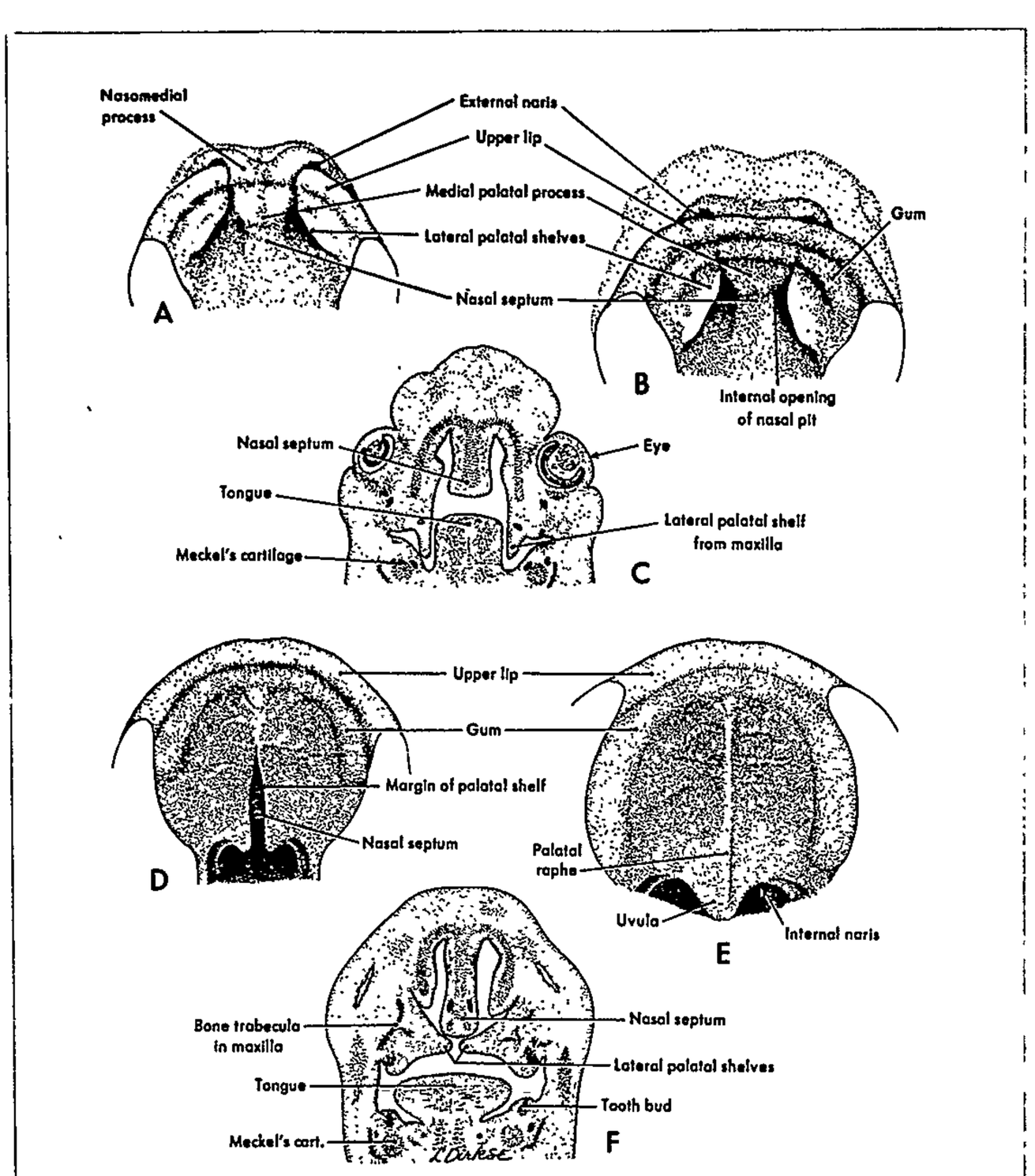
(c) Mechanisms of Cleft Formation

The 'classical' theory of middle face development allowed for clefting of the primary palate by simple failure of the fusion process. However, with the wide acceptance of the 'mesodermal penetration' ideas, cleft formation is now thought to be directly



Graphic summary of palatal fusion. (Redrawn from illustrations in Kraus, Kitamura, and Latham, *Atlas of Developmental Anatomy of the Face*, 1966, Hoeber Med. Div. of Harper & Row.) There have been some slight modifications especially of the younger stages on the basis of material in the Michigan Collection. There is considerable individual variability in such material. The measurements and estimated ages given should be regarded as averages for the condition shown.

FIGURE 2



The development of the palate. (The collaboration of Dr. James K. Avery in the shaping of this illustration is gratefully acknowledged.) A. Upper jaw and roof of the mouth as seen from below in embryos a little over 6 weeks old (C–R, 14 to 15 mm.). B. Similar view in embryos of about 7 weeks (C–R, 19 to 21 mm.). C. Projection drawing of a section through the head of a 7-week embryo to show how the palatal shelves at this stage are separated by the tongue. (University of Michigan Collection, EH 358; C–R, 19mm.) D. The palate during the ninth week; fusion of the lateral palatal

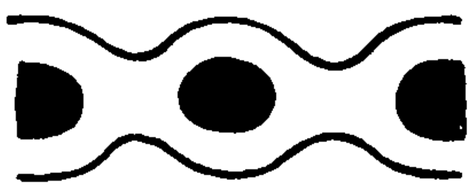
FIGURE 3

attributable to a lack of embryonic mesenchymal proliferation. This has been shown by Tondury (op. cit.) to be directly related to epithelial dissolution. Tondury's findings were similar to those of Steiniger (1939); the failure of epithelial fusion, dissolution and mesenchymal consolidation can occur at various stages of embryogenesis. It was noted even with the apposition of the epithelia of the facial processes. Thus, there can be a direct relationship between abnormal facial proportions and cleft formation.

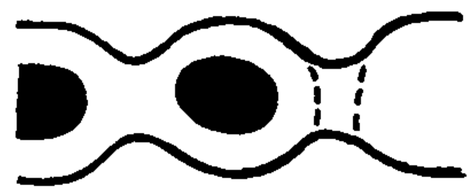
Stark (op. cit.) illustrates this concept of mesenchymal penetration as represented in Figure 4. He relates this rupture effect in the cleft areas to the presence of Simonart's bands and to the lack of lateral incisors; mesoderm is a precurser of pulpal tissues.

Clefting of the secondary palate is generally thought to be due to a lack of contact between the palatal shelves. Kraus (1970) maintains that the palatal shelves meet at about forty seven days and fusion is completed within one week. Palatal fusion of the maxillary and palatine bones in the midline occurs at about ten weeks. Kraus suggests that the majority of clefts in man are most likely to occur between seven and a half and ten weeks 'in utero'.

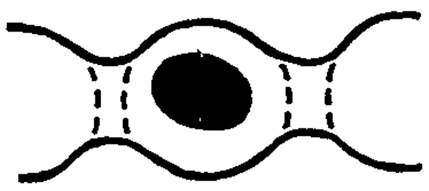
This author also refers to the presence of 'epithelial rests' in the midline at this stage. These may be found later as cysts. Burke et al. (1956) as well as Scott (1955) have noted these formations at a later stage of intra-uterine growth.



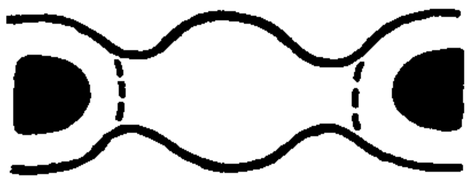
NORMAL



UNILATERAL HL



BILATERAL HL



MEDIAN CLEFT

The pathogenesis of harelip is presented diagrammatically. An epithelial wall exists initially as the anlage of the upper lip, premaxilla, and upper incisor teeth (primary palate). Three mesodermal volumes are located in it, one in the midline and two laterally. These grow and fuse, forming the normal upper lip, premaxilla, and four upper incisor teeth. If one volume of mesoderm is absent the epithelial wall will rupture and a cleft will occur in that area. The absence of a lateral volume will result in a unilateral harelip. Absence of the medial volume will result in a median cleft of the lip, a rare anomaly, and absence of the two lateral volumes will cause a bilateral harelip to occur.

FIGURE 4

Burdi and Faist (1967) found that these cysts were absent from the soft palate and propose this as evidence of soft palate formation by a process of merging rather fusion.

Johnston (1964) observed interesting cellular activity in the neural crests of chick embryos; extirpation of the neural crest on one side of the fore-brain frequently results in clefting. Thus, it could occur that clefting is associated with neural crest cell migration long before the facial processes are formed. Fraser (1968) extends this cellular concept to question the determinants of such migration in the neural crest, the sites of the nasal placodes and the topographical relations of the facial processes.

In summary, the normal embryonic primary and secondary palates appear to develop from a process of ectodermal fusion with the concomitant penetration and coalescence of mesoderm. The soft palate, however, would appear to form by a merging process.

Embryogenesis of the cleft lip and cleft palate is thus seen to be in terms of a breakdown in the proliferation process of the embryonic mesenchyme.

2. PRENATAL DEVELOPMENT OF THE UNILATERAL CLEFT LIP AND PALATE

The muscles of the nose and lips are formed from the primitive muscle cells which migrate from the hyoid arch region. This cellular organisation occurs in the sixth to seventh week of intra-uterine life. The result of this organisation and muscle

cell migration is readily seen in the muscle fibre directions in the orbicularis oris muscle. Myoblastic differentiation is in the horizontal plane conforming with the muscle layers of this muscle. (Ross and Johnston, 1972).

In the case of the unilateral cleft the arrangement of the muscle fibres is completely disturbed. Usually these fibres resemble those of normal lip musculature. Fara (1968), Pfeifer (1966). In this latter case cleft formation is thought to occur at a later date.

Muscle development in the secondary palate shows fibres coursing medially and inserting into the aponeurosis of the soft palate; this is commonly known as the midline raphe. Ross and Johnston (op cit.) illustrate this altered muscle growth in Fig. 5. Instead of seeking insertion in the aponeurosis, the fibres of the tensor and levator palati muscles find attachment in the posterior margin of the bony palate.

Alteration of the nervous and vascular tissue orientation follows a similar pattern. Both Fara (1968) and Pfeifer (1966) report that the respective terminal branches which would normally track to adjacent areas or anastomose, form into divergent pathways according the tissue distortion.

The developmental activity of primary and secondary clefts is essentially a localised phenomenon during the period of five to ten weeks I.U. (Kernahan and Stark, 1958). However, Wood (1972)

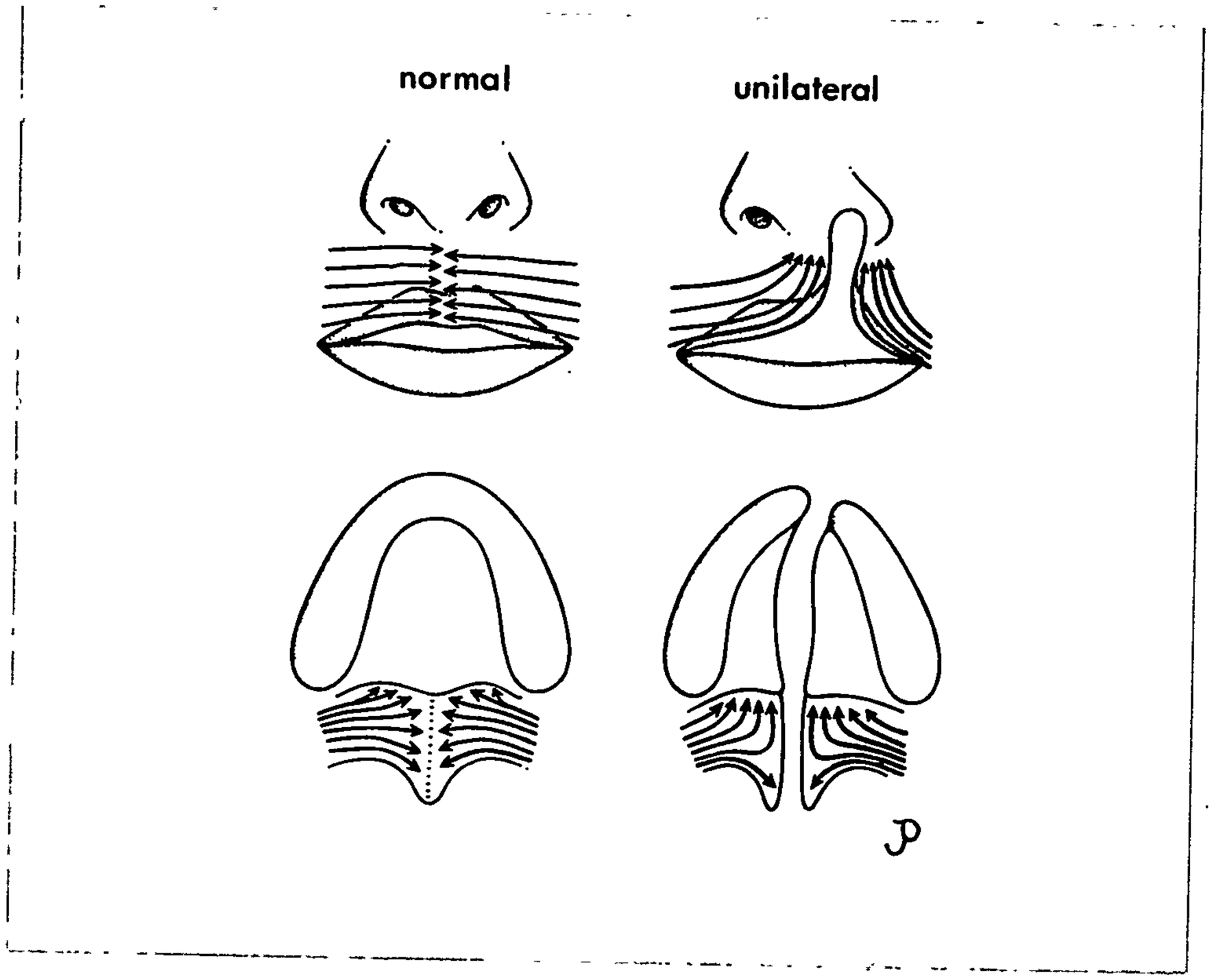


FIGURE 5. Migration paths followed by myoblasts entering a normal and bilateral cleft lip and palate.

