ORTHODONTIC EVALUATION OF OUTCOMES OF SURGICAL CORRECTION
OF COMPLETE UNILATERAL AND BILATERAL CLEFTS
OF PRIMARY AND SECONDARY PALATE

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fulfilment of the requirements for
the degree of Master of Dental Surgery.

Department of Preventive Dentistry, Faculty of Dentistry,
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Table of Contents

List of Figures ........................................ iv
List of Tables ......................................... vi
Introduction .......................................... vii

I. Chapter 1. Pathogenesis of Cleft Lip and Palate ......... 1
   1.1 Classification and Incidence of Cleft Lip and
       Palate ........................................... 1
   1.2 Cleft Lip and Palate Mechanisms ....................... 9
   1.3 Teratological Studies on Mechanisms of Cleft
       Formation ........................................ 16

II. Chapter 2. The Craniofacial Complex in Cleft Lip
       and Palate ....................................... 22
   2.1 The Maxilla ..................................... 22
   2.2 The Mandible .................................... 30
   2.3 Facial Proportions and Growth ....................... 32
   2.4 The Cranial Base ................................ 33
   2.5 The Nasal Septum and Nasal Airway ................. 34

III. Chapter 3. Presurgical Orthopedics in Cleft Lip
        and Palate ..................................... 36

IV. Chapter 4. Surgical Repair of the Cleft Lip ............. 47

V. Chapter 5. Effects of Lip and Anterior Palate
       Surgery ......................................... 64

VI. Chapter 6. Primary and Secondary Bone Grafting
       in Cleft Lip and Palate .......................... 77

VII. Chapter 7. Repair of the Cleft of Secondary Palate ... 87

VIII. Chapter 8. Effects of Cleft Palate Surgery ........... 95

IX. Chapter 9. Occlusion in Cleft Lip and Palate Patients 105
Summary and Conclusion .......... 129
Bibliography .................... 131
Acknowledgements ................ 145
List of Figures

Figure 1. The Kernahan and Stark Classification of 1955 ........................................ 4-5
Figure 2. Diagrammatic Representation of Stark's Theory of Pathogenesis of Harelip .......... 11
Figure 3. Scanning Electron Micrograph of Palatal Fusion ........................................... 14
Figure 4. Interaction of Genetic and Environmental Influences on Palatal Closure .......... 18
Figure 5. Diagrammatic Representation of Normal and Bilateral Cleft Naso-premaxillary Region ... 29
Figure 6. Presurgical Orthopedic Appliance as Used at Westmead Centre ......................... 44
Figure 7. Appliance in Situ ................................................................................................. 45
Figure 8. Serial Casts Showing Degree of Septal Straightening Achieved ......................... 46
Figure 9. Hofmann's Headcap ............................................................................................. 48
Figure 10. A Surgical Table from the Early Eighteenth Century .................................... 49
Figure 11. The Mirault Operation ......................................................................................... 51
Figure 12. Hagedorn's Operation ......................................................................................... 51
Figure 13. The Le Mesurier Repair .................................................................................... 52
Figure 14A. The Tennison Repair ...................................................................................... 53
Figure 14B. The Tennison Repair ...................................................................................... 54
Figure 14C. The Tennison Repair ...................................................................................... 55
Figure 15. The Millard Rotation Advancement Flap .............................................................. 56
Figure 16. Brophy's Technique for Moving the Premaxilla Back ...................................... 59
Figure 17. Surgical Removal of Part of the Nasal Septum .................................................. 60
Figure 18. The Blair and Brown Technique for Bilateral Cleft Lip Repair ....................... 61
Figure 19. The Millard Forked Flap ..................................................................................... 62
Figure 20. The Buccinator Mechanism .................. 65
Figure 21. The Decussating Fibres of the Orbicularis
Muscle ............................................. 66
Figure 22. Lip Pressure Profiles .......................... 68
Figure 23. The Muir Flap for Anterior Palate Repair .... 75
Figure 24. Early Bone Grafting ........................... 79
Figure 25. Radiographs Preoperative and Postoperative
Bone Grafting ....................................... 79
Figure 26. Skoog's Technique of "Boneless" Bone Grafting 82
Figure 27. Veau's Operation .............................. 89
Figure 28. Wardill's V-Y Palatoplasty ...................... 90
Figure 29. Dunn's Procedure for Using Vomer Flaps
for Palate Repair ..................................... 91
Figure 30. Vomer Flap Covered with an Autogenous Skin
Graft for Hard Palate Closure ........................ 92
Figure 31. Schematic Diagram of Possible Effect
of Postoperative Scar ................................ 96
Figure 32. Series of Cases Operated upon by the Delayed
Hard Palate Closure Technique ....................... 103
Figure 33. Tongue-Lip-Teeth Relations at Rest .......... 106
Figure 34. Photooopy of Maxillary Cast ................... 109
Figure 35. Arrangement for Stereophotographs .......... 110
Figure 36. Stereophotogrammetric Tracings of Palatal
Casts ............................................... 111
Figure 37. A Parabolic Curve Fit for Complete Unilateral
Cleft Lip and Palate .................................. 113
Figure 38. Arch Form .................................... 116
Figure 39. Segmental Divisions for the Numerical
Classification ...................................... 117
Figure 40. Canine Scoring ................................ 117
Figure 41. Molar Scoring ................................ 118
Figure 42. Incisor Scoring ................................. 118
List of Tables

Table 1. Incidence of Cleft Lip and Cleft Palate in Different Regions of the World 6

Table 2. Distribution of Cleft Cases by Sex and Cleft Type, (1964-1966, New South Wales) 7

Table 3. Observed and Expected Number of Cleft Cases by Sex, (1964-1966, New South Wales) 7

Table 4. Distribution of Cleft Cases by Sex and Cleft Type, (1967-1971, Royal Alexandra Hospital for Children) 8
Introduction

Clefts of the lip, alveolus and palate along with the nasal deformity represent a lack of normal development of this area of the face. The nature of the problem with which the clinician is faced is the growth and function of these defective or anomalous oro-facial structures and the aims and objectives of their correction to provide the best possible overall result.

Whether or not a child born with a cleft lip and palate has a chance of leading a normal life may depend entirely on the quality of the treatment provided. We must, therefore, examine the aims of treatment which are so easily stated yet not so easily achieved. They are to produce normal appearance, normal speech and normal dental occlusion. In order to optimize these results I believe it is necessary to concentrate the expertise of the surgeons, orthodontists and speech therapists under the same roof, seeing their cases together in follow-up clinics to allow for constant evaluation.

Chapter 2 looks at the craniofacial complex in cleft lip and palate with sections on the maxilla, mandible, facial proportions and growth, the cranial base, and nasal septum and airway. Chapter 3 examines the use of presurgical orthopedics as an associated rehabilitative procedure. Chapters 4 and 5 review the state of the art of lip and anterior palate repair and its effects.
Chapter 6 reviews the literature concerned with primary and secondary bone grafting as a part of the rehabilitative procedure, while chapters 7 and 8 deal with palatal surgery and its effects. Chapter 9 considers occlusion in cleft lip and palate.
CHAPTER 1: The Pathogenesis of Cleft Lip and Cleft Palate

1.1 Classification and Incidence of Cleft Lip and Palate

Malformations of the craniofacial complex abound with great variation whether they include cleft lip and palate or not. These malformations have been categorized by two means:

1. pathogenic mechanisms
2. anatomical conditions

Confusion has frequently arisen because the names of syndromes, for example, Crouzon and Treacher-Collins, do not fulfill the criteria necessary for either basic scientists or the clinician. The clinician requires simplicity combined with clinical applicability in a broadly based system which should, ideally, combine treatment goals and principles. The scientist, on the other hand, desires an embryologic or biochemical classification.

Of all facial deformities, classification of cleft lip and palate has received the most attention. Veau (1931) based his classification upon operative anatomical findings. Advances in understanding the embryology of cleft lip and palate have laid the foundations for the present classifications in use. These are:


The Kernahan and Stark classification is based on embryological studies in which they examined the criteria for classifying anomalies of the lip and palate. The so-called "primitive" palate includes the prolabium, premaxilla and primary septum, whereas the "definitive" or "secondary" palate makes up the hard and soft palate. According to Stark and Ehrmann (1958), the premaxilla of the adult develops from the primitive palate, a lip structure which is delimited by the nasal airways above. Because it is a lip structure, the premaxilla may be surrounded by a circumferential cleft in a complete bilateral cleft lip without a cleft palate. At this stage, the palatal shelves which will form the "definitive" or "secondary" hard and soft palates are rudimentary.

The fate of the premaxillary bone has long been a point of contention. Wood-Jones (1937) stated that the absence of the premaxilla in the human face was "a human specific character" because in early embryonic life the maxilla of each side grows forwards over the anlage of the premaxillae meeting in the midline of the upper jaw. Observations of Noback and Moss (1953),
quoted in Stark and Ehrmann (1958), indicate however, that the premaxillary bone fuses with the palatal shelves of the maxillary bones at the incisive suture. These investigators felt that the definitive premaxillary bone is the same in the adult as in the foetus.

According to the Kernahan and Stark classification, as palatal fusion occurs from the incisive foramen posteriorly, a palate that is cleft as far anteriorly as the incisive foramen represents a complete cleft of the palate. Involvement of the alveolus occurs only with a coexisting cleft of the lip. An alveolar cleft is a part of the lip anomaly and as such is not part of the cleft of the palate. These concepts will be dealt with in more detail in the following section.

Whitaker, Pashayan and Reichman (1981) have proposed a new classification for craniofacial anomalies based on the use of a clock face analogy as proposed by Tessier (1976). The point of reference for this system is the orbit, with clefts being found in two different hemispheres. Those of the lower lid region are facial clefts, while those of the upper lid are cranial. Thus clefts 0 through 4 which have downward extensions to involve the maxilla, fit into the usual cleft lip and palate classifications. Although this classification was produced for the American Cleft Palate Association, it seems that the Kernahan and Stark method, based on the incisive foramen, remains the most popular at the moment.
Figure 1. The Kernahan and Stark (1955) Classification

Unilateral, subtotal cleft of primary palate

Unilateral, total cleft of primary palate

Bilateral, total cleft of primary palate

Subtotal cleft of secondary palate
Total cleft of secondary palate

Unilateral subtotal clefts of primary and secondary palates

Unilateral, total cleft of primary and secondary palate

Bilateral, total cleft of primary and secondary palate
Chi (1974) analysed available birth data from hospital records in New South Wales for the period from 1964 to 1966. His findings revealed a ratio of one affected baby per 766 births or 1.3 per 1000 births. However, Chi encountered considerable difficulty in trying to classify the cases into the various groups due to the inaccuracy and ambiguity of information supplied by the hospitals. This highlights the need for maintenance of a central register of birth defects in New South Wales. See tables 1-4.