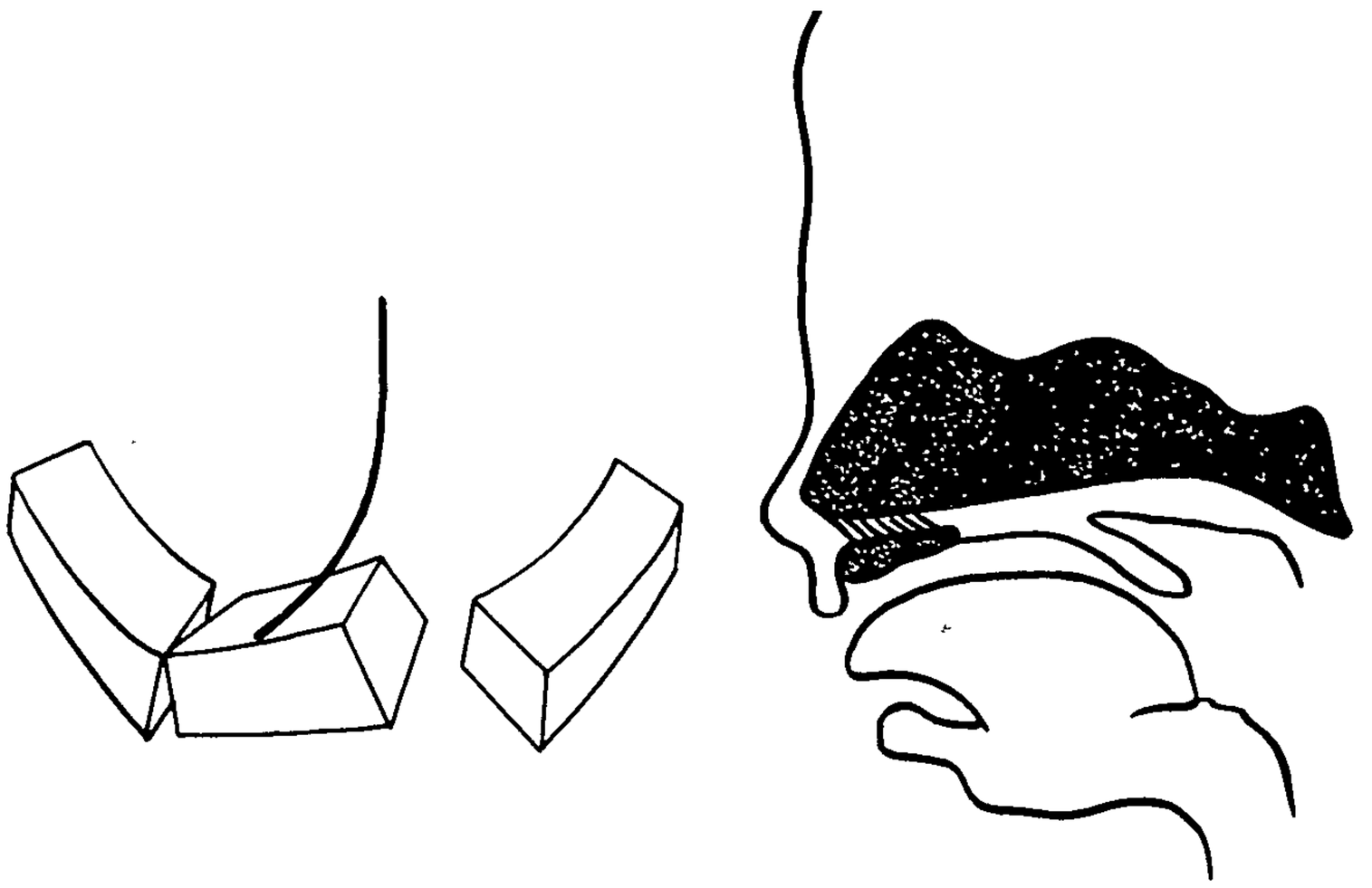
has found that during this period to twelve weeks I.U. the nasal septal cartilage commences a more forceful growth spurt. This is thought to stimulate downward and forward growth in the related bony structures. In the fourth month I.U. this growth activity slows down but there still exists a growth impetus in the facial middle third.

Septo-premaxillary Ligament

Associated with this nasal septum growth is the finding of Latham (1969). He describes an anatomical structure which forms a link between the cartilaginous nasal septum and the premaxilla. Termed the 'septo-premaxillary ligament', it is described as 'a clearly defined ligament.....coursing from the anterior border of the nasal septum to the anterior nasal spine and the median suture of the premaxillary region'.

Thus a mechanism has been identified which exists during foetal life allowing the concomitant forward and downward displacement of the septum and the maxilla. Fig. 6. after Wood (op. cit.) illustrates this structure as well as its relationship in the unilateral cleft of the primary palate.

Latham (op. cit.) referred to the 'intrinsic' growth mechanisms of later foetal and post-natal life which continue this facial development. This surface apposition and resorption as an expanding growth mechanism has been well described by Enlow and



Block illustration of the maxillary arch showing the forward, lateral, and upward displacement of the premaxilla.

Sagittal section of the fetal head indicating the directional pull of the septo-premaxillary ligament.

FIGURE 6

Bang (1965). Associated with this is the 'functional matrix' theory of Moss (1962); as the soft tissue structures begin to increase in size with developing body function, the related skeleton grows as well.

It can be concluded that the condition of unilateral cleft lip and palate in the neonate is the summation of redirected normal growth potentials in an embryologically disturbed area. Contiguous structures pursue less restrained growth and exhibit deformation until they become confined by the unaffected yet adjacent anatomical relationships.

3. AETIOLOGY OF CLEFT LIP AND PALATE

Embryonic disturbances can be exerted from two directions; they can be either genetic or environmental. An examination of the causative factors involved in cleft lip and palate embryogenesis presents a complex picture. There appears to be an interaction between these genetic and environmental factors.

(a) Genetic Factors

Fogh-Anderson (1961) refers to his earlier investigations in 1942 which showed the difference by sex of, and familial disposition to, cleft lip and cleft palate conditions in his Danish study. Cleft lip, with or without cleft palate, Cl(P), occurs most often in males as a recessive genetic character, while the isolated

cleft palate, CP, is found more frequently in females as a dominant characteristic.

In their Edinburgh study, Drillien, Ingram and Wilkinson (1966) noted that twice as many males with isolated cleft palate gave a positive family history as females and, in the case of the cleft lip with or without cleft palate, one and a half times as many females gave a positive family history as did the males.

Carter (1964) examined the incidence of cleft lip with or without cleft palate in familial populations. The incidence was found to decline rapidly from the first degree relationships to those of the third degree, at which level the incidence approximated general population levels. The degrees of incidence for first, second and third degree relationships were 3.5, 0.6 and 0.2 per cent.

Lip Pit Syndrome

This condition described by van der Woude (1954) appears to be the exception to this genetic independence of these two groups of cleft conditions, that is, the isolated cleft palate and the cleft lip with or without cleft palate. It is a dominant hereditary character but it exhibits a variable penetrance and expressivity. These mutations occur in about 1 to 2% of the CL(P) group. Ross and Johnston, 1972.

Chromosomal Variations

There are certain chromosomal configurations which cause cleft lip and palate formation together with other malformations. The most well known of these is Down's syndrome, or trisomy 21. These autosomal variants account for about 1% of the total cleft group found in the general cleft population. Ross and Johnston (op. cit.) are of the opinion that chromosomal abnormalities constitute about 10 to 12% of the total newborn cleft population but that few survive to become part of the clinic populations. Pateau et al. (1960) describe one such case of autosomal variation with cleft palate.

(b) Environmental Factors

Chromosomal relationship between monozygous twins was examined by Metrakos et al. (1963); they noted a lack in this relationship. In observations of clefting in monozygous twins, Ross and Coupe (1965), and Younglèson (1971) have both recorded a lack of concordance for cleft lips with or without cleft palate.

If complete genetic relationship were to be the case then we could expect 100% concordance. Thus we can assume that the environmental influence is of definite importance.

Physical Environment

Little is known of the physical influence of the uterine environment. Such factors as the foetal position, uterine pressure

and fluid content have yet to be fully investigated. Trasler et al. (1956) in mice experiments found a highly significant degree of cleft palate formation following loss of amniotic fluid. This was also noted by Poswillo and Roy (1965) who proposed that the loss of amniotic fluid and the change in the fluid dynamics can produce a temporary change in the position of the embryo, and the teratologic changes in palate formation.

Metabolic Environment

Kraus (1970) induced clefts in rodents with the presence or absence of agents. Such teratologic agents included riboflavine deficiency, folic acid deficiency, vitamin B 12 deficiency, hypervitaminosis A, trypan B, ionising radiation, hormones, salicylates and cortisone. Poswillo and Roy (op. cit.) noted this earlier with ionising radiation, as well as with the combined doses of vitamin A and cortisone.

In human subjects there is little evidence to support similar relationships. Harris and Ross (1955) reported cleft palate formation following high doses of cortisone during pregnancy. In a study of the offspring of diabetic mothers, Vallance Owen et al. (1967) noted a relationship between insulin antagonism and cleft deformities.

At an experimental level, McKenzie (1968) points out the importance of the embryonic blood supply. Any vascular anomaly could produce a failure of cellular differentiation with resultant teratogenic possibilities.

Viral disturbances, although unconfirmed by epidemiologic studies may effect embryonic malformations. Blattner and Heys (1961) showed a relationship between rubella in the mother and sibling malformations including cleft lip. Induced clefting in experimental animals has been noted, Ferm and Kilham (1964), but research in this direction is scant.

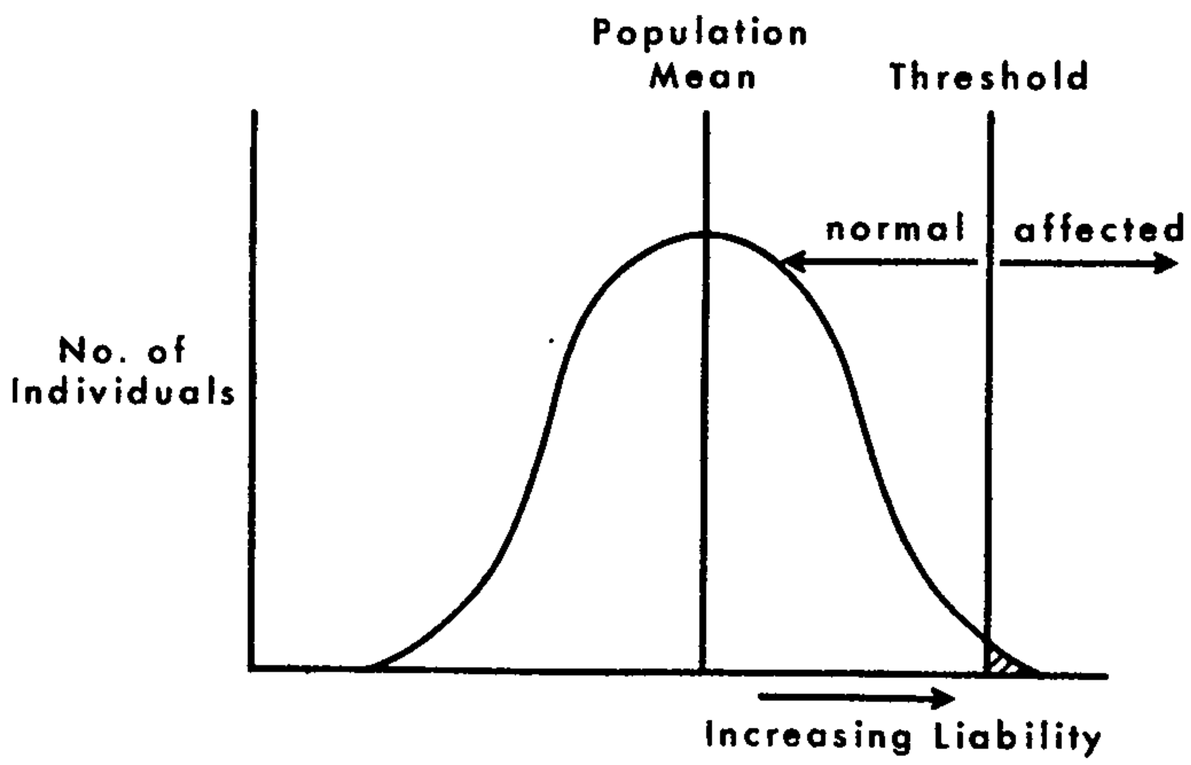
Ross and Johnston (1972) comment on the possibility of placental transfer of antibodies as is commonly seen with the Rh factor and suggest that this mechanism could produce cleft formation.

It would seem that the term of Fraser (1971), 'multifactorial' is the best description of the aetology as we know it today. Falconer (1965) refers to cleft formation as a 'threshold character', these threshold characteristics occur at the tail of the normal distribution curve. There is a point where the pattern of normal embryogenesis is not capable of effecting the correct relationship because of a number of factors. These include such conditions as shelf width and the degree of relationship to affected subjects. This is shown in Fig. 7.

4. EPIDEMIOLOGY

Studies indicate that the incidence of cleft lip with or without cleft palate is of the order of one per thousand live births. A review of earlier studies in Europe and the U.S.A. was presented by Sesgin and Stark (1961) and is reproduced in Fig. 8.

Evidence suggests that incidence is increasing. Fogh-Anderson (1964). There appears to be a positive increase as



Falconer's concept of liability and threshold for expression of a trait. The liability for the trait in each individual is composed of multiple genetic and environmental factors. If the factors exceed the threshold, the trait is expressed.

FIGURE 7

distinct from the expected increase from the improvements in data collection and recording, as well as the increased 'marriageability' of patients with more modern and aesthetic repairs.

Data collected by Chi and Godfrey (1970), is of particular relevance to this presentation; the incidence and distribution of cleft lip and cleft palate cases was compiled from information supplied from N.S.W. Hospitals.

The incidence of cleft defects in this state for the years 1964, 1965 and 1966 was 1.21 per 1,000 live births; this can be expressed as 1:821. This ratio is comparable with studies tabulated in Fig. 8.

This survey also found that there was a significantly greater incidence of clefts of the lip and palate in males, and in such cleft lip cases the left side was affected twice as often as the right side. These figures are in keeping with other reports. (Schade, 1971).

In comparing the incidence of cleft type within the total group of clefts, Chi and Godfrey (op. cit.) were again in agreement with other studies. The incidence of combined cleft lip and cleft palate is about 50% with isolated cleft lip and isolated cleft palate in equal proportions. This figure includes both unilateral and bilateral clefts.

Incidence of cleft lip and palate

Year	Investigator	Source	No. Total Patients	Incidence
1833-63	Frobelius	St. Petersburg, Russia	118:180,000	1:1525
1908	Risehbieth	London	39:67,945	1:1742
1924	Davis	Baltimore	24:28,085	1:1170
1929	Péron	Paris	106:100,889	1:942
1931	Schröder	Münster, Germany	28:34,000	1:1214
1931	Gunther	Leipzig, Germany	102:102,834	1:1000
1933	Sanders	Leiden, Rotterdam, Groningen, Holland	16:15,270	1:954
1934	Grothkopp	Hamburg, Germany	74:47,200	1:638
1934	Faltin	Finland		1:950
1934	Sanvenero-Roselli	Italy		1:1000-1:1500
1939	Edberg	Göteborg, Sweden	28:27,000	1:960
1939	Fogh-Andersen	Copenhagen, Denmark	193:128,306	1:665
1940	Conway	New York	32:22,513	1:700
1940	Henderson	Hawaii	35:18,021	1:550
1942	Grace	Pennsylvania	250:202,501	1:800
1935-44	Mueller	Wisconsin	736:567,504	1:770
1949	Hixon	Ontario, Canada	695:655,332	1:943
1950	Ivy	Pennsylvania	766:583,690	1:762
1951	Wallace *et al.*	New York		1:1265
1953	Wallace *et al.*	New York		1:1202
1950-54	Douglas	Tennessee		1:1694
1955	Lending *et al.*	New York		1:1342
1960	Sesgin and Stark	New York	21:27,087	1:1289

had Grace some 8 years before. A year earlier (1949) Hixon found the incidence to be 1:943 in Ontario, Canada.