### Table 1

<table>
<thead>
<tr>
<th>Author</th>
<th>Location</th>
<th>Period</th>
<th>Source</th>
<th>Number of cases</th>
<th>Sample size</th>
<th>Cases per 1,000 births</th>
<th>Ratio of cases to births</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rank and Thomson</td>
<td>Tasmania</td>
<td>1945-1957</td>
<td>Multiple sources</td>
<td>160</td>
<td>96.510</td>
<td>1:66</td>
<td>1:603</td>
</tr>
<tr>
<td>Fugl - Andersen</td>
<td>Copenhagen</td>
<td>1938-1942</td>
<td>Hospital records—live births</td>
<td>478</td>
<td>364.764</td>
<td>1:31</td>
<td>1:763</td>
</tr>
<tr>
<td>Moller</td>
<td>Iceland</td>
<td>1956-1962</td>
<td>Multiple sources</td>
<td>64</td>
<td>32.979</td>
<td>1:94</td>
<td>1:573</td>
</tr>
<tr>
<td>Greene, Verrill, Huy, Gibbons, Kerschbaum</td>
<td>California</td>
<td>1956-1960</td>
<td>Birth certificates</td>
<td>2,185</td>
<td>1,765.746</td>
<td>1:24</td>
<td>1:806</td>
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<tr>
<td></td>
<td>Wisconsin</td>
<td>1956-1960</td>
<td>Birth certificates</td>
<td>1,446</td>
<td>1,242.408</td>
<td>1:16</td>
<td>1:859</td>
</tr>
<tr>
<td></td>
<td>Birmingham</td>
<td>1940-1950</td>
<td>Multiple sources</td>
<td>692</td>
<td>485.104</td>
<td>1:43</td>
<td>1:701</td>
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<td></td>
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<td>285</td>
<td>218.693</td>
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<tr>
<td>Gilmore and Hofman</td>
<td>Wisconsin</td>
<td>1943-1962</td>
<td>Birth certificates</td>
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<td>1,755.044</td>
<td>1:23</td>
<td>1:87</td>
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<tr>
<td>Fugl - Andersen</td>
<td>Denmark</td>
<td>1953-1957</td>
<td>Hospital records</td>
<td>644</td>
<td>393.457</td>
<td>1:64</td>
<td>1:68</td>
</tr>
<tr>
<td>Hixon</td>
<td>Ontario</td>
<td>1943-1949</td>
<td>Hospital records</td>
<td>695</td>
<td>655.322</td>
<td>1:46</td>
<td>1:72</td>
</tr>
</tbody>
</table>

### Table 2

Distribution of cleft cases by sex and cleft type (1964–1966, N.S.W.)

<table>
<thead>
<tr>
<th>Type of cleft</th>
<th>Male</th>
<th>Female</th>
<th>All Cases</th>
<th>Percentage of series</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip</td>
<td>27</td>
<td>19</td>
<td>46</td>
<td>24.0</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>26</td>
<td>30</td>
<td>56</td>
<td>29.1</td>
</tr>
<tr>
<td>Cleft lip with cleft palate</td>
<td>58</td>
<td>32</td>
<td>90</td>
<td>46.9</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>111</td>
<td>81</td>
<td>192</td>
<td>100.0</td>
</tr>
</tbody>
</table>


### Table 3

Observed and expected number of cleft cases by sex (1964–1966, N.S.W.)

<table>
<thead>
<tr>
<th></th>
<th>Cleft lip</th>
<th>Cleft palate</th>
<th>Cleft lip with cleft palate</th>
<th>Cleft lip (palate)</th>
<th>All cases</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Observed</td>
<td>Expected</td>
<td>Observed</td>
<td>Expected</td>
<td>Observed</td>
</tr>
<tr>
<td>Male</td>
<td>27</td>
<td>24</td>
<td>28</td>
<td>27</td>
<td>58</td>
</tr>
<tr>
<td>Female</td>
<td>19</td>
<td>22</td>
<td>30</td>
<td>27</td>
<td>32</td>
</tr>
<tr>
<td>Total</td>
<td>46</td>
<td>56</td>
<td>58</td>
<td>46</td>
<td>90</td>
</tr>
</tbody>
</table>

\[ \chi^2 \]

\[ 0.7840 \quad 0.6436 \quad 6.4031^* \quad 6.6232^* \quad 3.0028 \]

* Statistically Significant at 0.005 level.

### Table 4

**Distribution of cleft cases by sex and cleft type (1967–1971, Royal Alexandra Hospital for Children)**

<table>
<thead>
<tr>
<th>Type of cleft</th>
<th>Male</th>
<th>Female</th>
<th>All Cases</th>
<th>Percentage of series</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip</td>
<td>35</td>
<td>33</td>
<td>68</td>
<td>28.8</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>36</td>
<td>34</td>
<td>70</td>
<td>29.7</td>
</tr>
<tr>
<td>Cleft lip with cleft palate</td>
<td>66</td>
<td>32</td>
<td>98</td>
<td>41.5</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>137</td>
<td>99</td>
<td>236</td>
<td>100.0</td>
</tr>
</tbody>
</table>


Brogan (1978) found a significant decline in the total incidence of cleft lip and palate combinations in Western Australia during the period from 1958 to 1975. An hypothesis was presented that these changes are linked to the changing fertility pattern throughout Australia, as the birthrate drops towards zero population growth.

Heyes (1980) surveyed cases of cleft lip and palate born on the Merseyside in England. While the total number of births in this area had been declining over a ten year period, the incidence of clefts actually increased. These results were thought to be due to increasing marriageability of affected adults through better rehabilitative procedures as well as an increase in infant survival rate.
Gregg, Zimmerman, Clifford and Gregg (1981) studied craniofacial anomalies in the Upper Missouri River Basin region of the United States using archeological and clinical evidence. The present frequency of cleft lip and palate anomalies in native Americans was found to be approximately 1:300 live births, considerably greater than the general population of the United States. Interestingly, no evidence of clefting in ancient times was found, in spite of an apparent favourable situation for perpetuation of faulty genes with supposed inbreeding. These authors surmised that this could be due to the practice of elimination of unfit or unwanted infants from the family, which still occurs in this area.

1.2 Cleft Lip and Palate Mechanism

The "classical" theory of normal development of the face separates formation of the "primitive palate" (prolabium, premaxilla, and primary septum) from that of the hard and soft palates. According to Dursy (1869) and His (1892), as quoted by Stark (1954), the face cephalad to the oral cavity is subdivided into numerous peninsular masses of ectoderm and mesoderm surrounded by free spaces or clefts. These are the paired maxillary processes laterally and the unpaired frontal process. It is maintained that these processes grow, meet and fuse in a manner similar to the healing of a wound. Clefting of the primary palate was thought to result from simple failure of the fusion process.
In the early part of this century Pohlmann (1910), as quoted by Stark (1954), raised doubts about the existence of these processes. Coincidental with this challenge came a newer theory of mesodermal penetration, which concerns itself with the primitive or prepalate only. According to this theory the prepalate or primary palate exists in its early form as an epithelial wall in which three masses of mesoderm are normally present which grow and fuse. No clefts or processes exist in the central third of the face. Stark (1954) suggested that if any of these masses of mesoderm is absent, or fails to develop adequately, a cleft will develop at that site. He relates this rupture effect to the occasional presence of Simonart's bands across the cleft of the lip, which are suggestive of a remnant resulting from traction or pulling apart of a region, and to the lack of lateral incisors, the development of which is also influenced by mesoderm.

Clefting of the secondary palate or definitive palate which includes the hard and soft palates, is generally thought to be due to a lack of contact between the palatal shelves which normally meet and fuse at about forty-seven days in utero. The mechanism, or mechanisms, which bring the palatal shelves into a horizontal position prior to fusion are not clear. However, significant progress has been made towards understanding what happens during the contact and fusion phases of palatal closure. Burdi (1977) identifies two consecutive events which must occur prior to successful palatal shelf fusion:
1) Adhesion of the palatal shelves to form the so-called epithelial wall or seam.

2) Subsequent disintegration of this seam.

Figure 2. Stark's theory of the pathogenesis of harelip is presented diagrammatically. The three mesodermal volumes located in the epithelial wall are shown. The absence of a lateral volume will result in a unilateral cleft lip. Absence of the medial volume will result in a median cleft, a rare anomaly and absence of the two lateral volumes will cause a bilateral cleft lip to occur. From Stark (1954).
Portois (1970) showed adhesion of rat palatal shelves to form a seam depends on an increase in adhesiveness of the surface epithelium at the edges of the converging shelves brought about by an increase in the glycoprotein layer at their surface. After incubating the palatal shelves in periodic acid, Portois hypothesized that failure of seam disintegration may occur by two different mechanisms:

1) Toxic action of the periodic acid may prevent the normal physiological state of tension that develops in the seam.

2) Some alteration in the mesenchyme might impair the differentiation or functioning of the histiocytes which may be instrumental in seam lysis.

Caution must be exercised in interpretation of this data as they result from an in vitro experiment in rats.

Mato, Smiley and Dixon (1972) showed that the plasma membrane of human superficial epithelial cells increased in density with a "specific coating" observed immediately before contact of the shelves. Pratt, Goggins, Wilk and King (1973) demonstrated an increase in acid mucopolysaccharides present in the palate at the time of shelf rotation and fusion, while De Paola, Drummond and Miller (1975) noted a large increase in concentration of glycoproteins on the free medial epithelial surfaces prior to contact and adherence.
Whilst these palatal shelf surface phenomena are no doubt important, attention has also been focused elsewhere. Mato, Smiley, and Dixon (1972) found specific epithelial changes in the presumptive regions of fusion both before and during secondary palate formation in humans. These changes were found in the mitochondria, the epithelial cell arrangement, and the appearance of lysosomes. There were also signs of cell degeneration at the medial edge of the palatal processes and at the lower surface of the nasal septum before fusion. Smiley and Koch (1972) corroborated these observations. Their results indicated that epithelial cell-death, and hence "micro-disruption", are normal events in mouse embryos and therefore, cellular contact between palatal processes is not a prerequisite for midline epithelial disruption.

Many factors must be considered in interpretation of developmental changes in palatal processes grown in vivo, as the biological responses could be varied by changes in experimental parameters. Goss (1975) continued to investigate possible differences between rat in vitro and human in vivo research and succeeded in producing a cleft palate in the rat by rupture of an intact palate which later healed normally by epithelial covering of exposed mesenchyme. Subsequently, Goss and Avery (1975) suggested surface contact of the two epithelial surfaces is essential for fusion as complete failure occurs when shelf epithelium is placed in direct contact with lateral shelf mesenchyme.
Waterman, Ross and Meller (1973) examined the surfaces of the palatal shelves in the A/Jax mouse embryo with a scanning electron microscope prior to and during palatal fusion. Changes were noted in the surface cells along the future medial edge of the area of presumptive fusion. The fact that these changes both preceded closure and were restricted to the region of subsequent contact and fusion between the shelves probably indicates they are a prerequisite for normal palatal fusion and represent an expression of acquisition of fusion potential. See figure 3.

Figure 3. Drawings of palatal shelves from specimens at critical stages examined by Waterman, Ross and Meller, (1973). The distribution of observed surface alterations at each stage is indicated by heavy black shading along the medial edges of the shelves. Redrawn from Waterman, Ross and Meller, (1973).
Interest has also centered on the spatial arrangement of structures in the oral cavity at the time of palatal closure. Ross and Lindsay (1965) demonstrated a relationship between the vertebral column in the cervical region and facial structures. In the eighth or ninth week in utero, the embryonic tongue lies between the vertically oriented palatal shelves. Shortly prior to secondary palate formation, the head is lifted from the pericardial region allowing the mandible and tongue to drop, permitting the palatal shelves to rotate, meet, and fuse in the midline. Ross and Lindsay maintain that lengthening of the neck is particularly important to this change in head position, and whenever the cervical vertebrae are grossly abnormal, the neck remains short and the mandible compressed with the tongue caught between the palatal shelves during the time palatal closure occurs, thereby preventing normal palatal shelf rotation and leading to a cleft of secondary palate. Postwillo (1966) produced cleft palate in rats following puncture of the amniotic sac probably by causing an alteration in fluid pressure resulting in a similar mechanical change in posture.

Nanda (1970) observed a delay in palatal process transportation in a condition of hypervitaminosis A, possibly as a result of maxillo-mandibular ankylosis caused by heterotropic cartilage leading to partial or incomplete mobility of the mandible. He has described this condition as "first branchial arch syndrome" which in many ways is comparable to the Pierre Robin Syndrome in humans with its cleft of secondary palate and characteristic small mandible.
Wragg, Smith and Borden (1972), and Wragg, Dieuwert and Klein (1972) suggested that shelf closure is initiated by tongue contraction, whilst the dorsal movement of the shelf tissues creates room for the widening tongue. The foetal rat myoneural apparatus is functional at the time of palatal closure with the genioglossus, hyoglossus and transversus muscles narrowing to depress the arched tongue prior to closure. These authors question the requirement put forward by other authors for the jaw to drop to allow the tongue to be removed from between the separated palatal shelves, as the tongue did not appear to descend, but merely to flatten. No space formation whatsoever, nor external muscular activity, seemed essential to closure.

Greene and Kochar (1973) studied cortisone-induced cleft palate in mice, observing a delay of as much as twelve to fourteen hours in shelf rotation. When the shelves finally reached the horizontal, they contacted the nasal septum and each other in a normal fashion. However, normal fusion did not occur, possibly due to some intracellular influence on the midline epithelial cells which prevented either adhesion, epithelial breakdown or both.

1.3 Teratological Studies on Mechanisms of Cleft Formation

While current efforts to explain normal and abnormal dentofacial development are targeted toward man, there are obvious limitations on the use of human material for experimentation. This has forced researchers toward appropriate animal types whose early
Dentofacial development is thought to be in compliance with the multifactorial model and its genetic-environmental interplay. Fraser, Walker and Trasler (1957) emphasised this concept of a developmental defect resulting from a genetic and environmental background. Finding a genetic influence, however, is only helpful in so far as it defines the problem a little more clearly. What we really need to know is what the genes are doing to bring about this reaction. Questions still remain on the actual similarities in development between mammals. However, studies continue in which normal fusion is disrupted using teratogens such as Lithium (Loeby, 1973), tranquilizers (Walker and Patterson, 1974), and cortisone (Hackman and Brown, 1972), (Spain, Kisieleski and Wood, 1975) and (Shah and Travill, 1976).

Recently Brogan (1980) noted an increased incidence of cleft lip and palate in country regions of Western Australia, as well as an unexpected seasonal variation. An association with the agricultural use of herbicides and insecticides seems possible, although this is by no means proven at this stage. Goldman, Herold, and Piddington (1981) found cortisol inhibition of lysosomal-mediated cell death on the medial edge of palatal epithelium in an in vitro mouse model, possibly brought about by alterations in enzyme synthesis.

Since treatment with barbiturates and tranquilizers clearly induces cleft palate in mouse foetuses, the question of mechanism can be raised. Some of the possibilities include:
1) Direct action of the drug on the physiological activity of the neuromuscular system.
2) Modification of foetal tissues by a chemical action of the drug apart from sedation.
3) A maternal effect due to the consequences of the drug treatment on maternal metabolism.

Figure 4. A diagram depicting some of the ways in which genes (G) and environmental factors (E) can influence the process of palate closure. From Fraser, et al (1957).
The underlying rationale for the variety of teratological approaches is that if a suspected mechanism of normal development can be interfered with under controlled experimental conditions, inferences can be drawn with regard to the importance of that system in normal morphogenesis.

Saxen (1973) found low concentrations of hydrocortisone prolonged palatal closure time in in vitro studies in mice. He raised the possibility of a genetic difference of metabolism between strains of mice, although he was unable to pinpoint whether the action of the teratogen is during, or after, shelf "horizontalization".

Several researchers have endeavoured to discover the changes which occur within the shelves during their progressive "horizontalization". Pratt and King (1971) used the presence of hydroxyproline in whole rat foetuses and isolated palates to calculate the rate of collagen synthesis from the thirteenth day of gestation to the first neonatal day. The amount of collagen per unit dry weight increased exponentially during the time of palatal closure. Pratt and King (1972) showed the lathyrogenic compound B-aminoproprionitrile has a dramatic and reversible inhibitory effect on collagen cross-linking in the rat embryo, which presumably interfered with the rotation of the palatal shelves from their vertical position beside the tongue.

Steffek, Verrusio and Watkins (1972) supported these findings, while Mato, Uchiyama, Aikawa and Smiley (1975) subsequently showed suppression of the so-called "programmed cell death" of
abutting shelf epithelium prior to mesodermal proliferation under the influence of B-aminoproprionitrile. Mato et al concluded that B-aminoproprionitrile decreased the "connection capacity" between the embryonic mesenchyme and epithelium resulting in palatal clefting. Although no definite conclusions can be drawn regarding the pathogenesis of B-aminoproprionitrile-induced cleft palate, it appears that the normal regressive changes in the leading-edge palatal epithelium do not occur even if the shelves are in contact.

Diewert (1976) demonstrated the need for an adequate blood supply to support the rapid proliferation of mesenchymal tissue during the process of shelf rotation and fusion. The severe epithelial anomalies which appear to be produced by doses of vitamin A and cortisone may contribute to a distortion of palatine process growth by restricting the migration of the neurovascular bundle. This bundle may be one of a number of factors contributing to movement and re-orientation of the palatine processes. However, the author advocates caution in interpretation of results, as morphologic associations in time may not necessarily reflect cause and effect relationships.

Cleft lip and palate have probably been the target of more research on developmental causes and mechanisms than have most other congenital malformations. Although detailed accounts of primary and secondary palate formation have been put forward by various authors, such factors as in vivo versus in vitro experimental differences still need to be reconciled before specific
cause and effect mechanisms can be isolated. Continued epidemiological studies are necessary to evaluate changing patterns of incidence in the community and to provide government and health bodies with a means of assessing the need and demand for rehabilitation.
CHAPTER 2: The Craniofacial Complex in Cleft Lip and Palate

In consideration of growth in cleft lip and palate individuals, one must be careful not to focus merely on the areas which appear to be affected by the defect, as this view may mask the possibility that the whole craniofacial skeleton has been involved to some extent. The reason for continued interest in growth and development of the craniofacial skeleton arises from the growing awareness that the palate does not exist in isolation from even relatively distant structures and that normal growth and development of each component of the craniofacial complex depends on concomitant growth and developmental integrity of all associated structures. (Moss and Salentijn, 1969). In cleft lip and palate there is increasing evidence that many regions of the face and cranial base are subject to variances from normal size, shape and relative position. (Krogman, Mazaheri, Harding, Ishiguro, Bariana, Meier, Canter and Ross, (1975)).

2.1 The Maxilla

Nakamura, Savara and Thomas (1972) emphasized the necessity of studying homogeneous groups with respect to sex, age and type of cleft. Due to the relatively small differences in measurement involved, any combination of these above factors will tend to distort means and variances, thus possibly obscuring the real differences. These authors studied mixed longitudinal samples of
boys and girls with repaired clefts of the lip and palate finding larger than normal maxillary width measurements in areas distant from the site of surgery. Their explanation for this was a combination of a normal or slightly wider than normal maxilla at birth in cleft patients, and a differential effect of surgery. This had an inhibitory effect on growth in the area immediately adjacent to its site, especially in the antero-posterior direction, but a diminished effect at more distant sites. Surgery could also be suggested as a variable in this study.

Fish (1973) found the growth in width of the maxillary arch of cleft palate children less than normal, whether or not presurgical orthopedic appliances were used to improve the position of the segments prior to surgery. He contended that there was a deficiency of palatal tissue at birth with both lateral and vertical displacement of palatal shelves combining to produce a wider maxilla than normal. Postnatal growth of the palatal shelves reduced cleft width. Fish suggested that more favourable changes would occur in this area as a result of using obturator appliances which facilitated palatal growth, so making surgical repair less difficult. By three years of age the mean posterior and intercanine widths of arch in cleft group were significantly less than normal. Other authors such as Wada and Miyazaki (1975) have suggested that the major growth disturbance in cleft lip and palate is in the anterior alveolar region and is characterized by anteroposterior growth insufficiency following lip repair.
Mapes, Mazaheri, Harding, Meier and Canter (1974) studied a sample of 40 patients, both prior to and following surgery, for complete unilateral cleft lip and palate. With the aid of photo-copied casts, they found a high degree of asymmetry between affected and unaffected sides, suggestive of a tissue deficiency which is slow to be replaced by hard and soft tissue. This asymmetry was located mainly in the anterior region. The posterior segment of the unaffected side was shorter than the affected side suggesting hyperplasia of this segment on the cleft side. The maxillary growth rate was found to lag during the time intervals when surgery was being carried out, but accelerated following surgery. This is the concept of "catch-up" growth. Mapes maintained that properly timed, conservative surgery allowed the maxilla to normalize itself.