In the last decade, studies from vital statistics of the City of New York have been shown by Wallace, Hoenig, and Rich, to be 1:1265 in 1951, and 1:1202 in 1953. The figures by Lending, Jacobziner, Grossi, Kugler, and Rich in the City of New York were 1:1342 in 1955.

FIGURE 8

In a specific investigation at Royal Alexandra Hospital for Children the percentage of operated cleft defects which were unilateral cleft lip and cleft palate can be seen from Fig. 9.

Distribution of Infants Undergoing Primary Surgery at Royal Alexandra Hospital for Children During the Second Halves of Four Consecutive Years (1966 to 1969)

Type of Cleft	1966	1967	1968	1969
Left cleft lip only	6	9	4	2
Right cleft lip only	3	1	—	—
Bilateral cleft lip only ..	—	1	1	1
Left cleft lip with cleft palate	8	10	11	4
Right cleft lip with cleft palate	2	4	4	2
Cleft palate only	12	16	19	10
Bilateral cleft lip with cleft palate	3 (8·8%)	6 (12·8%)	4 (9·3%)	6 (24·0%)

FIGURE 9

Section Two

1. SURGICAL REPAIR OF THE UNILATERAL CLEFT LIP AND PALATE

Lip Repair

Surgery of the soft tissue cleft in the lip is the first major step in the rehabilitation of the cleft lip.

The development of cleft surgery appears in retrospect, to have gone through a 'dark age' in the years up to the second half of this century. Even with the use of such techniques as 'flap' procedures by Mirault in 1844 and Hagedorn in 1892, the mutilating methods of Brophy (1923) gained prominence.

The publication of LeMesurier's technique in 1949 (Fig. 10) was a milestone in this aspect of surgery. With admitted deference to the earlier procedures of Mirault and Hagedorn, LeMesurier (1949), Steffansen (1949), 1953) and Tennison (1952) have greatly influenced our present concepts of this type of surgery. Such methods which employ the benefits of 'Z-plasty' philosophy have enabled surgeons to produce results in accordance with Steffensen's criteria. (1949).

For satisfactory repair of the single cleft lip there should be:

(a) Accurate skin, muscle and mucous membrane union.

(b) Symmetrical nostril floors and vertical nostrils.

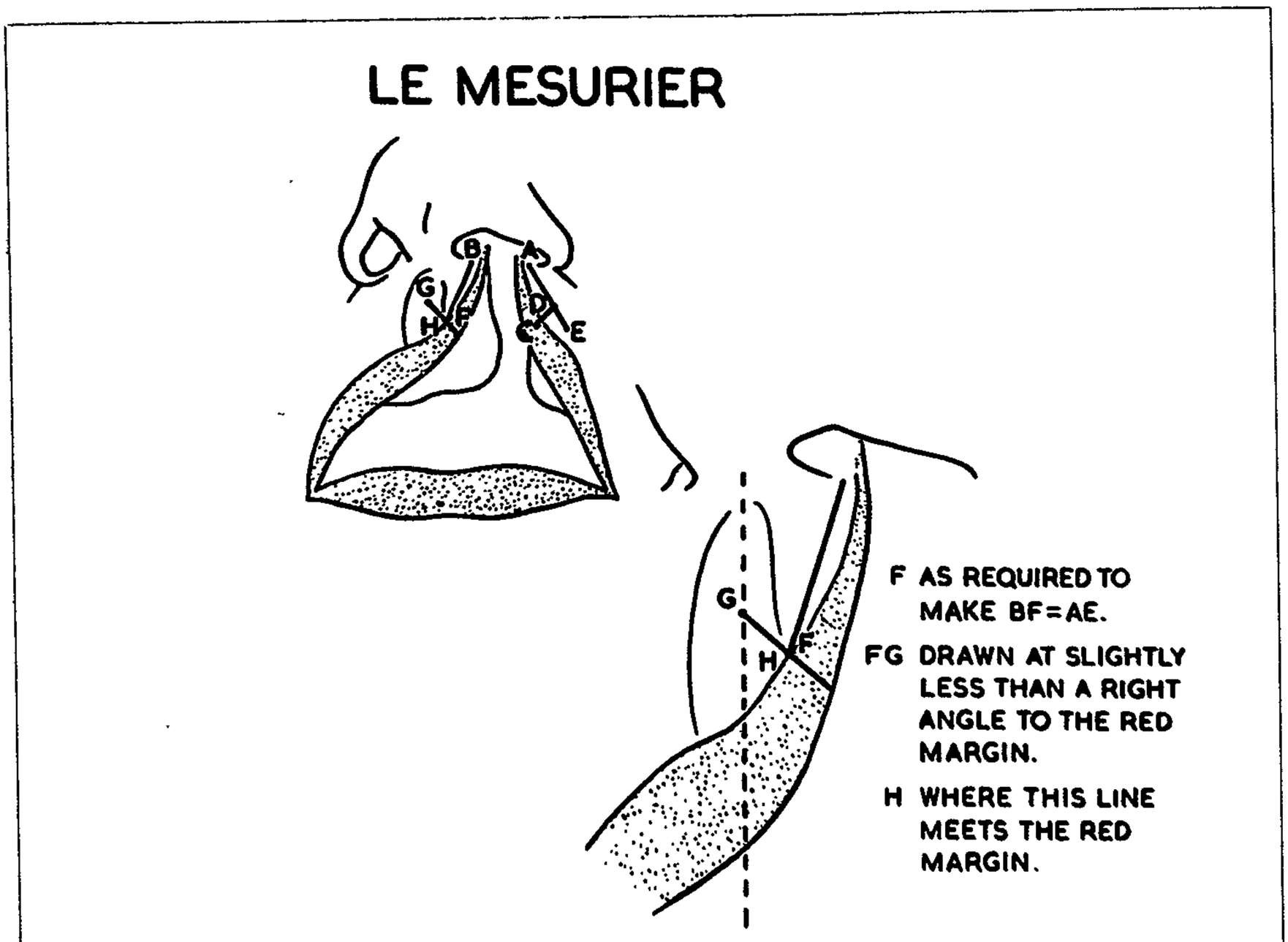

LE MESURIER
A
B
C
D
E
F
G
H
F AS REQUIRED TO MAKE BF=AE.
FG DRAWN AT SLIGHTLY LESS THAN A RIGHT ANGLE TO THE RED MARGIN.
H WHERE THIS LINE MEETS THE RED MARGIN.

FIGURE 10

(c) Symmetrical vermilion border and Cupid's Bow reproduction.

(d) Slight eversion of the lip.

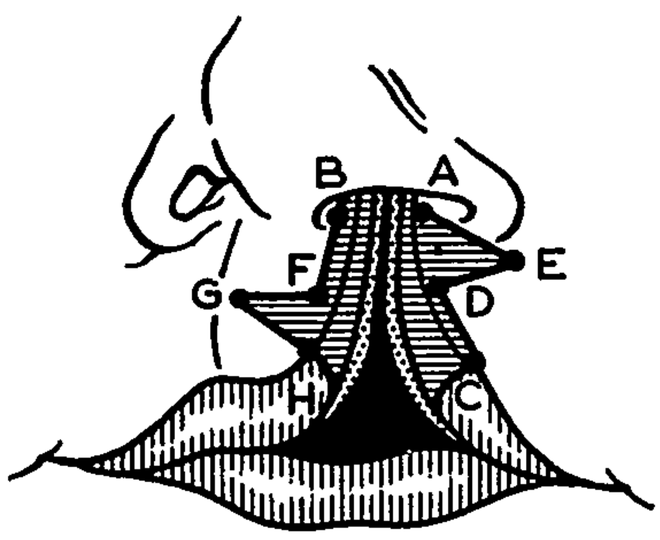
(e) Minimal scarring which by its contraction will not affect the above criteria.

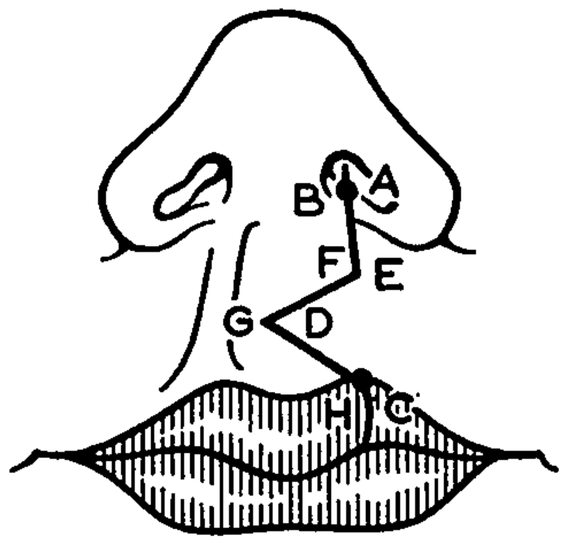
The method of surgery used on the subjects in this survey is based on the Tennison method of lip repair. This is represented in Fig. 11.

The recently advocated technique of Millard (1964) and its later refinements (1967) appears to avoid the zig-zag scarring of previous techniques by following the philtram. However as Tennison (op. cit.) points out, surgical complexity may present a challenge to the practised surgeon but is directioned away from the original concept of operational standardisation by technical simplicity.

Palate Repair

Closure of the cleft palate provides separation of the nasal and oral cavities. This aids in the development of normal speech, as well as improves masticatory function and the efficiency of deglutition. The success of surgical treatment up to the middle of the nineteenth century relied on the stability of mucosal suturing. The excessive tensions produced by mere appositional techniques resulted in severe limitations of the maxillary growth potentials,





Yoder.

The Tennison operation.

A, B. Points at the nostril margin, approximation of which will constitute a satisfactory floor.
H. Peak of the Cupid's bow on the cleft side.
F. So that BF is half the height of the lip on the uncleft side (usually 4 to 5 mm.). F is usually 1 or 2 mm. from H.
G. So that GH equals FG equals BF. (BG should be 2 mm. longer than BF).
C. On the red margin where vermilion is of normal thickness.
D. On the red margin so that CD equals BF.
E. So that AE equals DE equals CD. (CE should be 2 mm. longer than CD.)

FIGURE 11

and led to the development of lateral incisions to ease these tensions. This innovation of Dieffenbach in 1826 was further modified by Von Langenbeck in 1861; the complete mucoperiosteum was freed and sutured across the cleft leaving a raw palatal bone surface to heal by granulation cover. In 1871 Billroth suggested the fracture of the hamular processes to relieve this posterior tension in the soft palate.

Mention must be made of the technique of the American surgeon Brophy who practised his own type of early osteouranoplasty. (Brophy, 1927). The use of silver wire and lead plates to effect closure of the 'normally' developed but separated areas of the palate had long term disastrous results. Mutilation of the dentition and growth of the maxilla was common with his method of palate repair; this was most frequently seen in North America where his influence was widespread. (Graber, 1949).

Veau in 1931, published a collection of case histories which conclusively showed the benefits of his modifications to the Von Langenbeck method. Speech assessment indicated his success with methods of palatal repair incorporating the nasal mucosa; scar contracture was much reduced with improvements in the mobility and length of the soft palate.

Wardill (1937) and Kilner (1937) both described methods for palate lengthening. This has since become known as 'V-Y Retropositioning' or more simply as 'V-Y Pushback'. This method

enables the palate to be closed and lengthened simultaneously. Lateral incisions, in most cases hamular fracturing, as well as freeing of the palatal mucosa are incorporated in the technique. The significant difference is the rotation and advancement of the flaps posteriorly. Continuity of the palate is not lost and it is significantly lengthened.

According to the degree of clefting this operation uses three or four flaps as shown in Figs. 12a and 12b. The 'V-Y Pushback' has been used exclusively in this group of patients selected for this survey. This operation is usually one year after the lip has been repaired.

2. ASSOCIATED TECHNIQUES IN CLEFT LIP AND CLEFT PALATE REHABILITATION

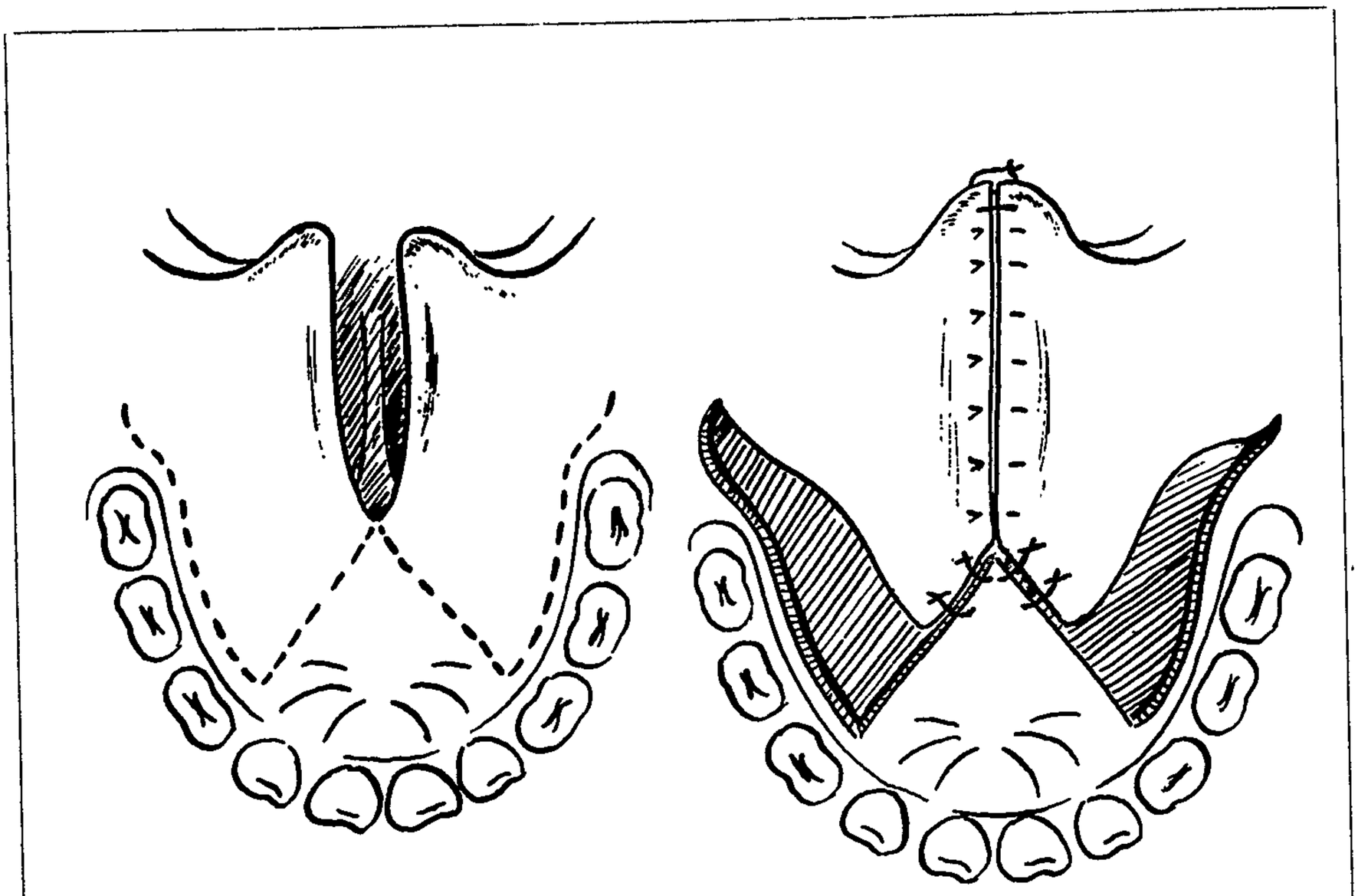
Although the patients selected for this survey have been treated with the two described techniques only, a brief resume of two associated techniques is presented. These are:

(a) Pre-surgical orthopaedics, and

(b) Bone grafting procedures.