Krogman, Mazaheri, Harding, Ishiguro, Bariana, Meier, Canter and Ross (1975) disputed the evidence that surgical intervention in cleft lip and palate had a growth stultifying effect upon the maxillo-palatine complex, claiming that cleft lip and palate are not isolated defects but have ramifications into the skull base and the skull vault. They also noted this catch-up growth phenomenon and recommended similar surgical principles to Mapes et al (1974).

Hayashi, Sakuća, Takimoto and Miyazaki (1976) investigated growth in unilateral complete clefts using lateral cephalograms. The movement of growth in the maxilla from one age to another was smaller in the cleft group than in normals especially after eight
years of age and, additionally, the whole maxillary complex was located in a retruded position. The residual growth rate of maxillary depth in the cleft group was also lower, suggesting a lower maxillary growth potential generally, or an effect on appositional growth at the tuberosity.

Recently some Scandinavian studies have been carried out using metallic implants following the method described by Bjork (1968). Dahl (1979) found an inhibition of transverse maxillary growth probably as a result of a combination of surgery and the disturbance in the palate suture system. In unilateral cleft lip and cleft palate, the median palatal suture has an abnormal position lateral to the midline and the untreated cleft side segment has no sutural connection with the maxilla or the non-cleft side. (Dahl and Fossh-Andersen, 1974) quoted by Dahl (1979). In the bilateral situation, the mid-palatal suture is completely absent with segmentation of the maxillary complex into three parts. Dahl pointed out that these results may have been influenced by orthodontic treatment being carried out during the study.

Perhaps these statements should be qualified bearing in mind that the condition of cleft lip and palate is itself a variable condition. The "median palatal suture" would occur only at the alveolar process in those cases with bilateral posterior cleft with unilateral anterior cleft.
Rune (1980) studied the movement of the cleft maxilla using implants with the aid of a final reference, the frontal bone, established outside the maxillary bones to eliminate the influence of treatment procedures which had clouded Dahl's (1979) study. The movement of the maxillary segments showed no individual pattern and could not be related to timing, type of surgery, or cleft type.

Johnson (1980) studied craniofacial growth in nineteen unilateral and nine bilateral cleft patients following relatively early correction of palatal defects. He found the amount of midfacial deficiency demonstrated was related to failure of maxillary bone development, rather than a disturbance in forward growth potential of the midface region. If a maxilla deficient in tissue content demonstrated a normal growth potential in terms of horizontal development, it would still never attain the dimensional size of a normal skeleton. Johnson reached this conclusion after finding comparable horizontal displacement of the maxilla in both the test and normal groups.

Several studies have also been undertaken of individuals with unoperated cleft lip and palate. Bishara (1973); Bishara, Krause, Olin, Weston, Van Ness and Felling (1976); Bishara, Olin and Krause (1978); Crabb and Foster (1977). Caution is necessary in interpretation of results from these studies as they have usually involved very small samples of a particular race and may not be applicable universally. Bishara et al (1976) studied twenty individuals with unoperated clefts who belonged to the
same facial group. Whilst the maxillary complex was not found to be significantly different in either size or relationship, there was a tendency for a relative protrusion of the anterior part of the maxilla in the cleft group, but the cranial base and skeletal face were not extensively malrelated. Crabb and Foster (1977) concurred with these views.

Bishara et al (1978) analysed two cases of unrepaired complete bilateral cleft lip and palate. Both showed a severe prepalatal protrusion caused by a severe maxillo-mandibular discrepancy with maxillary incisors bodily displaced anteriorly but with reasonable axial inclination in both cases. The cranial base in both subjects was relatively normal.

Considerable discussion has taken place regarding the origin of this so-called prepalatal protrusion in bilateral cleft lip and palate, with some surgeons referring to the existence of a "pre-vomerine bone". Pruzansky (1971) reported on the overgrowth of the premaxillary-vomerine suture which he claimed contained proliferating cartilage cells. Friede (1973) found only small cartilaginous "islands" or conglomerates of cartilage cells which he interpreted as being secondary to the overgrowth of the premaxillary-vomerine complex.

Atherton (1974) hypothesizes that the prominent "premaxilla" arises in man because the cleft allows the face to assume a normal mammalian pattern of growth. According to this theory,
the cleft acts as a premaxilla-maxilla suture and the peculiarity of man is the growth of the normal face which shows restriction of forward growth of the anterior part of the maxilla. The superior part becomes attenuated to form the anterior nasal spine by its attachment to the nasal septum through the septo-premaxillary ligament described by Latham (1970).

King, Workman and Latham (1979) studied the structure of the medial crura of the alar cartilages and their relationship to the cartilaginous nasal septum and protruded prepalate in bilateral cleft infants. See figures 5A, 5B. They found that part of the abnormal protrusion of the prepalatal process is represented by the advanced position of the basal premaxillary bone and partly by the gross protrusion of the alveolar process. The position of the basal premaxillary bone appears to be determined by a fibrous attachment to the nasal septum the "septo-premaxillary ligament".

In so far as "mechanical" concepts have been used as a possible explanation for retrusion of the primary palate after primary surgery, the protrusion of the prepalate in unrepaired clefts could be similarly explained in mechanical terms as a propelling effect of the tongue without the restriction of an intact lip, i.e., "functional matrix theory".
Figure 5A. Diagrammatic outline of normal naso-premaxillary structure. Soft tissue profile has been added to show relationship to underlying cartilage and bone. Major structural features of midline are identified anteroposteriorly: A. Medial crus of alar cartilage; B. Free inferior border of cartilaginous nasal septum; C. Anterior nasal spine; D. Alveolar process. Note anteroposterior arrangement A-D.

Figure 5B. Diagrammatic lateral view of bilateral cleft naso-premaxillary region. Comparable structures figure 5A are identified by the same letters A-D. Note superimposition of premaxillary bone (D) on columellar cartilage (A). From King, Workman and Latham, (1979).
2.2 The Mandible

The notion that the primary and secondary palate do not exist in isolation appears to be born out by evidence of effects of the condition and/or its treatment on other structures. In studies of unrepaird complete clefts carried out by Bishara et al (1976) and Bishara et al (1978), the mandible was found to be relatively shorter and the mandibular plane was steeper when compared with normal individuals. This was accompanied by a significantly larger lower face height. Nakamura et al (1972) conjectured that this shorter mandible could also be part of the cleft-producing process by interference with palatal shelf closure and fusion as their postoperative cleft lip only group had mandibles of relatively normal length, although this brings up the old question of whether the chicken or the egg came first.

Krogman et al (1975) found a normal corpus and ramus with a more obtuse gonial angle in the cleft group. The temporomandibular joint was also more retro-positioned than normal. Horowitz (1976) found all mandibular angles were increased in individuals with cleft lip and palate. While Hayashi et al (1976) found that forward undergrowth of the mandible became more evident with increasing age. Mandibular prognathism as measured by the SNB angle was also smaller in the cleft group than the controls. This fact is interesting in view of the 93% incidence of anterior crossbite in their cleft samples.
Graft-Pinhus (1976) studied thirty-nine children with repaired unilateral clefts of lip and palate in comparison with a carefully matched control sample. His results indicated wide variability. In the two cases which he judged to be "most cleft", one showed midfacial hypoplasia, pseudoprognathism and total crossbite by age eight years, while the other neither developed hypoplasia nor crossbite and had a well balanced facial appearance by age seven. Perhaps his message here is not to draw too many conclusions about the difficulty of treatment based on cephalometric evidence alone.

Vora and Joshi (1977) compared mandibular growth in twenty-five individuals with repaired cleft lip and palate, twenty-five controls without clefts and twenty-five additional individuals with normal occlusions. Their results showed retrusion of the anterior limits of the mandibular apical base in the cleft group measured by a smaller SNB angle and higher Y-axis angle. The vertical growth of the ramus was also poor with a higher gonial angle. There was a greater tendency towards a retruded skeletal chin in relation to cranial base, with retroclination and lingual placement of mandibular incisors.
2.3 Facial Proportions and Growth

Bergersen (1972) examined the relationship between skeletal maturity estimated from hand radiographs to the facial adolescent growth spurt and found a significant correlation between onset of the male adolescent spurt of all facial dimensions studied as well as standing height. The skeletal age estimate of the male adolescent spurt onset has a third of the variation of chronological age estimate and is a more accurate indicator of timing of the spurt than chronological age. Skeletal maturation is significantly correlated with the onset of the spurt in the face from at least five years prior to onset through one year following. Nakamura (1972) found similar growth rates between children with cleft lip and palate and normal children. Krogman (1975) found greater anterior upper face height and lower face height than normal, while Hayashi (1976) disagreed after finding shorter upper face heights and slightly larger lower face heights. Vora and Joshi (1977) found anterior upper face height normal with an increased lower facial height in the cleft group.

Johnson (1980) found compensation for the decreased vertical development of the maxilla evidenced by changes in the angular and dimensional make-up of the mandible resulting in an increased lower anterior face height and overall longer anterior face height. Analysis of posterior face height revealed a decrease in dimensional attainment for both the upper and lower components.
Review of this literature highlights the obvious lack of unanimity of opinion on facial proportions in cleft palate individuals. Possible reasons for this are differences in methodology, particularly in relation to measurement point selection or differences in study samples. If growth rates are not significantly different in normal and cleft palate individuals, but facial proportions do differ significantly, then surely the facial characteristics must be different to begin with.

2.4 The Cranial Base

The growth and development of the cranial base complex involves cartilaginous synchondroses whilst that of the facial complex involves intramembranous sutures, which may both be regarded as growth and adjustable sites. It is fairly well accepted that areas of cartilaginous growth are primary and important growth sites, while areas of sutural growth are secondary or accommodating growth sites.

Krogman et al (1975) indicate that S-N (anterior cranial base) length and Ba-S (clival length) tend to be larger in children with clefts than in normal individuals. Sella angle (Ba-S-N) showed greater flexion in clefting. They felt that all this pointed to basion and foramen magnum as a major area of adjustment to clefting, and that the entire anterior cranial base complex is harder hit, and shows a greater divergence from the normal pattern.
Bishara et al (1976) found the cranial base and skeletal face were not extensively malrelated in individuals with unoperated unilateral cleft lip and palate when compared to matched normal individuals. However, they noted distinct differences in dentoalveolar and skeletal relationships between the cleft group and normals. A similar result was noted for bilateral cases by Bishara et al (1978).

Hayashi (1976) found no difference in length or growth rate of anterior cranial base at any age regardless of sex. The anterior cranial base in cleft subjects seemed to grow in the same manner as in non-cleft subjects. However, they did find a slightly larger cranial base angle in the cleft group than the controls. This difference, they thought, may be related to greater angulation of medial pterygoid plates.

Johnson (1980) could not demonstrate statistically significant changes in the cranial base angulation with bilateral or unilateral clefts, although a tendency was noted for the angle to be smaller than the value found in normals.

2.5 The Nasal Septum and Nasal Airway

Drettner (1960) recorded the size of the nasal airway in 53 cases of repaired clefts of lip and palate, or palate only, and found narrowing in 45% often associated with thickening of the nasal mucosa. Unilateral cleft lip and palate were usually accompa-
nied by narrowing of the nasal airway on the same side caused by pronounced deviation of the septum towards the obstructed (cleft) side. Irritation from food and saliva was the probable explanation of hyperplasia of the nasal mucosa leading to upper respiratory tract infections and inflammation. Chaudhuni and Bowden-Jones (1978) studied 245 children with mixed clefts and found 51% with deviated septum. 24% of these nasal obstructions were bad enough to require submucous resection when older. These authors emphasized the importance of considering the physiological respiratory function of the nose along with the cosmetic problems involved in the treatment of cleft lip and palate. Latham's work in this area has been reviewed in a previous section.

The recent literature indicates that some of the craniofacial growth deficiencies observable in cleft lip and palate are apparently unrelated to surgical trauma, while other studies suggest that surgery may retard maxillary growth temporarily or perhaps permanently. Certainly not all deviations from the "normal" can be assigned to the negative effects of surgery. In some cases, surgery may have only a temporary negative effect, or possibly even a positive effect, on subsequent growth and development.
CHAPTER 3: Presurgical Orthopedics in Cleft Lip and Palate

Presurgical orthopedics as originally proposed by McNeil (1956) aimed to bring about a non-surgical closure of the cleft by mechanical stimulation of bone growth using gentle pressure brought to bear over a large area of the hard palate. If control of the dental arch could be attained in early infancy, McNeil claimed, normal function would occur with regard to respiration, swallowing, mastication and speech and hence, result in a more normal growth pattern. This control and correction of displaced maxillary segments prior to surgery was intended to assist the surgeon by presentation of a normal bony facial skeleton at operation.

Probably no other aspect of cleft lip and palate rehabilitation has been the subject of as much debate as presurgical orthopedics. The term "presurgical orthopedics" has come to not only mean the movement of entire jaw segments, that is, the teeth, bone and soft tissues as one unit, but also encompasses use of passive holding appliances which prevent the tongue from entering the cleft and facilitate feeding. Pruzansky (1964), a dissenter, stressed the need for reliance on factual evidence rather than notions of what we would like to believe, when evaluating and deciding to employ these techniques. He has claimed McNeil erred in failing to consider the role of the muscles and nasal septum in guiding growth as well as basing his claim for improvement of maxillary shelf deficiency on a premise which Pruzansky says does not exist.
Whilst advocating a joint approach of the paediatrician, surgeon and orthodontist in treatment plan formulation, Burston (1965) stated the principal factors for consideration as feeding problems, other associated abnormalities, the desirability of early orthodontic correction, the family background and whether the child should be treated at home or in a hospital. Godfrey (1982) believes facilitation of feeding is one of the main advantages of these appliances. The sucking plate provides the infant with an intact palato-alveolar base against which a feeding nipple can be compressed, at the same time keeping the tongue out of the cleft. This in turn separates the oral and nasal cavities and facilitates their separate functions, leading towards better tongue posture, activity relating to future speech and swallowing, as well as a possible "functional matrix" effect related to growth. (Godfrey, 1982.)

Robertson (1971) found considerable variation in the nature of changes produced by presurgical orthopedics as distinct from growth, in a study using tantalum implants and a cephalometer. These changes were in part dependent on the design and construction of the appliances used in any given case; also, there was the possibility of reduction in palatal cleft width without decrease of overall arch width.

Hotz (1969) and Hotz and Gnoinski (1979) advocated use of these techniques to create and maintain good initial palatal width and occlusion, so as to prevent adverse effects of primary surgery by providing mechanical retention. The latter study was a follow-up
evaluation of ten years of treatment, a very important feature of any research in the field of cleft palate rehabilitation, where the effects of any procedure performed early in life are often not fully apparent for some time. Hotz concluded that a combination of orthopedic guidance together with suitably timed primary surgery has a beneficial effect on maxillary development and, therefore, facial contour. She does not recommend routine palatal expansion except in some cases of bilateral clefting.

Glass (1970) questioned the advantages of presurgical orthopedics in bilateral cleft lip and palate due to the necessary delay in lip closure and failure to either stimulate maxillary growth or produce clinical or bony junction of the segments. The added burden imposed on the family both in terms of travelling to the clinic and possible guilt feelings of the mother through failure of the baby's acceptance of the appliance were also factors cited against presurgical orthopedics. It is difficult to justify Glass' criticism of surgical delay as a disadvantage of this technique. The most common age for lip repair is three months, which seems to be a compromise between the emotional needs of the parents and the medical condition of the child. At Westmead Centre presurgical orthopedics per se is only carried out at the request of the surgeons to reposition the anterior segment.

Georgiade and Latham (1975) described an arch alignment appliance for use in bilateral cases for both maxillary expansion and rapid prepalate retraction based on a pinned screw mechanism. This
involved placement of a pin in the "premaxilla", which is a fairly major and exacting procedure with significant attendant surgical risks. Latham (1980) has continued to advocate a similar strategy in unilateral clefts for anterior movement of the minor segment to improve the position of the depressed alar base, while Berkowitz (1981) disputes the existence of such retropositioning of the lesser segment.

Using increasingly complex analytical methods, Huddart (1967), (1977) and (1979) has concluded that presurgical orthopedics is beneficial in narrowing the width of the alveolar and palatal clefts thereby facilitating surgery, whilst it has a constrictive effect on arch growth and a retardant effect on palatal tissue growth. Huddart has also stressed the advantages of tongue confinement to a normalized space, particularly when the appliance is placed within a few days of birth.

Rosenstein (1975) has also emphasized the need for long term clinical evaluation to document whether presurgical orthopedics does any harm or good. His team's approach involved stabilization of segments by means of an autogenous bone graft in addition to presurgical orthopedics, following the method earlier advocated by Nordin and Ohlsen (1960). Although his results show no growth attenuation in the anterior-posterior dimension, it is doubtful whether he shows sufficient justification for using these procedures.
Hellquist (1971), following Skoog (1967), advocated presurgical orthopedics combined with a periosteoplasty operation to accomplish expansion in cases of marked primary collapse of the lateral maxillary segments. In bilateral cases orthopedic backward movement of the prepalate is achieved without tilting of the segment, which avoids possible damage to the growth centres of the nasal septum, sometimes caused through the use of excessive pressure.

According to Pruzansky and Aduss (1967) two questions are central to the argument regarding presurgical orthopedics:

"1) Is arch collapse an inevitable sequel to repair of the cleft lip and palate? If not, what proportion of cases do collapse? Correlatively, what is the prevalence of malocclusion particularly crossbite in children where the reconstruction did not include pre- or post-surgical orthopedics and bone grafting?

2) If some arches collapse and others do not, within the surgical experience of a school of surgeons using essentially similar procedures for all of their patients, what are the factors which favour or prevent such collapse? Is it possible that variants in morphology peculiar to the individual patient are the determining factors for ultimate arch form?"

Factors determining collapse of the arch in the population they studied were shown to include:
1) Size and shape of the alveolar process

2) Size and configuration of the palatal shelves

3) Size and shape of the inferior turbinate on the side of the cleft

4) Size and shape of the nasal septum

Aduss and Pruzansky also showed the prevalence of malocclusion, particularly crossbite, in their untreated sample was substantially less than that reported by other authors who had used presurgical orthopedics.

Rune, Sarnøs and Selvik (1979) used metallic implants in a roentgen stereophoto-grammetric study to highlight the difficulty of accurate identification of movement of segments by use of surface measurement points. Along with Huddart (1967), (1974) and Robertson and Fish (1975) they found orthopedic expansion treatment prior to palate repair had little influence on the position of the cleft segment in unilateral cases and only resulted in a limited lateral translation which relapsed completely within seven months. Changes brought about by the orthopedic appliance appeared to have been restricted mainly to movements of teeth and supporting alveolar bone. Paradoxically, rotational movements of the cleft segment occurred mainly in opposite directions to the changes measured on the casts.
Recently, Peat (1982) investigated the effects of presurgical oral orthopedics on children with bilateral complete clefts of lip and palate. He found a highly significant reduction in incisor crossbite incidence in his treated group compared with the untreated group, in both the deciduous and mixed dentition although. There was no significant difference in either buccal segment crossbite or skeletal analysis between treated and untreated groups.

At Westmead Centre presurgical orthopedics, per se, is carried out at the request of the surgeons to improve the position of the prepalatal segment in bilateral cleft lip and palate or, in cases of unilateral cleft lip and palate, to improve the position of the lesser segment prior to lip repair. In bilateral cases this anterior segment is often protruded and/or rotated on the nasal septum at a rather bizarre angulation. The origin of this deviation is possibly tongue pressure in utero. Thompson (1982) is of the opinion that simple closure of the lip on the side distant from the rotation, without the benefit of presurgical orthopedics, will not achieve septal straightening even if the lip is repaired in two stages. He feels this will only serve to cause further tension and distortion of the prolabium and possible postoperative wound breakdown. See figure 6.

Acceptance of these appliances by the babe seems to be difficult to predict and, to a certain extent, seems unpredictable depending on the attitude of the parents and the experience of the nursing and medical staff. Hotz (1964) carries out the entire
treatment on an out-patient basis. The treatment is started as early as possible, generally at the age of three to four weeks. He admits that his procedures cause a delay of one to two months in lip repair. Graf-Pinthus (1964) treats all patients ambulant-ly except a few that must be hospitalized due to other malforma-
tions. He finds that the parents of his small patients usually work with him very reliably and are glad to be able to do something for their children. Dreyer (1964) attempts to complete and adjust the appliance before the first feed. Our own chance of success seems to be enhanced if the appliance is placed during the first week and the mother and baby remain in hospital until confidence is gained in handling it.
Figure 6. Typical presurgical orthopedic appliance as used at Westmead Centre in cases of complete bilateral clefts of primary and secondary palate. Note the presence of fins on the tissue fitting surface of the appliance which can be selectively adjusted to apply pressure to the cartilaginous nasal septum as required. From Godfrey, (1982).
Figure 7. Appliance in place. A layer of micropore is laid down on the skin prior to appliance placement. This can be changed at infrequent intervals without causing undue irritation. Elastoplast strapping is then placed in order to both secure the appliance and exert a distal pressure on the premaxillary segment. This method of fixation ensures that the strapping will not be used without the appliance, a situation which may lead to disastrous collapse of the maxillary segments. From Godfrey, (1982).
Figure 8. Serial casts of patient shown in figure 7, showing degree of septal straightening which can be achieved prior to surgery. From Godfrey, (1982).
CHAPTER 4: Surgical Repair of the Cleft Lip

Surgical repair of the cleft lip is the first major step in rehabilitation of the cleft lip and palate patient. The following requirements of lip repair have been outlined by Tennison (1952):

1) Adequate muscle approximation with the muscle of the lip brought into as near normal transverse alignment.