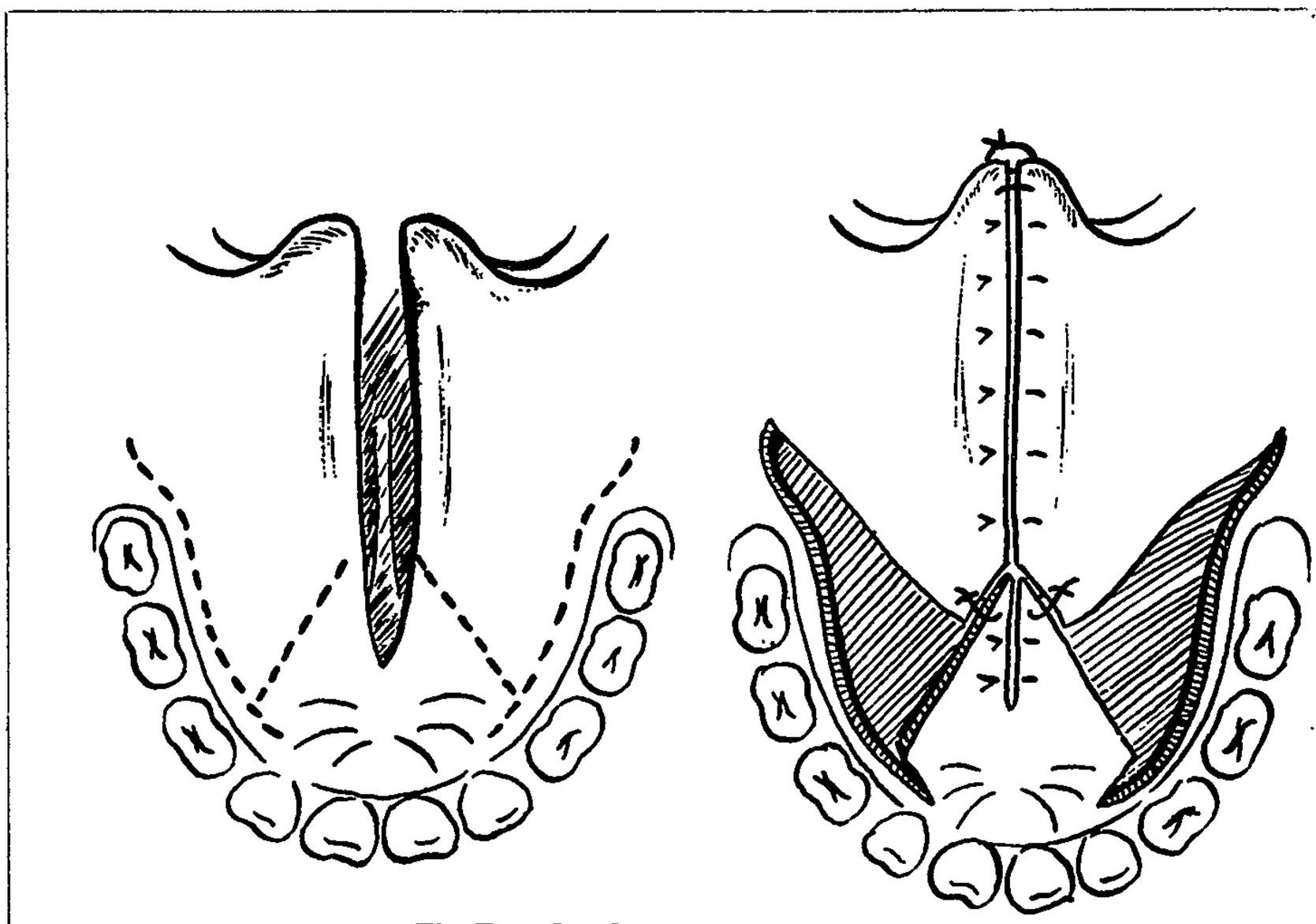
(a) Pre-surgical orthopaedics

The publication of McNeil's thesis in 1954 and his other contributions in 1950, 1956, and 1964 introduced a new concept in cleft lip and cleft palate rehabilitation. McNeil devised a type of sucking plate with a symmetrical arch form that would enable the moulding of neonatal irregularities.



The Three-flap Operation (Wardill) for shorter cleft.

FIGURE 12a



The Four-flap Operation (Wardill) for extensive cleft.

FIGURE 12b

In its original concept, the 'McNeil' plate exerts an orthopaedic force on two parts of the maxilla. Alignment of the malposed segments occurs and in this way maximum advantage is taken of this period of rapid growth. Surgical repair of the lip follows completion of this arch alignment (usually at three to six months) then McNeil issues a 'stimulation' plate until palatal repair is completed.

Burston (1958), a proponent of this technique, maintains that this moulding provides a firm basis for surgical lip repair as well as for a more normal growth of the nasal septum. McNeil (1964) quotes the investigations of Haupl in 1955. In an histological appraisal of the bony margins of the cleft area, following the use of a stimulation plate, it appeared that bone growth had occurred. Ohlsson (1966) refers to work of Hotz and Graf-Pinthus which suggests that the lack of tongue interposition could produce this result.

There is a paucity of long term studies on the effects of presurgical orthopaedics on both the maxillary growth as well as the development of the dentition. Graf-Pinthus and Bettex (1970) have presented evidence that realignment of the segments prior to surgery up to the age of 2½ years produces a relatively stable result. There is less likelihood of the development of cross-bite in the early mixed dentition.

In the view of Mr. K. Godfrey, University of Sydney, the use of this type of plate is of immediate value as a 'sucking'

plate (1973). During this difficult period of maternal adjustment the benefits of a device to occlude the palatal defect during feeding is very reassuring. The infant can effect a palatal seal and can therefore be easily bottle fed.

Subtelny (1964) however, does make the point that the extended period of presurgical orthopaedic therapy as well as the unrepaired lip can be an added psychologic trauma for the parents.

It would seem that there is a greater use of presurgical measures in Europe. In the United States there has been a deal of controversy. Pruzansky (1964) completely discounted all forms of treatment while Subtelny (op. cit.) presented a more general viewpoint. Rosenstein et. al. (1972) after the original method of Nordin and Johanson (1955) considers the combination of early bone grafting following orthopaedic moulding to be most desirable.

The concept of presurgical orthopaedics is well founded. In the ideal situation it is an acceptable method of ensuring correction of the maxillary segment alignment, easier surgical adaption as well as improved feeding of the infant. Success, however, is dependent on a number of factors; these include the availability of co-ordinated personnel and facilities as well as the comprehension and co-operation of the parents.

(b) Bone grafting procedures

There have been numerous attempts to restore the bony integrity of the cleft area. The original ideas of Nordin and Johanson (1955) and Schmid (1955) aimed at the replacement of the deficient bony tissue by bone-grafting.

It was felt that there could be improvement in the basic stability of the maxilla and this in turn would ensure a better development of the dentition. Cross-bite in the cleft region could be prevented; teeth could be expected to migrate through the grafted bone and there would be a more stable base for the alar cartilages. Robertson (1971).

The concept of 'boneless bone grafting' is even more appealing. Skoog (1965, 1967). This technique involves two layer periosteal flaps which cover a blood clot containing absorbable gauze (Surgicel) and thus fill in the area between the maxillary segments. The contained area of the blood clot reorganises as bone and bridges the cleft. In this way there is no need for a donor site.

Epstein et al. (1970) suggest three groupings of bone-grafting techniques according to the timing of the procedure:

1. Early or Primary grafting, prior to palate closure,
2. Intermediate bone grafting, after the closure of the palate but prior to the eruption of the late secondary dentition (usually from 2 to 9 yrs.) and

3. Delayed or Secondary bone grafting, after the eruption of the secondary dentition, and after orthodontic correction.

Current appraisals of such a relatively new technique (particularly when viewed in terms of human growth) lack true scientific comparisons. Success and failure in terms of jaw relationships at a given stage of skeletal development may indicate an existence of a trend. However, there is a need for long term longitudinal studies before any true correlation of the growth potentials can be made.

The recent work of Jolleys and Robertson (1972) examined the status of early bone-grafting in a controlled study. After five years their results showed that the control group which had no bone grafting was significantly better in terms of mid-facial growth.

A more recent report from the National Institute of Dental Research (1973) notes that primary bone-grafting is seldom practised today as the results have generally been less than desired. The techniques are involved; they are multidisciplinary as well as hazardous.

3. GROWTH IN THE UNREPAIRED CLEFT LIP AND PALATE

In endeavouring to assess the relative success of surgical repair of clefts it would be valuable to have unrepaired clefts at maturity for comparison. The progress of modern society has made the access to such material increasingly more difficult. It is only by observation of the more primitive societies that any evaluation

can be attempted; even then this could be expected at a purely quantitative level.

Mesrre et al. (1960) in observations in Peurto Rico thought that unoperated unilateral clefts had a potential for normal growth, however Innis (1962) after examining the unrepaired clefts of the Dusan tribes of North Borneo felt that there was diminished growth potential in the cleft areas. This growth lack resulted in the alteration of the alveolar segment resulting in cross-bites, lateral open bites and anterior open bites.

Van Limborgh (1964) presented a series of skulls ranging from the foetal, the newborn to the adult. With particular reference to the development of the adult this author examined four cases of complete unilateral clefting. Constant findings of a slight forward maxillary rotation on the unaffected side were noted while the affected side was slightly flattened and retrognathic.

This paucity could allow only very generalised observations. It did suggest however, that growth in cleft cases was a slower process but that the growth potentials were essentially normal. The exception of course, was the cleft area showed no attempt to close spontaneously and was relative in size to degree skull development.

Pitanguy and Franco (1968) examined a series of cleft subjects in South America. Isolation from adequate corrective facilities enabled these investigators to assess both the unrepaired as well as the poorly repaired cases. Using cephalometric techniques

as a method of quantitative evaluation, all non-operated clefted cases had a certain amount of maxillary protrusion which was seen to increase with bilateral clefting. This protrusion decreased with age.

From these references it is obvious that critical evaluation is impossible. 'Over-processing' of skull samples leads to the inherent errors of extrapolation. Conversely, the limited means of accurate recording of primitive human subjects can lead to similar errors. True evaluation of growth is a longitudinal in method; clearly this is an almost impossible proposition.

Thus, our present knowledge of the status of growth in the unoperated cleft subject is based on infrequent and chance observations. Coupe and Subtelny (1960) examined the question of whether clefting produced a deficiency or displacement of tissue. Where clefting affected the hard palate, tissue was deficient; the amount of tissue deficiency and displacement was least in unilateral clefts.

Deficiency and displacement must be related to the development and interdependence of skeletal and soft tissue elements. Atherton (1967) noted that the bones on the cleft sides of fifteen unilateral cleft palate skulls were smaller in width, length and vertical dimensions. He may well assert that bone and cartilage are laid down in an abnormal position and thereby producing an abnormal developmental pattern; this can in fact be expected embryologically.

It would seem more relevant to stress developmental factors in terms of the functional Matrix' concept of Moss (1962). The altered force vectors which affect the developing deformity are merely deviations from the accepted concepts of functional anatomy. Deformation is determined by the degree of counterbalancing required to allow a balanced growth pattern. In simple terms, the degree of mesodermal lack in the embryo dictates the proportionate lack of foetal development which is reflected to maturation. The addition of a constraint, as in surgical repair, redirects the force vectors towards more acceptable norms.

4. FACIAL GROWTH IN REPAIRED UNILATERAL CLEFT LIP AND PALATE

Graber's thesis (1949) was the first scientifically orientated appraisal of the status of facial growth in surgically repaired clefts of that period. It has been maintained by Hagerty and Hill (1963) that there was earlier criticism of the traumatic surgery of Brophy and his followers. This criticism however, was based on a qualitative assessment rather than on the quantitative methods of Graber.

Cephalometric evaluation provided Graber with the evidence that mid-facial growth retardation did occur. It is also noteworthy thai there was developmental trauma in terms of distorted alveoli and obliterated tooth buds.

In summary Graber found:

1. There was deficient maxillary growth, both vertically and antero-posteriorly; mandibular growth was normal.
2. There was increased freeway space; this was interpreted as an attempt by the musculature to approximate vertical jaw relationships towards the norm.
3. Hypertonic activity of the orbicularis oris muscle was associated with excessive retroclination of the lower incisors.
4. Maxillary growth in width is 5/6 completed by the end of the fourth year.

In a later study (1964) Graber again examined the question of cranio-facial growth. Without standardisation of the cleft type and the method of surgery employed, it was shown that major growth was in an antero-posterior direction in the region of the pterygo-maxillary fissure.

Slaughter and Brodie (1949) made parallel assessments of the surgical involvement in growth retardation. They too related this growth inhibition to the degree of surgical interference; without such interference the deformed areas grew at normal rates. A relationship with the integrity of the blood supply was also claimed; this was in keeping with Graber's findings but was refuted by Jolleys (1954). Growth retardation thought the latter, was due more to the constricting influence of the surrounding

fibrous tissue; there was agreement with the concept of minimal surgical trauma.

Although Hagerty and Hill (op. cit.) attempted to question the value of Graber's assertions, their findings could in fact be interpreted as substantiation. They found no gross differences between non-operated and operated cleft palates in an investigation of growth in a series of unilateral cleft lip and palate cases. The standard procedure of surgical repair was the Langenbeck method by a single operator. This in no way replicated the technique of Brophy.