2) Good skin coverage of the lip with the suture line placed in such a manner that subsequent scar contracture is reduced to a minimum.

3) The production of approximately normal anatomy of the lip with the preservation of the "Cuspid's Bow". Any minor residual deformity should be easily correctable.

4) The lip should have a full red border with normal "pouting" protrusion of the lower portion.

5) Adequate nostril floor with correction of as much nasal deformity as possible at the primary operation.

6) Elimination of as much undermining of the face as is consistent with good closure.

7) A simple means of arriving at the locations of the incisions so that standardization of the procedure is possible."
The earliest attempts to repair the cleft lip consisted of a simple "freshening" of the tissue edges as described by Celcus in 1000 A.D. "... For hare-lip, pound mastic very small, add the white of an egg and mingle as thou dost vermillion, cut with a knife the false edges of the lip, sew fast with silk, then smear without and within with the saliva." Quoted by Shearer, (1980).

Figure 9. An ingenious head cap devised by Hofmann in 1686 to relieve tension along the repair by means of a simple system of clasps, around which threads were drawn and tensed, depending upon the discretion of the surgeon and the amount of cheek-bolstering required. Redrawn from Shearer, (1980).
Figure 10. A surgical table containing most of the instruments considered essential for harelip repair in the early 18th century (1712) by Dionis. Redrawn from Shearer, (1980).
In unilateral cases, the cleft of the lip is usually quite wide especially at the lower edge, so that one of the main problems is to place sufficient tissue into this area. Mirault (1844) attempted to solve this problem by cutting a flap extending up one margin of the cleft and swinging it down and over to the other side. See figure 11. Hagedorn (1892) refined this technique with the introduction of his quadrilateral flap to create a Cupid's Bow. See figure 12. This was a great advance on previous procedures. Le Mesurier (1949) suggested a further modification which produced symmetry of the Cupid's Bow. See figure 13. This was followed by the Tennison repair of 1959, which standardized the procedure of locating incision points. See figures 14A, B, C. These methods were the first to employ the so-called "z-plasty" philosophy which is characterized by the transposition of two triangular flaps. According to Converse, (1977), the "z-plasty" technique as applied generally to plastic surgery has three main purposes:

1. "Lengthening of linear scar contracture

2. Dispersal of the scar, thus breaking up the straight scar, and

3. Realigning the scar within the lines of minimal tension. Elongation and interruption of the straight line and release of linear scar contracture to prevent its recurrence."
Figure 11. Diagrams available to show the Mirault operation. From Le Mesurier, (1949).

Figure 12. Hagedorn's Operation. From Le Mesurier, (1949).
Figure 13. The Le Mesurier repair. The height of the cupid's bow on the two sides. On the lateral side this height depends on how close the point A is placed to the mucocutaneous line. On the medial side it depends on how far above the mucocutaneous line the point B is placed. From Le Mesurier, (1949).

Dissatisfaction with the Cupid's Bow results of previous operations led to the rotation advancement flap repair of Millard (1957). This represented an attempt to transpose the normal non-cleft component into the correct position by its release from the abnormally high attachment to the columella base. This left a triangular defect in the upper third of the lip to be supplied by a tissue filler from the cleft side. Most techniques employed today seem to be variations of the Millard repair.

Figures 14A. A measurement is made on a twisted alloy steel wire equal to the length of the columella side of the cleft.
Figure 14B. The wire is divided into thirds and bent into a "Z" so that the upper angle points to the cleft side. It is then placed with its upper end at point A.
Figure 14C. The skin incisions are made according to the stencil and the sides of the cleft are brought together and sutured as desired.
Surgery for correction of the bilateral cleft lip is often complicated by the lack of attachment and protrusion of the prepalatal which is advanced on the nasal septum. (Latham, 1979). This complete separation of the central fronto-nasal component of the prolabium and anterior palate influences the nose, philtrum,
musculature, vascularity and nerve supply as well as the growth and development of the maxilla. (Millard, 1977) The nasal tip is flattened with columella absence and separation of the medial crura of nasal cartilages. Several methods of dealing with this problem have been proposed.

In 1556 Franco advocated excision and discarding the protruding anterior segment prior to lip closure, followed by a prosthetic reconstruction of the anterior segment. This technique fell into disrepute because the secondary deformity created seemed as bad as the original defect. Recently, however, Motohashi and Pruzansky (1981) have revived interest in this method in certain carefully selected cases dictated by the likely prognosis of a severe facial convexity. However, this has not been advocated as a primary procedure and is never carried out before the age of seven years.

Browne (1949) also suggested a surgical setback of the "premaxilla" due to what he claimed was the existence of a cartilage-filled suture line between the vomer and what he called the "prevomerine" bone. If the prepalate was not moved into a normal position and fixed firmly Browne felt there would be great difficulty closing the lip over it resulting in a very ugly profile. Later Matthews, (1952) supported this method but warned against any disturbance of the "prevomerine suture line", lest retardation of the subsequent development of the area would occur.
Bauer, Trusler and Tondra (1959) advocated simple early closure of the lip clefts to provide a retrograde molding action on the prepalate. They acknowledged that this molding action could lead to medial collapse of the maxillary segments resulting in anterior trapping of the prepalate, as well as placing a significant initial stress on the repaired lip segments. Slaughter and Pruzansky (1954) noted the importance of re-establishment of the prime function of the orbicularis oris muscle as a force responsible for "favourable" reconfiguration of the bony skeleton of the middle third of the face.

Gillies (1957) suggested a compromise solution to the problem, in the form of a partial premaxillectomy operation, in which the anterior mucous membrane was peeled back from the "premaxilla" and the anterior bone and tooth-buds were cut away. In this way arch form preservation was encouraged.

Several authors have also proposed shortening of the septovomerine stalk. Brophy (1923) used a mechanistic approach including an oblique incision in the nasal septum to move the "premaxilla" distally, followed by attainment of a bony union with the maxilla using wires, sutures and lead plates for fixation. Huffman and Lierle (1949) suggested premaxillary setback after removal of a portion of the "overly developed" nasal septum. This was carried out to avoid endangering the suture line by tension developed from the lip repair. See figure 16.
Figure 16. Brophy's technique to move the "premaxilla" back using an oblique incision. From Bishara and Olin, (1972).
Figure 17. Surgical removal of part of the nasal septum. From Bishara and Olin, (1972).

For the bilateral lip repair many surgeons originally merely adapted their unilateral repair designs by doubling them. However, the bilateral cleft lip is not merely a right or left unilateral fissure with its mirror image on the opposite side, but tends to vary a great deal in the amount and shape of the deformity.

Brown, McDowell and Byars (1947) admitted that repair of the bilateral cleft lip is about twice as difficult as the unilateral and the results about half as good. These authors recognized the problem with shortness of the prolabium and introduced a pair of triangular flaps beneath its inferior border gained from the
lateral elements of the lip. They also recommended later closure than for unilaterals because of the greater magnitude of the operation and the fact that kinks in the nasal septum behind the "premaxilla" are sometimes so marked as to occlude one or both nasal airways at birth. See figure 18.

Figure 18. *The Mirault operation as modified by Blair and Brown for the bilateral cleft lip repair. From Millard, (1977).*
Figure 19. Millard's use of forked flaps for columella lengthening to improve nasal appearance. From Millard, (1977).

Millard (1967) described a procedure which lengthened the short columella, reduced the unattractively-wide prolabium to more natural proportions, and released the depressed nasal tip. He claimed this provided a possibility of correcting the nasal deformity at the same time as lip closure, to allow growth to proceed unimpeded. The previous plans had included closure of the lip in one or two stages followed by columella lengthening at five years. This had tended to result in a flat nose. The lip scar results also tended to be improved with a single stage
procedure. Millard noted one possible problem with this procedure as interruption of the blood supply to the prolabium and "premaxilla" which could lead to necrosis. Perhaps one criticism which could be levelled at Millard is his drawing of conclusions, apparently based on his experience with relatively few cases.

When considering the effects of lip repair, it is necessary to remember that the lip makes up only one part of the primary palate of a child with a cleft of the primary and secondary palate. There seems to be a tendency to concentrate on the lip surgery which has reached a state of technical excellence and to overlook the importance of obtaining a satisfactory repair of the anterior palate. This natural tissue separation of the oral cavity from the nasal cavity is important not only for health reasons, but also to obtain the growth stimulus provided by the functional continuity of skeletal parts. These aspects will be discussed in a following chapter.
CHAPTER 5: Effects of Lip and Anterior Palate Surgery

The repair of the lip musculature establishes the integrity of the so-called "buccinator mechanism". Graber (1971) describes this as a system of muscles consisting of the orbicularis oris muscle, buccinator muscle, the pterygo-mandibular raphe and superior constrictor muscle of the pharynx. This muscular complex is opposed by the tongue which in normal circumstances provides a state of muscular equilibrium on which balanced growth of the maxilla and articulating bony relationships are dependent. Coupe and Subtelney, (1960) showed a definite tendency toward a deficiency of hard palate tissue in all clefts which involve the palate, with minimal deficiency in unilateral cases and maximum deficiency in cases of bilateral and posterior clefting. It would, therefore, seem reasonable that in unilateral clefts simple re-direction of muscle forces from lip repair would produce the maximum reduction in width of the cleft. Pruzansky (1955) also noted this beneficial reduction in cleft width following lip repair.

Mazaheri, Harding, and Nanda (1967) attributed this reduction in cleft width to growth and possible downward displacement of palatal shelves as well as muscle moulding. They also found that lip surgery had no effect on bimaxillary or bituberosity width. Mapes et al (1974) found that growth rate of the maxilla lags behind normal during the time intervals in which surgery occurs, but is accelerated in intervals following surgery. However, the incisor to canine length segment of the affected side did not
show this acceleration rate after lip surgery. This was explained as a result of tissue compression and moulding following repositioning of segments and also as a result of tissue deficiency.

Figure 20. The buccinator mechanism. Note continuous muscle band that circles the dentition and is anchored at the pharyngeal tubercle. From Graber, (1961).
Figure 21. The decussating fibres of the orbicularis oris muscle, the anterior component of the buccinator mechanism. Note the sphincter-like or "purse-string" functional possibilities. From Graber, (1961).