Thus from these earlier investigations into the relationships between growth potential and surgical methods, certain guidelines have become clear. Surgery could be very disfiguring but with newer skills and the avoidance of growth inhibiting procedures these disfigurements could be minimised.

Although these gross maxillary deformations may well be historically recorded sequelae of earlier methods of repair, there are still persistent features directly associated with the facial growth potential and the cleft lip and palate condition. These are the incomplete antero-posterior growth of the maxilla, the mandibular posture and the distortion of the dento-alveolar relationships.

(a) Maxillary growth and mandibular posture

It is now accepted that there is an absence of tissue in the maxillae of cleft lip and palate subjects. Coupe and Subtelney (1960), Huddart et al. (1969), and Robertson (1971). In these unilateral cleft lip and palate cases there appears to be a still greater restriction of the maxillary growth potential. Pitanguy and Franco (1968) examined a number of unoperated facial clefts and noted that the unilateral cleft lip and palate exhibited the greatest amount of maxillary retrusion. This may be directly related to the nature of the cleft; the bilateral case has little to influence the correct alignment of the premaxilla and is in fact in a protrusive position. On the other hand the unilateral condition has a greater amount of musculature which can influence the alignment of the two segments; in this way the unilateral cleft lip and palate case might appear to be relatively retruded.

The repaired unilateral cleft lip and palate was examined in a longitudinal growth survey by Chapman (1966). This was during the six to sixteen year growth period and it was observed that there was a general retrusion of the maxillary complex which progressively worsened with age. Fig. 13.

Harvold (1961) related this retrusion to the bundles of scar tissue which exerted their influence between the hard palate and the pharyngeal muscles. He compared this phenomenon to the similar effect of tight lip musculature on dental retrusion. This concept has been

FIGURE 13. A boy with a complete unilateral cleft lip and palate at five years (A) and at nineteen years (B). The excellent convex pro-file became unfavourably concave with normal mandibular growth but inadequate maxillary growth. (Ross and Johnston, 1972).

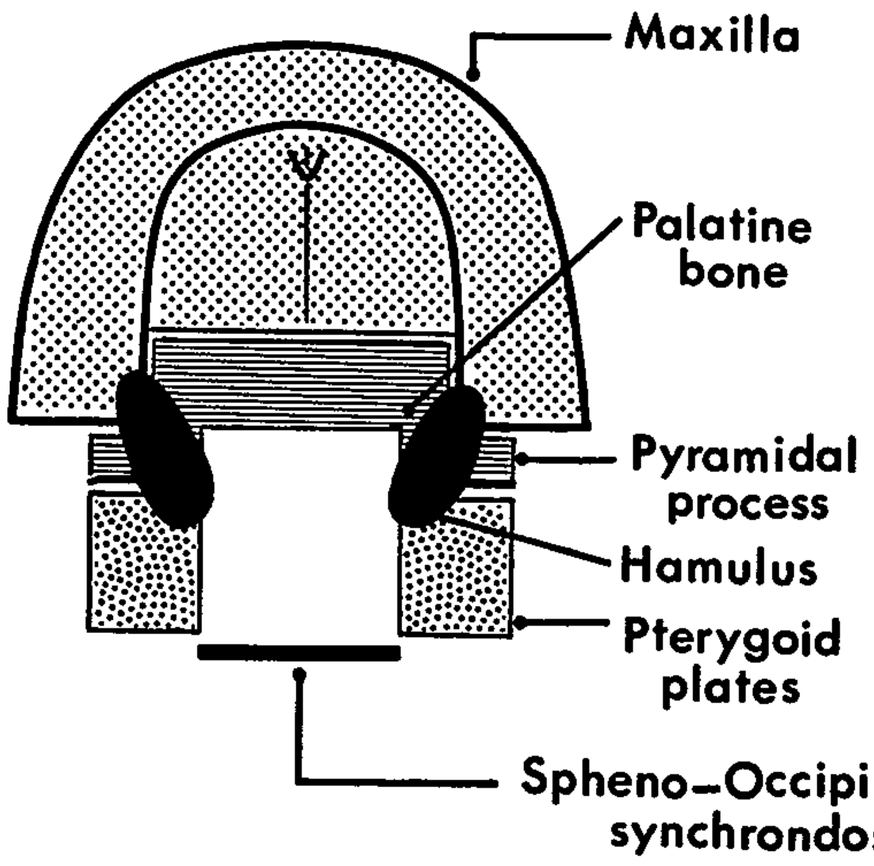
expanded by Ross (1970) and Ross and Johnston (1972) alters the pull of the tensor palati muscles. In such a way a 'continuum of scar tissue joins the maxilla, the palatine bone and the pterygoid plates of the sphenoid. This is described as 'maxillary ankylosis'. Fig. 14.

This band can have a direct influence over maxillary growth be it sutural, or appositional in the tuberosity region. Scott and Symons, 1958; Enlow and Bang, 1965.

Mandibular posture has been extensively examined by Harvold (1960, 1961, 1968, 1973) who noted a downward and backward displacement of the anterior region of the mandible, associated with an increase in the size of the gonial angle. Chapman (op. cit.) also recorded a significantly retruded chin point in his growth study. Harvold related this change in the position of the mandible to the altered tongue position. When the palate is repaired there is a reduction in the maxillary arch size as well as a decrease in the depth of the palatal vault; this is believed to effect a change in the rest position of tongue. Harvold (1968, 1973) has provided experimental evidence on monkeys to demonstrate this inter-relationship between the tongue position and mandibular posturing.

(b) Dento-alveolar relationships

The dental occlusion and alveolar relationships are closely integrated with the growth of the maxilla and the posture of the mandible. Pruzanzky and Aduss (1964) examined the arch form and



Diagrammatic representation of the continuum of scar tissue in a repaired cleft palate which unites the bones of the maxillary complex and pterygoid plates of the sphenoid bone, inducing a mild maxillary ankylosis. This is a fibrous ankylosis, not the true bony type.

FIGURE 14

and the deciduous occlusion of a series of complete unilateral cleft patients. The results indicate that following the repair of the lip there was no maxillary collapse in 11 of the 33 cases. Collapse was noted in 13 of the 33; there was buccal crossbite in 7 cases and buccal with anterior crossbite in 3 cases. If we assume that the realignment of buccal segments is a prime objective, then this was indicated in this survey in only 10 of the 33 cases.

There was also noted that in a sample of 564 non-cleft children 2% exhibited anterior crossbite compared with 3% in the cleft group; 5% had buccal crossbite in the non-cleft group compared with the 30.2% in the experimental group.

It should be pointed out that there is conflicting data on the degree of crossbite in samples of repaired unilateral clefts of the lip and palate. This paper is relevant as it is a longitudinal observation without the intervention of presurgical orthopaedics or bone-grafting techniques.

Dental relationships as distinct from alveolar positioning is another relatively common factor in the realignment of the dentition of the cleft lip and palate patient. Fig. 15 shows the buccal contraction of the permanent dentition. Dey (1973) relates this phenomenon to the surgical trauma to the mucoperiosteum of the alveolar crest.

On the other hand Ross and Johnston (1972) suggest that the distortion of the tooth position is related to the attachment of the supporting fibres (the periodontal ligament) which become

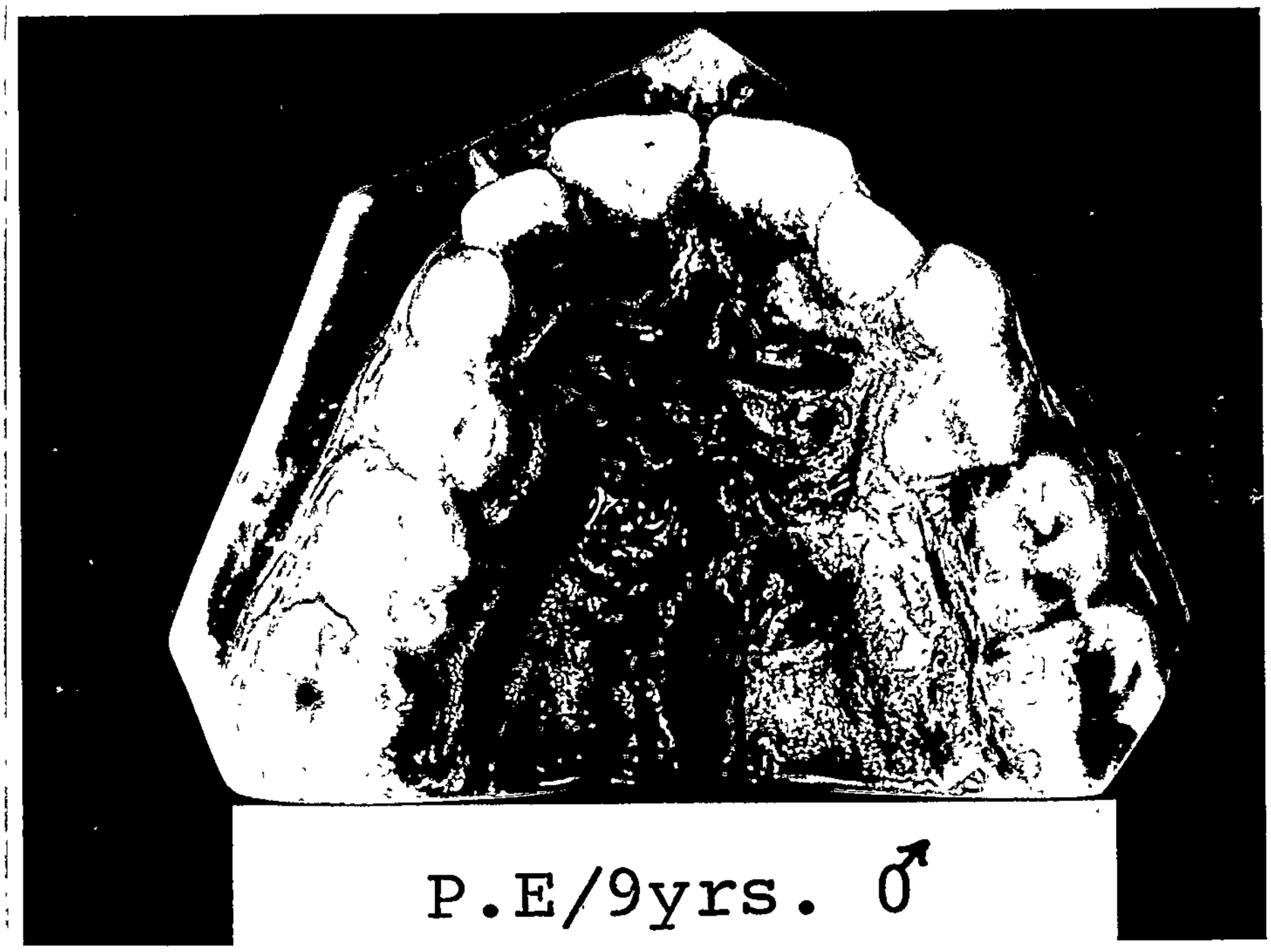


FIGURE 15. Buccal contraction of the permanent dentition following palatal surgery.

included in the scar tissue formation of the involved mucosa. This seems a little anachronistic as the periodontal ligament formation would appear to occur with tooth eruption and not, as suggested, at the time of palatal healing after surgery.

Lip surgery has reached a state of technical excellence; it is in the approach to palatal surgery that greater precision seems necessary if we are to avoid iatrogenic growth disturbances. Scar tissue avoidance is of prime importance if we are to think in terms of improved prognoses. The observations at the Royal Alexandra Hospital for Children by Dey (op. cit.) seem particularly applicable and call to question the between operator variance. Ross and Johnston (op. cit.) place emphasis on the constancy of surgical technique and noted that one surgeon's results were seen to constantly improve.

Thus facial growth, or lack of it, is dependent on the inter-relationship of the tissues of the area be they muscle, bone or tooth structure. To this can be added the concept of function which will be discussed later. One common fact emerges however; there is a basic tissue deficiency. Attempts have been made to reconstruct the absent tissue but it is evident that the results are limited. It would seem logical to harness what nature has left us, treat it as delicately as possible in our attempts to bridge the deficiency and utilise maximum growth potential.

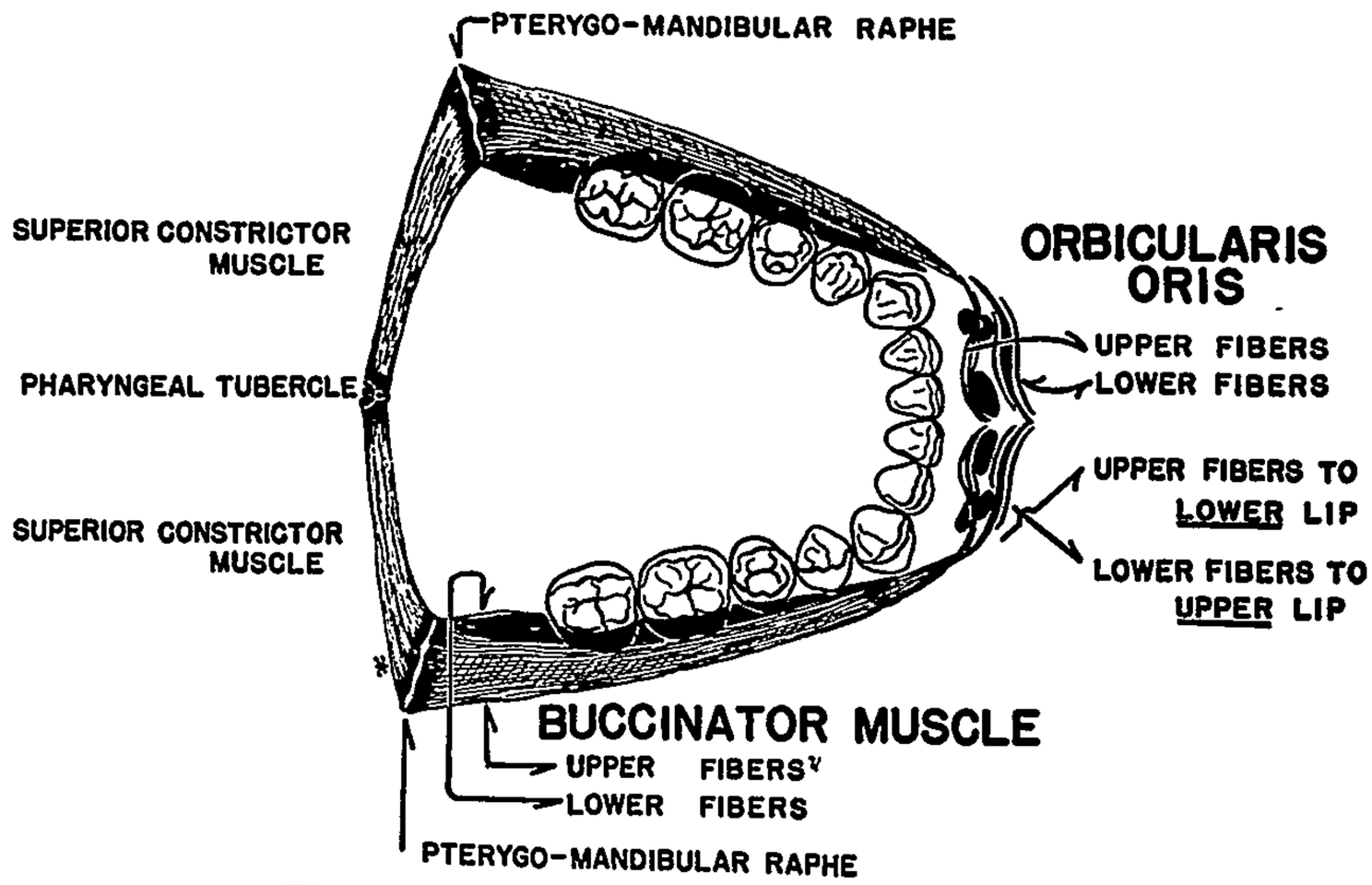
9. IMMEDIATE EFFECTS OF REPAIR ON GROWTH

(a) Lip repair

The repair of the lip musculature establishes the integrity of the 'buccinator mechanism'. Graber (1971) describes this as a system of muscles consisting of the orbicularis oris muscle, the buccinator muscle, the pterygo-mandibular raphe and the superior constrictor muscles arising from the pharyngeal tubercle. This is seen in Fig. 16.

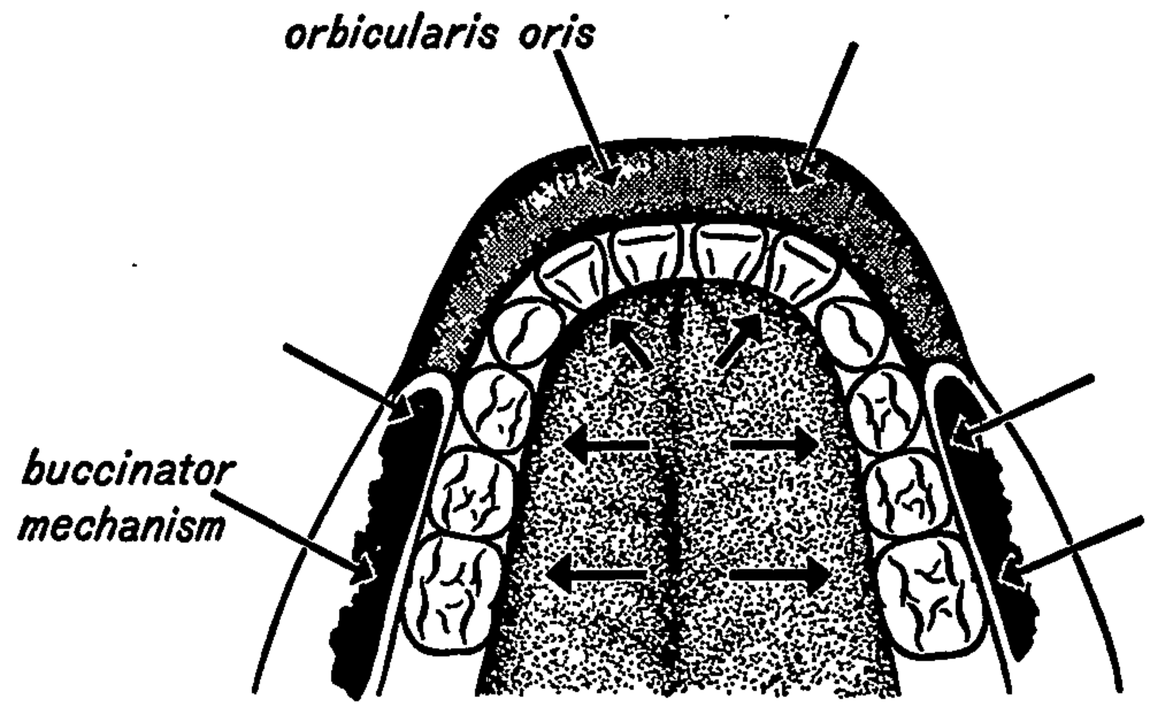
This muscle complex is opposed by the tongue which under normal circumstances provides a state of muscular equilibrium interposed by the developing jaws and teeth. Fig. 17. Thus for balanced growth, the maxilla and the articulating bony relationships are dependent on intergrated muscular balance. There is no pivotal point at the malar bones as Brophy (1923) had inferred; consequently the separation of the tuberosities with coaption of the alveolar segments has been discounted. Fig. 18.

Coupe and Subtelney (1960) found that while hard palate tissue was deficient in cleft patients, it was of a minimal amount in the case of unilateral clefts. It was also evident that the displacement was minimal as well. Therefore it can be expected that lip repair can produce the maximum reduction possible in the width of the cleft by simple redirection of the muscle forces. Aduss and Pruzansky (1968) noted this reduction in the width of the tuberosity, as well as further reduction following the repair of the palate.



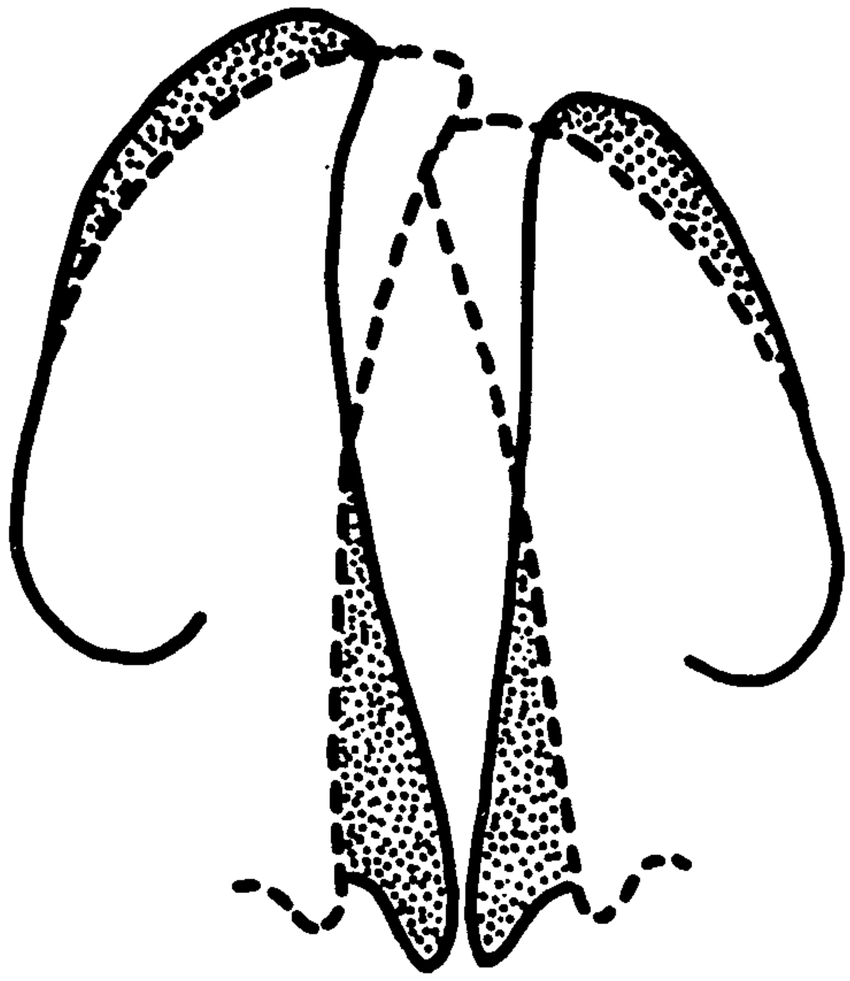
The buccinator mechanism. Note continuous muscle band that encircles the dentition and is anchored at the pharyngeal tubercle. (Modified from J. Jarabak.)

FIGURE 16



The molding pressures on the dental arch, exerted by the contiguous musculature.

FIGURE 17



—— before surgery

--- after surgery

Diagramatic representation of Brophy's concept of the changes in the palate that follow cheiloplasty. (Modification of Brophy's original illustration from, Pruzansky, S.: *Plast. Reconstr. Surg.*, *14:* 10–29, 1954.)

FIGURE 18

Reduction of the cleft width following the repair of the lip was noted qualitatively by Pruzansky (1955). However, Mazaheri et. al. (1967) produced quantitative evidence which supported the view that this reduction was due to growth of the palatal shelves as well as by the moulding effect of the restored intact musculature.

The sphincteric nature of the peri-oral muscles can be seen to have a strong moulding influence on the underlying bony structures. Before repair the force vectors are disorientated and effect a separating movement of the maxillary segments; this activity is augmented by the forward thrusting of the tongue and the laterally superior pull of the septo-premaxillary ligament.

It would seem logical to prevent these distorting forces as soon as possible. This infers the establishment of an intact musculature by surgical repair. Tolerance is required by the neonate and is delayed until the conditions are suitable. At RAHC it is flexible, but as a guide the increasing body weight is about 4.5 to 5 Kg, the Haemoglobin 10 gms. % and the age about 10 weeks.

Following surgery adaptation of the distorted alveolus is rapid. In the report of Pruzansky and Aduss (1964) they noted that there was sufficient tissue present in 80% of cases for complete approximation of the alveolar segments.

(b) Palate repair

The closure of the cleft palate is completed at RAHC around the child's first birthday. For the purposes of this review only the status of growth will be considered. However, in broad terms,

palatal repair involves the occlusion of the hard and soft palate defect. The palatopharyngeal ring of muscle is in this way restored and soft palate movement can be attempted. This area of reconstruction is of key importance in the functions of swallowing and speech. Of more relevance to the orthodontist is the hard palate repair and its relationship to the growth and function of the maxilla.

The surgical techniques in vogue today denude areas of the palate. The closure of the cleft requires that tissue be 'cribbed' from some area; the obvious choice is the adjacent mucosa. The great disadvantage is the resultant scar tissue formation. This was discussed earlier as a related factor in maxillary underdevelopment rather than the concept of Graber involving growth retardation by maxillary ischaemia. Jolleys, 1954; Grundt, 1950.

Jolleys also noted that there was a significant reduction in the soft palate length when the cleft closure was completed after three years of age. It may well have been that the lack of use of the palato-pharyngeal ring of muscle caused the velum to be foreshortened. Perhaps later repair and the consequent increase of scar tissue is related to this poor prognosis.

Scar tissue may, however, have a moderating effect on the initial tendency for the cleft palate to expand excessively. A limited contractile force can reduce such over-expansion to a more acceptable relationship. It must be remembered that any rearrangement of the skeletal structures is in terms of a balance of operating forces.

Aduss and Pruzansky (1968) noted such factors as:

1. the size and shape of the alveolar processes,
2. the size of the palatal shelves,
3. the size and shape of the inferior turbinates, and
4. the size and degree of deflection of the nasal septum.

Such structures are acted upon by functional forces. We could consider these forces to be,

1. the repaired buccinator mechanism,
2. the repaired palato-pharyngeal ring,
3. the functional jaw musculature,
4. the tongue muscle activity, and
5. respiration, and whether it is nasal or oral.

The ideas of Herfert (1963) could well be applied; improvements on the growth potential may well be integrated with improvements in function.

Once the tissue defects have been restored as closely as possible to normally accepted relationships, it would seem obvious that future development could occur within the confines of accepted growth norms. This of course assumes that there is an absolute minimum amount of time lost in restoring the physical environment noted above.

It is on this basis that orthodontic treatment in all its aspects has an important role in the multidisciplinary approach to cleft lip and palate rehabilitation.

PART TWO

This project set out to examine the facial growth status of a group of children who had been treated for unilateral cleft lip and cleft palate. All the patients within this group were treated by the same surgeon who used the same basic surgical methods in each repair.

Initially it was thought that this investigation would be essentially a subjective and qualitative assessment of the amount and direction of growth in the hard and soft facial tissues. Such records as plaster casts, full face and profile photographs, and radiographs of the cleft area allow only a visual appraisal of alteration in the skeletal and integumental tissues. To extend this survey to the level of a quantitative, objective evaluation, cephalometric radiographs were analysed.

Selection of Subjects

All the patients in this investigation were selected from the public and private cases of the surgeon and had been treated in the Royal Alexandra Hospital for Children at Camperdown.

Public patients were located through the Medical Records Section of R.A.H.C., whereas the private group was taken from the personal records of the surgeon. All these children were born between June 1957 and June 1963; they were therefore, in the 10 to 16 year age group and most had full eruption of the permanent dentition.

Whenever possible, suitable appointments were made at the University of Sydney Dental School where the necessary records were

collected. A number of cases were precluded from the survey because of their distance from Sydney.

For the means of comparison, a similar number of children were randomly selected from the files in the University Department of Preventive Dentistry. From a similar age group, all these children had an Angle Class I occlusion, pleasing profiles, and a minimum of crowding of the dentition.

Results of the location of subjects

Of the 15 private cases who were approached,

- 6 took part,
- 2 agreed but delayed their reply beyond the available time limit,
- 1 declined,
- 1 was subnormal and uncooperative and
- 5 failed to contact.

Of the 21 public cases who were approached,

- 9 took part,
- 1 declined,
- 2 were deceased but were unrecorded as such by R.A.H.C.
- 1 was subnormal, and
- 8 failed to contact.

In total there was an initial group of 15 cases available for investigation.

Methods of Recording

Patients and parents were interviewed to explain the investigation. A short history was obtained and the aspect of orthodontic correction was discussed with those parents who appeared unaware of the need for such treatment in their child.

Records were taken:

- Study casts with a wax bite in centric occlusion,
- Photographs of the full face and right and left profiles, and
- Radiographs: Periapical
  Vertex occlusal of the cleft area
  Lateral headplate in centric occlusion.

In the comparative group, records were limited to lateral headplates for the cephalometric analysis.

EXPERIMENTAL METHODS AND OBSERVATIONS

1. Evaluation of the Arch Form and Occlusion:

Recent work by Huddart and Bodenham (1972) attempted to evaluate arch form and occlusion on an objective basis using a scoring method. This appeared to have a low level of consistency and reliability when compared with such descriptive methods as suggested by Pruzansky and Aduss (1964) and more recently by Matthews et al. (1970).

Pruzansky and Aduss (op. cit.) divided the occlusion into six categories:

1. no crossbite present,
2. canine crossbite only,
3. buccal crossbite only,
4. anterior and buccal crossbite,
5. anterior and canine crossbite,
6. incisor crossbite only.

Matthews et al. (op. cit.) on the other hand classified the occlusion as follows:

1. Class A, where all the segments of the maxilla are in normal occlusion with the mandible.
2. Class B (1), the tooth bordering the cleft is in lingual occlusion and requires minor orthodontics to be corrected.
3. Class B (2), normal occlusion of the greater segment but lingual occlusion, which may be only very slight, of the lesser segment.

4. Class B (3), the maxillary arch is perfect but is too small.
5. Class C, these are failures; there is an overall Class III of all segments of the maxilla and in addition there is collapse of some part of the small maxillary arch.