Bardach et al (1977, 1978, 1979a, 1979b, 1980) have conducted a series of animal experiments on the influence of lip and palate surgery on facial growth. In the first three studies the hypothesis was tested that cleft lip repair inhibits facial growth in rabbits. Observations during the 20-week postoperative period and the results of skull measurements after sacrifice at the termination of the experiment demonstrated that significant inhibition occurs in the anterior-posterior dimension of the facial skeleton following cleft lip repair. These findings support the hypothesis that lip repair is an important factor in the inhibi-
tion of anterior-posterior facial growth. Lip pressure was found to be less in normal animals with unrepaired cleft lips and became greater than normal in the early weeks after lip repair. Comparisons between two differing methods of lip repair showed significant intergroup differences in patterns of pressure change over the period of observation.

Bardach et al (1978) by direct cephalometry found inhibition of facial growth in the surgically created unilateral cleft lip, alveolus and palate group of rabbits without repair. They also found inhibition of facial growth by primary lip repair. Bardach et al (1979a) found a correlation between the increase in lip pressure following repair and the degree of midfacial growth inhibition which occurred.

Rintala and Haataja (1979) advocated the use of a lip adhesion procedure in cases of wide alveolar clefts in order to create more favourable anatomical conditions to facilitate the final repair of the lip and nose, in effect, creation of an artificial Simonart's band. The width of the unilateral complete cleft alveolus diminished by two-thirds on the average, depending upon the extent of deviation of the alveolar segments. The effect on the width of the bilateral cleft was clearly less predictable than in unilateral cases. In fact, several cases of collapsed lateral segments occurred, resulting in lack of space for the protruding anterior segment.
Kaplan (1978) investigated growth of the unilateral cleft lip and noted growth discrepancies which frequently followed surgery for repair of clefts of lip. He attributed these discrepancies to variations in the technique of repair. The study showed that a
Cleft lip has a ten to twenty per cent greater growth in the transverse direction parallel with the orbicularis oris muscle than in the vertical direction perpendicular to the muscle. The procedures which transpose tissue from the transverse to the vertical will lead to excessive vertical growth.

The sphincteric nature of the perioral muscles can be seen to have a strong moulding influence on the underlying bony structures. Prior to lip 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 by the superior pull of the septo-premaxillary ligament laterally. (Latham, 1979). It is logical, at least in so far as the unilateral case is concerned, to prevent these distorting forces as soon as possible, by establishment of an intact musculature by surgical repair.

The classical teaching by Holdsworth and Millard is that the lip should be repaired when the baby weighs ten pounds, is thriving and has a haemoglobin concentration of ten grams per cent. This usually results in the operation being undertaken at three months of age, when the child is not only in fit condition to undergo surgery, but the parts are sufficiently large and robust to allow a reasonable degree of precision in the repair. It should also be noted that there is usually considerable pressure from the parents for the lip repair to be carried out as soon as possible. In the bilateral cleft lip, it may be necessary to deal with the protruding anterior segment prior to lip repair.
When considering the effects of lip repair, it is necessary to remember that the lip makes up only a part of the primary palate of the child with a cleft of primary and secondary palate. It is very easy to concentrate on lip surgery and overlook the importance of obtaining a satisfactory repair of the anterior palate. To obtain a natural tissue separation of the oral cavity from the nasal cavity is important not only for health reasons, but also to obtain the growth stimulus provided by the functional continuity of skeletal parts which transmit and distribute the complex patterns of stresses and strains associated with oro-facial activities. Moss (1968) and Skoog (1967) drew attention to the principle of the procedure of "boneless bone grafting" involving the establishment of periosteal continuity across the cleft, using local flaps from the bordering maxillary segments.

In the case of the bilateral cleft, the surgeon is often faced with difficulty in obtaining skilful conjunction of the three lip elements in his repair by the presence of a prominent anterior segment. The older surgical manoeuvre put forward by Brophy (1923) of resecting the vomer in order to retroposition the "premaxilla", had disastrous effects in restricting growth of the middle third of the face.

Godfrey (1969) categorized four approaches to surgical closure of the primary palate in cases of complete bilateral clefts:

1) Primary repair of the lip only with or without late closure of the anterior nasal floor.
2) Primary repair of the lip only, but with closure of the anterior nasal floor in early childhood.

3) Primary repair of anterior palate and lip simultaneously.

4) Primary repair of the alveolar gap with subsequent closure of the lip and anterior palate.

Approach (1) can result in inward collapse of lateral alveolar segments behind a still prominent "premaxilla". That is, the anticipated restricting and moulding influence of the repaired lip musculature, which is such an asset in the unilateral cleft lip, does not occur (Brown, 1949).

Godfrey (1969) believes there has been a false notion that the prepalate is held protruded simply by collapse of the two lateral alveolar processes posteriorly. He has not observed orthodontic expansion of these two processes to be automatically followed by any substantial retrusion of the "premaxilla". On the contrary, such treatment is liable to increase the size of the fistulous opening into the nose, making later closure of this fistula a difficult procedure. Whilst the maxillary incisor teeth may be retained in a satisfactory state, they may be over-erupted and show poor inclination, which will make it most difficult to adjust them orthodontically to a satisfactory cosmetic and functional level. This is the reason for Motohashi and Pruzansky's secondary "premaxillary" excision.
Approach (2) has an identical early picture to approach (1) although the fistulous opening around the "premaxilla", even after orthodontic expansion, may not have become so large. The tying together of the central maxillary and two lateral elements for the remainder of childhood and adolescent facial growth is also beneficial, although according to Godfrey the attendant malocclusion would only be less in degree than that associated with approach (1) repairs.

The third approach presents the most favourable outcome in terms of facial growth and degree of malocclusion. The expected medial collapse of the alveolar elements is not as great and orthodontic expansion need not be impeded by any apparent tying-in action of the primary surgical closure. The maxillary incisor teeth and alveolar segments are much better placed than in the first two types and the external profile is more satisfactory. Godfrey cites the action of the Simonart's band, linking a cleft lip as a particular example of what could be the initial surgical objective in retruding the prominent "premaxilla". The Simonart's band frequently serves to tie the "premaxilla" to the lateral element, thereby controlling the direction and amount of its growth as compared with the lateral elements. Thus, there could be a case for initiating this anterior nasal floor repair prior to primary lip repair as indicated by the fourth approach.
Thompson (1982) has pointed to a further advantage of this technique. The surgery for the anterior palate can be carried out with good access without being hampered by the presence of a tense repaired lip. In this way there is less possibility of residual fistulae in this area which will be difficult to close surgically later.

Manchester (1970) raised the question of allocation of priorities in surgical repair of the various components. He believed that an inordinate amount of time and effort was being devoted to the attainment of good dental occlusion at the expense of speech and a satisfactory appearance. He advocated a two stage repair with the objective of the first operation to leave nothing more than a cleft of soft palate to be repaired subsequently. This author proposed that the large volume of literature devoted to treatment of velopharyngeal incompetence, which is a cause of poor speech, resulted from nothing less than initial surgical failure due to misplaced priorities.

Muir (1966) also favoured early repair of the anterior palate noting that the cause of the collapsed arch, often seen after primary lip repair, should not be attributed to tension from the repaired lip, but is due to the frequently employed method of alveolus closure using a single nasal layer. This leaves a raw surface on the buccal aspect often up to one centimetre wide, which is left to granulate and heal by secondary epithelialization. Contraction of scar tissue results in rapid collapse of
the lesser alveolar segment leading to arch collapse. Burian (1957) first advocated the turning back of a small flap of mucosa from the buccal aspect of the lip. Muir's operation involved the use of mucosa from the free edge of the lip which is usually discarded at operation to provide a buccal layer for repair of alveolar cleft. He claimed no patient subjected to this technique developed an anterior fistula and that the alveolar gap closes by the time of completion of the primary dentition with a good arch shape maintained. One possible drawback of his technique was caused by the residual gap between the maxillary segments which made closure of the hard palate more difficult, sometimes requiring a second stage repair using a Veau Flap.

Dey (1974) is in agreement with these principles. He cited as the main advantage, the ease of access to carry out a repair back into the hard palate at the primary operation, thereby avoiding any unwanted fistulae. His technique is similar to Muir's but differs in detail in the size and formation of the flap. Dey also cautions against use of any incision which transgresses the smooth alveolar mucoperiosteum on the ridge before the teeth erupt. He feels there will not only be localized dental and alveolar deformity, but also an unnecessary posterior narrowing of the maxilla following completion of palatal closure.
Figure 23. The Muir flap for anterior palate repair.
A. A left cleft lip and palate showing the markings for a triangular flap type of repair.
B. The incisions. The tissue of the free edge of the lip on the lateral (left) side of the cleft will be used for the flap to repair the alveolar cleft.
C. View of the posterior aspect of the lip to show the design of the flap.
D. The flap has been raised on a narrow base.
E. The nasal layer has been sutured and two "A" stitches have been passed through the flap.
F. The flap in position at the completion of the operation. From Muir, (1966).
Mention must also be made of the so-called "Slaughter Procedure" put forward by Slaughter and Pruzansky, (1954). This involved the repair of the soft palate or velar cleft as a primary procedure with subsequent closure of the hard palate and lip. Perko (1979) and Hotz and Gnoinski (1979) report good results with soft palate closure at eighteen months of age following lip closure at approximately six months. The hard palate is left untouched until five to eight years. These patients are routinely treated with early maxillary orthopedics as an integral part of the rehabilitative process since its introduction in 1969. Final outcomes are no available as yet as growth is not complete.

It is clear that the mechanics of lip repair have reached a standard of technical excellence. The main dispute between authors such as Bardach, Kaplan, and Perko and Hotz relates more to the timing of the various elements of the repair rather than the actual techniques which appear comparable. As Perko (1979) and Hotz (1982) have pointed out, the really important issue is not how you carry out the repair so much as what works best in your own hands, having due regard to the needs of the individual patient. Constant evaluation of the outcomes is of paramount importance for every "team" involved in this work.
CHAPTER 6: Primary and Secondary Bone Grafting in Cleft Lip and Palate

Cleft bone grafting was introduced by Nordin (1957) as a means for providing a substitute for a lack of tissue in the cleft region. An attempt was made to estimate the amount of bone deficiency in the alveolar process and to replace this with free bone grafts from the tibia of the patient. In addition to improvement in maxillary stability, Nordin found that the shape of the upper dental arch, intermaxillary relationship, and facial appearance relative to skeletal support also improved following his procedure. However, Nordin's cases were only followed for six to eight months. By his own admission, long-term follow-up is necessary to enable a critical appraisal of the technique. This same criticism could be levelled at other workers prior to that time, when a great deal of experimental surgery was being undertaken without the benefit of long term evaluation, for example, Brophy (1923).