This latter method was used in this investigation. It seemed to classify the various states of the malocclusions in cleft palate cases with a clearer degree of assessment in terms of the total occlusion.

Findings:

Class A	5 patients were found to satisfy the criteria of this category. One of the group had never had any orthodontic treatment.
Class B (1)	2 patients were in this group.
Class B (2)	5 patients were in this group, one of whom had no history of orthodontic treatment.
Class B (3)	None were found to meet the requirements of this group.
Class C	3 patients were found in this group. None had had any form of orthodontic treatment, although one was under observation for future expansion.

2. Photographic evaluation

Full face and left and right profile photographs were taken of each of the fifteen patients. These photographs were subsequently presented to a group of ten people selected from the general population at random; they were asked to comment on the appearance of these cases.

The classifications used in this survey were:

1. Good. Patients in this category were considered difficult to distinguish any difference from an unaffected case.
2. Acceptable. Some difference could be distinguished in these cases but it was insufficient to appear easily noticed.
3. Poor appearance.

Findings

The total scores have been expressed as percentages for each classification.

Full frontal appearance:

Good	59%,	Acceptable	37%,	Poor	4%

Profile appearance:

Good	66%,	Acceptable	28%,	Poor	6%

3. Radiographic evaluation

A. Periapical and vertex-occlusal views of the cleft area.

This method of investigation was designed to provide some information on the integrity of the cleft area. Any formation of a bony 'bridge' across the cleft was noted; an example of this type of union is seen in Figure 19.

Findings

Bone bridging was found in two cases, both of whom were in Class B (2) of the occlusal classification.

FIGURE 19. Bone 'bridge' between the major and minor segments in a unilateral cleft palate.

B. Cephalometric Investigation

This method of investigation was used as an objective evaluation of the growth of the skeletal and integumental profile. Although it was recognised that facial form can be expressed in terms of two planes in space, the frontal planeand the sagittal plane, the latter was selected as more representative of any changes in the facial growth pattern.

Cephalometric radiographs were taken of both the experimental and the comparative groups after the method of Burstone (1958). In summary, patients were positioned as follows:

1. the sagittal plane was at right angles to the path of the x-rays,
2. the teeth were in centric occlusion, and
3. the lips were lightly closed, neither overly relaxed nor tightly closed.

Duplicate tracings were made until all measurements agreed or if they differed, this difference was no more than one unit of measurement. These readings were then averaged.

The number of cases in this cephalometric study was reduced to 13 from the original 15 as the x-ray plates from R.A.H.C. were unsuitably developed. The experimental group was composed of 8 males and 5 females of mean age 12.8 years (SD 1.47). The comparative group, 7 males and 6 females of mean age 12.2 years (SD 1.06).

Landmarks employed

Bony landmarks used in this study were well known points used in cephalometric analysis. They included Sella (S), Nasion (N), Pogonion (P), and Point A. To establish integumental landmarks the line joining sella to nasion, S-N, was extended to intersect the overlying soft tissue at a point which was called 'soft tissue nasion' (NS). The most anterior point of the soft tissue chin was designated 'soft tissue pogonion' (PS), and the point at which the nasal septum between the nostrils merged with the upper cutaneous lip in the midsagittal plane, subnasale (SB).

Reference lines and angles

Any angular and linear evaluation relies on the accuracy of these established points. With the assumption of such points, lines were drawn to form segments and angles that could be related to the components of the profile.

In this study, reference lines were formed between points S and N to establish the cranial base line S-N. To establish the relationships between the maxilla and the mandible to this cranial base line, angles were formed by lines drawn from the relevant reference points. Hard tissue reference angles used in this study were S-N-A and S-N-P. For soft tissue measurements the angle S-NS-PS was used. These angles are shown in Figure 20.

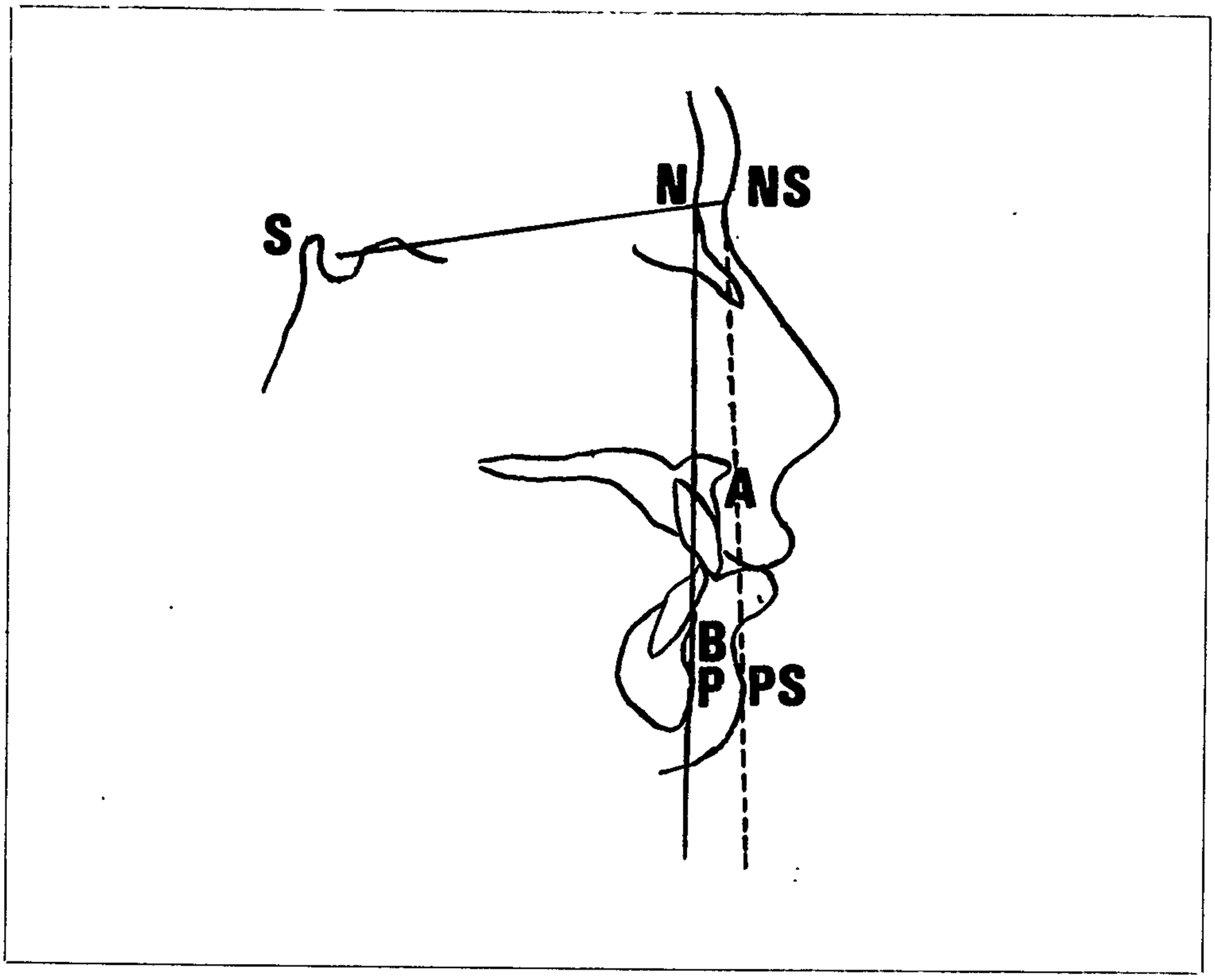


FIGURE 20. Landmarks employed in the cephalometric profile analysis.

As any convexity of the skeletal profile could be inferentially assessed from the angles S-N-A and S-N-P, the convexity of the soft tissues only was determined by a separate measurement. This angle was formed by soft tissue nasion (NS) - the tip of the nose - and soft tissue pogonion (PS); it was designated as 'angle theta' (θ). Figure 21. This was based on the work of Subtelny (1959) who pointed out that the nose has a marked and sometimes direct influence on the total cosmesis of the soft tissue profile.

Linear measurements were taken of the thickness of the upper and lower lips. Upper lip thickness (UL) and lower lip thickness (LL) were measured by the perpendicular distance from N-P of the thickest part of the lips in this plane. Figure 22.

Changes in the total facial height were related to the distance between soft tissue nasion and soft tissue pogonion. This reference line (NS-PS) was intersected by a perpendicular line drawn from subnasale (SB). Two measurements were taken; these were the upper face height (UF) and the lower face height (LF) and expressed as a ratio UF/LF. This was termed the Facial Ratio. Figure 23.

Findings from the cephalometric study

This comparative study was based on the angular and linear values of measurements calculated from the landmarks just described. Both the hard and soft tissue profiles have been related to the mean value for each attribute. The comparison was analysed statistically with Student's 't' test.

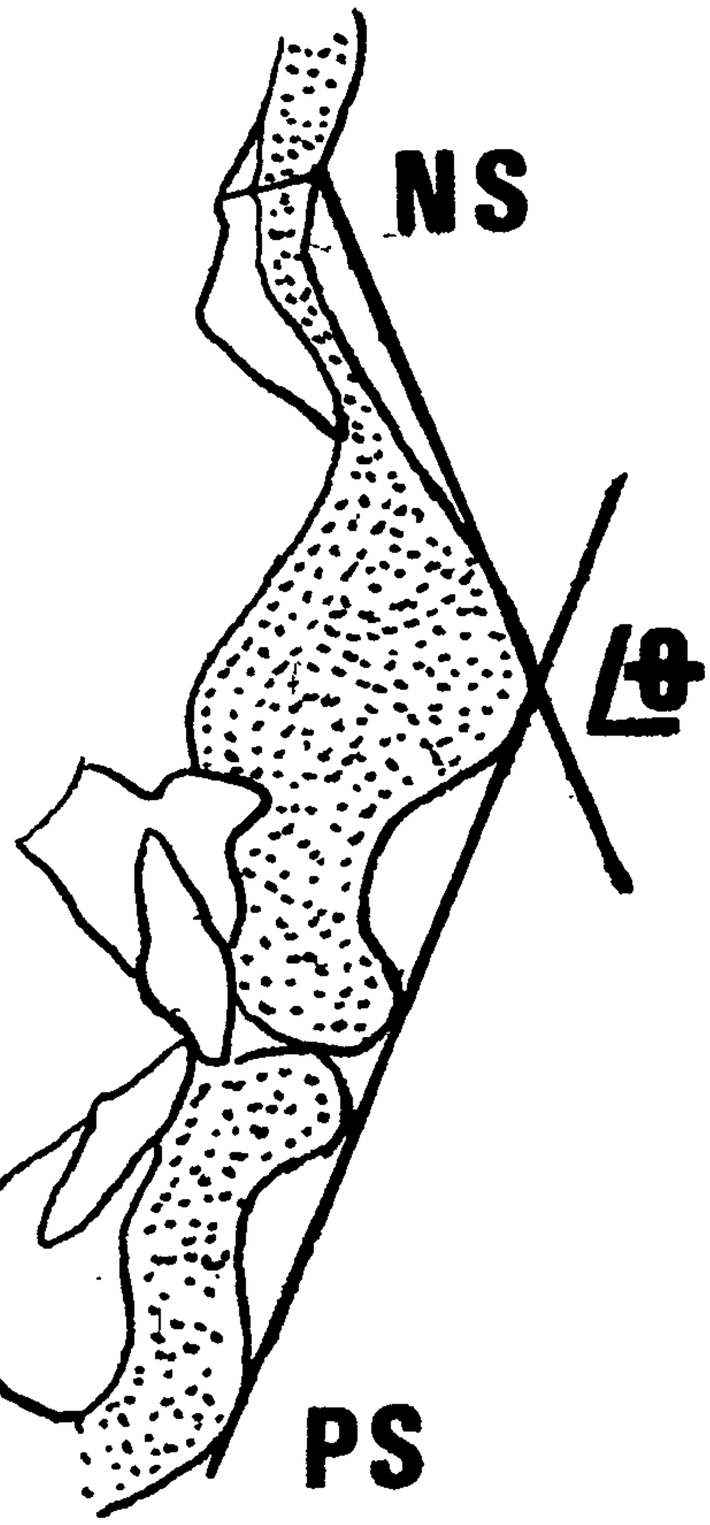


FIGURE 21. Total profile convexity measurement. ($\angle\theta$).

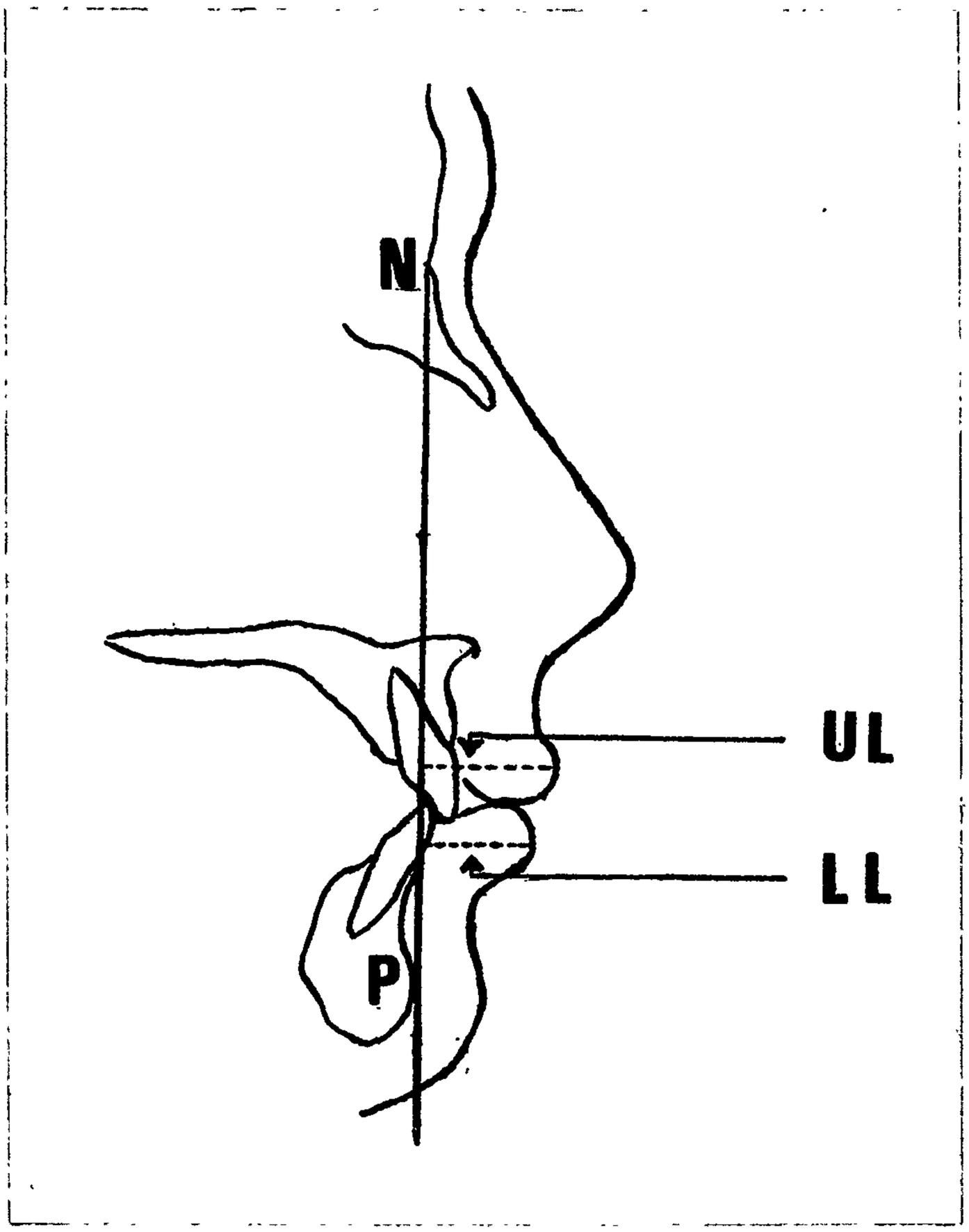


FIGURE 22. Thickness measurement of upper lip (UL) and lower lip (LL).

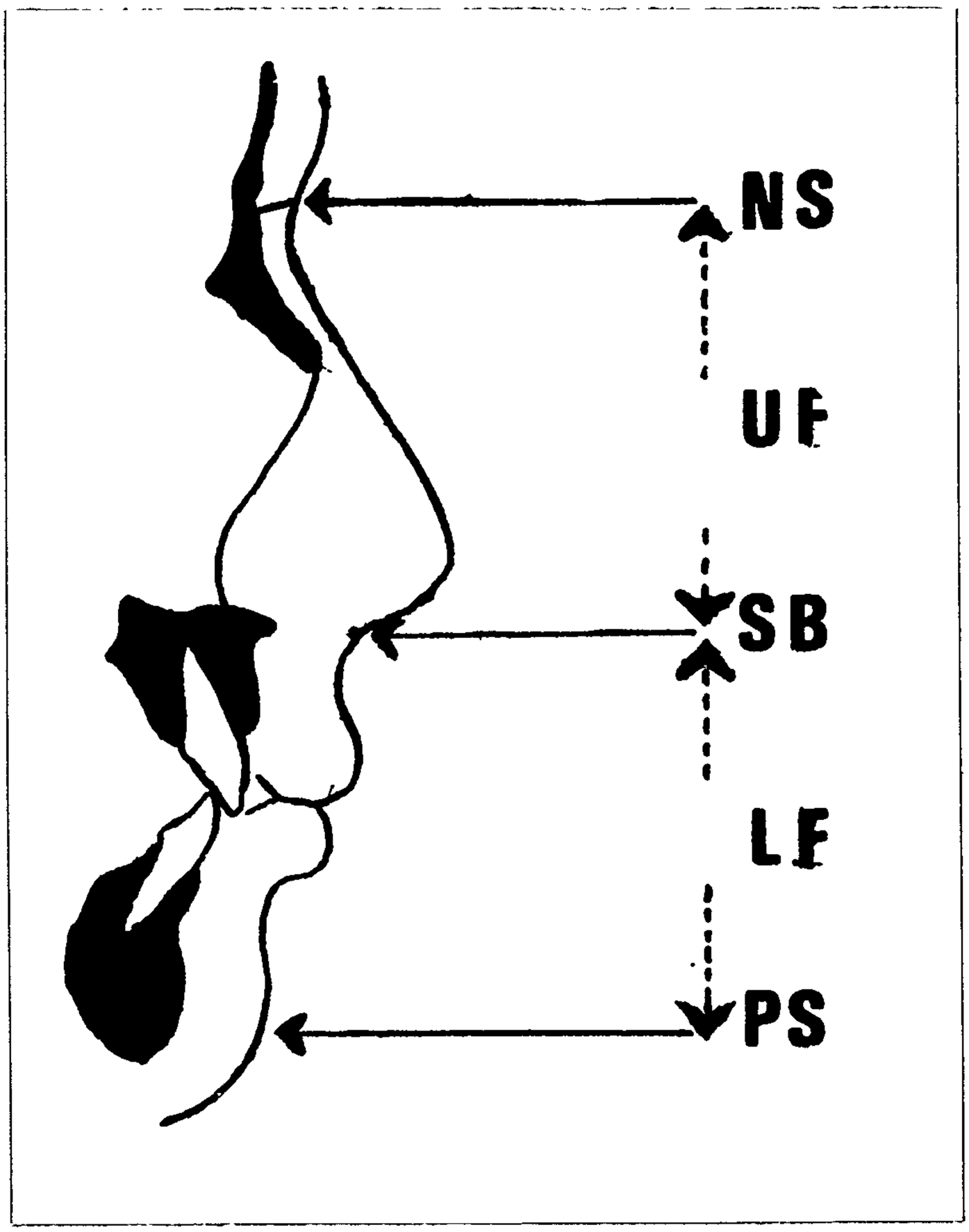


FIGURE 23. Anterior facial height measurement. Upper face height (UF) and lower face height (LF).

Skeletal analysis

S-N-A was found to decrease in the experimental group by

4.4 degrees from 80.2. degrees

P = less than .001

S-N-P was found to decrease in the experimental group by

5.0 degrees from 79.9 degrees

P = less than .001

Integumental analysis

S-NS-PS was found to decrease in the experimental group

by 4.8 degrees from 81.8 degrees.

P = less than .001

Angle θ was found to increase in the experimental group

by 3.1 degrees from 136.7 degrees.

P = approximately .04

UL of the comparative group was 17.4 mm. and 16.8 mm.

in the experimental group.

The difference was not significant statistically.

LL of the comparative group was 15.3 mm. and 16.6 mm. in

the experimental group.

The difference was not significant statistically.

Facial Ratio was 1.13 in the comparative group and

1.08 in the experimental group.

The difference was not significant statistically.

The results of this cephalometric study are tabulated in Figure 24.

FIGURE 24.

	Comparison Group		Experimental Group		
	Mean ($\bar{X}$)	S.D.	Mean ($\bar{X}$)	S.D.	P.
Age (in years)	12.2	1.06	12.8	1.47	
S-N-A	80.2	2.74	75.8	2.24	less than .001
S-N-P	79.9	2.56	74.9	3.83	less than .001
S-NS-PS	81.8	2.64	77.0	4.11	approx. .001
$\angle\theta$	136.7	3.55	139.8	3.72	approx. .04
UL	17.4	3.01	16.8	2.50	approx. .55
LL	15.3	1.90	16.6	2.47	between .1 and .2
Facial Ratio	1.13	0.07	1.08	0.11	between .1 and .2

DISCUSSION

Arch Form and Occlusion

At the particular time of this investigation, twelve of the fifteen cases (80%) had a functional occlusion. A normal occlusion was observed in five cases (33%) and in seven (47%) there was some form of crossbite of the minor segment.

Of the five cases which had not been treated orthodontically, three were found to be in the worst category. These had a total crossbite and collapse of the maxilla. Of the remaining two, one had complete crossbite of the minor segment while the other had a normal interarch relationship.

It could be conjectured that with no orthodontic treatment only one of this group may have had a normal occlusion while the remainder may have been progressively worse. The fact that three from the group of five who had never been exposed to any form of orthodontic treatment needed such attention warrants comment. It would appear that there is a need for more positive interdisciplinary and within-disciplinary referrals.

Photographic evaluation

While such an investigation can be no more than a summary of observations it does indicate a high degree of public acceptance of the appearance of the unilateral cleft lip and palate patients treated in this survey. Although profile views appeared to be more

successfully treated than did the full frontal views, it became evident that observers could have been biased by the full frontal photographs. If profile views alone had been presented, the scores for the 'good' category very likely would have been higher. Of more relevance perhaps, is that only four per cent of the profile scores and six per cent of the full frontal scores were in the 'poor' category.

Apart from the nasal distortion which is not corrected at R.A.H.C. until facial growth is in the main completed, there are many variables in such a photographic evaluation. Such variations as the degree of the initial insult, the general physiognomy and growth potential of the patient, and observer variance ensure that these observations are in no way definitive.

Radiographic Investigation

A. Periapical and vertex-occlusal views of the cleft site.

Little seems to be known about this phenomenon of 'bone bridging'. Whether it appears as an incomplete breakdown of the cleft area or as an attempt by nature to repair the cleft is unknown. However it does seem that the presence of a bony bridge in these cases was in combination with an unerupted tooth, as a supportive measure.

It could be expected that bone bridging might be seen more frequently in cases with a minimum of maxillary arch distortion. Both cases in this investigation showed a total collapse of the minor segment.

B. Cephalometric comparison

As Osborne (1966) points out, the study of a cleft palate individual's adolescent growth can be little more than a study of variables. He included such factors as differences in adolescent growth and the type of treatment carried out to correct the basic defect which in itself can be of varying degrees of severity. To eliminate the surgical variable in this investigation the same surgeon used variations of the Tennison lip procedure and the 'V-Y Pushback' method of palate repair.

In their investigation into the effects of orthodontic treatment on growth, Ross and Johnston (1967) found that treatment in the primary and mixed dentitions had little effect on the basic facial morphology. Noted exceptions were those with very severe maxillary collapse and an altered tongue position due to a lowered tongue posture. In this survey none of the cases were from this category.

Skeletal Analysis

Skeletal assessments of any cleft group appear to form two main groups. Subsequent to Graber's findings (1949) many pursued these in their own institutions. Then, following the revision of rehabilitative procedures and the advent of less growth inhibiting methods of the 1950's there have been further evaluations of the effects on growth. Olin (1971), for example, noted marked

improvement in maxillary retrusion (angle SNA) when he compared groups treated in 1947/1948 with 1956/1958. These however, were composite groups of bilateral and unilateral clefts.

For a complete assessment of growth, the serial method of analysis presents the most comprehensive view. Coccaro and Pruzansky (1965) and Chapman (1966) have examined the skeletal profile in unilateral cleft lip and palate cases. Both these investigations indicated a retrusion of the maxilla and the mandible although Coccaro and Pruzansky related the increase in the facial convexity to the later forward growth of the chin point. Neither of these investigations made any attempt to standardise the initial method of surgical repair.

The results presented in this analysis corroborate these findings. Point A was retruded in the cleft group by 4.4 degrees and the chin point, Pogonion, was retruded by 5.0 degrees which gives support to the concept of a posteriorly rotated mandible. These results were statistically highly significant.

Integumental analysis

Although controversy exists concerning the inter-relationship of soft and hard tissue profiles, Reidel (1957), Burstone (1958), the effect of a 'pleasing profile' is aimed for by all orthodontists. Subtelney (1959) examined the soft tissue profile in a longitudinal study using a method of cephalometric analysis which formed the basis of a later investigation by Coccaro and Pruzansky (op. cit.) into

unilateral clefts. The most recent published investigation was that of Sadowsky, Aduss and Pruzansky (1973). This was longintudinal study with minimal operation variance but a team of twenty-four operators.

Both these latter investigations have been based on the work of Subtelney (op. cit.) and have all measurements based on Basion-Nasion and the extrapolation to Soft Tissue Nasion. In the evaluation of the R.A.H.C. group all measurements are based on Sella-Nasion and extrapolated to another Soft Tissue Nasion point. Any attempt at comparison must be with the data of Sadowsky et al. (op. cit.) since this investigation alone extended to the mature profile.

The mandible

Both the soft and hard tissue profiles of the mandible decreased in the cleft group, 5.0 and 4.8 degrees respectively; these differences were statistically highly significant. Unlike Sadowsky et al. (op. cit.) who noted general mandibular retrusion but a greater degree of protrusion of the soft tissue chin relative to the cranial base when compared with the underlying skeletal chin, the differences in this study appeared parallel.

Total profile convexity

In the experimental group Angle θ was formed by the angle subtended from the tip of the nose to Soft Tissue Nasion and to Soft Tissue Pogonion. In this study of the total profile the cleft

group was 3.1 degrees less convex. This finding concurs with that of Sadowsky et al. (op. cit.). Alar distortion was evident in the full frontal photographs and partly explains the higher degree of 'poor' assessments in the previous subjective evaluation. As was pointed out, none of the cases in this study had been treated for nasal correction; this would have affected this result.

Soft tissue thickness of the lips

Lip thickness in this study was investigated for both the upper and lower lips and were not found to be statistically different from the comparison group. The notion of Harvold (1954) that the tightness of the upper lip inhibited the forward growth of the maxilla does not appear to be fully supported. This finding when related to the photographic assessment is in keeping with the generally favourable response to soft tissue surgical repair. Lips are an important feature of both the profile and full frontal appearance; the fact that they appear such within subjective and objective norms is just comment on the excellence of the surgical methods.

Facial height

The increase in anterior vertical height has been widely observed, particularly in older children. Ross and Johnston (1972) Chapman (op. cit.). If this is related to mandibular posturing whether from a shallower palatal vault suggested by Harvold (op. cit.) or from enforced mouth breathing by nasal obstruction, then

it could be expected that there would be an increase in the lower face height (LF). This would produce an increase in the value of the ratio UF/LF. No significant difference was found between the two groups in this study. It would appear that any posterior positioning of the mandible was insufficient to produce facial elongation.

CONCLUSIONS

This study formed two parts; a subjective as well as an objective assessment of this group of patients has been presented. While every attempt has been made to compare and evaluate, the limitations set by the failures of sample size and the makeup of the comparison group must be recognised.

At the subjective level the great majority of patients in the study had been surgically repaired with a high degree of pleasing aesthetic result. While only two of the fifteen patients had any form of naturally occuring bony support across the cleft and therefore, the possibility of increased maxillary stabilisation, most seemed to have a favourable orthodontic prognosis. The need for more liaison with referring dental, medical and surgical personnel was evidenced by the 20% of the group requiring orthodontic treatment.

The quantitative cephalometric evaluation has supported the findings of others. Both the maxilla and the mandible were found to be retruded in this group of cases; this was reflected in the retruded soft tissue profile of the mandible. The total soft tissue convexity of the profile was seen to be slightly flattened although there was no difference in the thickness of the lips. No difference was observed in the anterior facial height.

Although some of the quantitative findings were at a statistically significant level of difference, these differences were small and

were within the range of generally acceptable skeletal profiles which constitute the norm. The qualitative assessments offered sufficient evidence of the overall success of the surgical techniques applied to this group of patients.

From an orthodontic viewpoint, only one patient had been completely treated and had been fitted with a prosthesis; hence no real assessment could be made.

A later study at the completion of all rehabilitative treatment would be required to enable a full appraisal of these particular patients.

SUMMARY

The literature relevant to the development, aetiology, epidemiology and treatment of the complete unilateral cleft lip and palate was discussed. This was related to the treatment of patients selected for this study.

The findings of the quantitative and qualitative investigations were presented and discussed. In particular, the quantitative findings were reviewed with reference to other studies.

Conclusions were presented.

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