Jolley and Robertson (1972) highlighted some confusion regarding terminology in the division of classification of bone grafting procedures into primary and secondary. "Primary" has been used to indicate bone grafts inserted at the time of original mucosal repair, and "secondary" to indicate that the graft was inserted into the bony gap after the mucosa had been repaired. As this classification took little account of the age of the patient, the authors suggested the term "primary bone grafting" be restricted
to grafting undertaken at around the age of one year or the time of secondary palate repair.

Matthews, Broomhead, Grossman and Goldin (1970) were enthusiastic regarding both primary and secondary bone grafting procedures. They claimed a very high survival rate of the primary graft, as evidenced radiographically, together with a subsequent reduction in the need for conventional orthodontics in order to establish a perfect occlusion. However, no control series without bone grafts was provided for comparison to justify this claim. Secondary bone grafting was carried out by these authors for the purpose of:

1) Correction of malocclusion in a collapsed maxilla and eruption of teeth into the gap so created.

2) Relief of an obstructed nasal airway or airways.

3) Improvement of the patient's appearance by the augmentation of the middle third of the face, elevation of the alar base, and stretching of the tight upper lip.

These authors set eighteen years of age as an optimum to allow the maxilla to occlude with the fully developed mandible and hence maintain the correct relationship throughout adult life.

Figure 25. A. Preoperative view of cleft and adjacent teeth. B. Migration of teeth into the bone graft one year later. From Robinson and Wood, (1969).
Robinson and Wood (1969) and Wood (1970) found primary bone grafting prevented collapse of the lesser maxillary segment by acting as a splint. This is achieved by bringing this segment under the influence of the growth stimulus of the nasal septal cartilage, which seems to be important in maxillary growth and development in the first six years of life. Scott (1967). The teeth formed at the margins of the cleft also tended to migrate into the grafted zone.

Rosenstein (1969) achieved butting of the alveolar segments with an orthopedic appliance, prior to placement of an early bone graft. To fill out the "alveolar base and enhance facial symmetry", Rosenstein (1975) carried out a cephalometric and intra-oral evaluation of patients, from six to nine years of age, who had been treated by this method. He found no evidence of either growth attenuation in the anteroposterior direction or an increase in crossbite incidence when this sample was compared to a group treated with other procedures. Enany (1981) carried out a cephalometric study on a sample supplied by Rosenstein, an exponent of bone grafting, finding a retropositioning of both the maxillary and mandibular apical bases but a "harmonious" dental base relationship.

Jolleys and Robertson (1972) provided the first disquieting evidence of detrimental effects of primary bone grafting in their five year study. Prior to this the literature was full of optimism, with most authors taking what could be termed a mechanistic approach, to show how easily the surgery could be performed with
minimal risk to the patient. Robertson and Jolley's found clear
evidence of deleterious effects on maxillary growth of children
who had undergone primary bone grafting. These authors postu-
lated that the graft caused prevention of growth in the gap or
bone adjacent to the gap and even reduced the width of the gap by
contraction. This limitation of maxillary growth was manifested
by a reduced anteroposterior development, increased crossbite
incidence, and reduced upper jaw area. Thus the technique was
abandoned by these authors.

Skoog (1967) described the technique of "boneless bone grafting"
which involved the establishment of periosteal continuity across
the cleft utilizing local flaps from the bordering maxillary
segments. His procedure was founded on three main assumptions:

1) The periosteum covering the maxillary segments pos-
sesses normal growth potential.

2) Denuded bone in this area will regenerate normal
periosteum similar to other bones.

3) The re-established interaction between growth cen-
tres on the medial and lateral sides and the biomechan-
ics of the environment will determine the growth and
development of the united maxilla.

In a refinement of this technique, Skoog added the haemostatic
material "surgicel" to augment the blood clot of the periosteal
"sandwich" and so to augment bone formation. However, due to the
difficulty of obtaining tight closure of the pocket formed by the twin periosteal flaps in complete clefts, he later recommended a two stage procedure as a safety measure. Skoog felt that the classical method of bone grafting, per se, was unnecessary as the grafts became completely replaced by new bone. Recently the role of "surgicel" in stimulation of bone formation has been questioned by Nappi and Lehman (1980) following evidence of a detrimental effect on periosteum and endosteal bone.

Figure 26. Skoog's technique of "boneless" bone grafting. Operative procedure for double-layered periosteal flap repair of clefts of the primary palate. (A) Outline of incisions along the borders of the cleft carried through mucosa and periosteum. (B) The nasal floor has been reconstructed by suturing mucoperiosteal flaps mobilized from the deep aspect. Extension of buccal gingival sulcus incision for exposure of the maxilla, and outline of a periosteal flap on the outer aspect of the maxilla are also shown. (C) Maxillary periosteal flap rotated into position across the cleft and sutured.
Wood (1970) criticized Skoog's techniques due to what he thought was an unnecessary delay in bone formation which allowed medial collapse of the lesser segment and loss of growth potential by failing to connect the segments rigidly to the nasal septal cartilage. Perhaps he showed some insight in recommending presurgical orthopedics for correct segmental positioning to obviate the chance of perpetuation of the deformity.

Skoog's teachings have continued to find popularity in Scandinavia, with several follow-up evaluations appearing in the literature. Thilander and Stenström (1974) found encouraging results with regard to maxillary growth and dental occlusion in cases followed up for eight to twelve years following "surgicel" implantation.

Ranta, Oikari, Rintala and Haataja (1974) found narrowing of the alveolar cleft in unilateral cleft lip and palate in most cases after periosteoplasty, in cases followed up for three years after palate repair. The extent of this narrowing seemed to vary according to the original width of the defect, with the rate of narrowing being more pronounced in the first year after lip repair. This narrowing occurred irrespective of formation of any radiographically apparent bone bridge between the segments.

Hellquist and Skoog (1976) studied the effects of periosteoplasty longitudinally from infancy to five years, using a consecutive series of patients with complete unilateral clefts of lip and palate. This five year follow-up study was not found to demon-
strate any significant difference in maxillary growth and development when compared with control material. Descriptive analysis of occlusion revealed an increase of buccal crossbite in the experimental group with the wider clefts. The results were better in narrower clefts. A trend towards increased incidence of anterior crossbite became evident as the study progressed. (Hellquist, 1979). Although the results of cephalometric analysis for buccal crossbite incidence remained favourable. Interestingly, Hellquist and Ponten (1979) found a significantly high frequency of normal permanent maxillary lateral incisors in the periosteoplasty group, leading them to question the role of surgery as an aetiological factor in dental abnormalities, as put forward by Zilberman (1973).

There has also been a revival of interest in secondary bone grafting, particularly in relation to repair of residual fistulae. Lehman, Curtin and Haas (1978) and Ames, Ryan and Maki (1981) use an autogenous particulate cancellous bone marrow graft to restore "form" and "function" to the alveolar cleft. Their criteria for successful grafts include radiographic evidence of bone formation bridging and obliterating the alveolar cleft, clinical evidence of complete oronasal fistula closure tested by probing, and finally, long term orthodontic follow-up showing eruption of teeth into the previously grafted site where applicable. These authors make no mention of possible deleterious effects on growth.
Using a roentgen stereophotogrammetric study with the aid of metallic implants, Rune, Sarnäs and Selvik (1980) analysed the movement of the maxillary segments after expansion and/or secondary bone grafting. The expansion seemed to bring about complicated movements of the maxillary segments. Stability of the final relationship between the segments was not achieved by secondary bone grafting.

Eldeeb, Messer, Lehnert, Hebda and Waite (1982) are fascinated by the possibility of normal canine eruption into grafted alveolar defects, claiming a higher success rate when the root of the canine is one-fourth to one-half formed at age nine to twelve years. There is some probable justification for this procedure if it facilitates subsequent orthodontic treatment and retention, and if the operation is carried out for the conjoint purpose of lip, nose or palate revision. The authors even suggest the possibility of a further operation to facilitate complete eruption. I would contend that further surgery is a very high price to pay for good canine positioning.

It would seem that the technique of primary bone grafting is in decline probably because it not only fails to achieve the objectives outlined by its originators, but also appears to actually cause harm in some situations which actually worsens with time. The surgeons have apparently adopted a "sculpturing" approach to their plastic surgery and tended to treat the face and jaw bones like clay to which bits could be added or subtracted to give
shape, with only a simplistic view of the functional biology of bone. However, the techniques of periosteoplasty and secondary bone grafting continue to be popular, with an increasing number of necessary long-term evaluative studies appearing in the literature.
CHAPTER 7: Repair of the Cleft of Secondary Palate

The aim of closure of the cleft palate is to separate the nasal cavity from the oral cavity. This serves as an aid to normal speech development as well as improved mastication and efficiency of deglutition. Blocksm, Leutz and Mellerstig (1975) have provided the following set of criteria for evaluation of a satisfactory approach to cleft palate repair:

1) Minimal tissue loss

2) Minimal undermining

3) Minimal scar formation

4) Minimal jeopardy to the growth potential of the palate and adjacent structures

5) Minimal bone exposure

6) Satisfactory functional result in speech, respiration and glutation

7) Protection of the Eustachian tube by obtaining a complete separation of the nasal cavity from the oral cavity.
The original methods of repair relied on variations of the simple apposition of tissues and mucosal suturing. Since Dieffenbach's description of a rather radical closure of the cleft palate in 1826, many surgeons who have performed palatoplasties over a long period have described secondary oral and facial deformities. In fact, the present high level of interest and experimentation in the field could be a reflection of the widespread frustration with the current techniques.

In 1861, Von Langenbeck proposed a more sophisticated approach using freed mucoperiosteal flaps sutured across the cleft, leaving a raw palatal bone surface to heal by granulation cover. Veau (1931), quoted by Wardill, recommended that raw surfaces should be covered as far as possible, claiming sutured flaps which were prevented from adhering to the underlying bone of the hard palate would lead to excessive wound contracture. Wardill (1937) and Kilner (1937) severed all bony attachments of the soft palate in the hope that the greatest length of the soft palate would be secured along with a better speech result, provided the operation was carried out before speech age. This operation has since become known as the "V-Y pushback" procedure consisting of three or four flaps depending on the degree of clefting present, and is probably the most commonly used operation today. It is intended to provide a lengthening of the soft palate to allow correct function with the pharyngeal wall, thereby eliminating the nasal air escape which results in the classical poor "cleft palate speech".
These conventional techniques were questioned by Dunn (1952) as being responsible for detrimental effects on the development of the maxillary arch and dentition. He proposed using a vomer flap to close the hard palate cleft, combined with simple closure of the soft palate without pushback. Dunn described this as a minimum surgical intervention technique which would allow the teeth to erupt into their proper positions.

Figure 27. Veau's operation. The flaps were designed to allow the palate to ascend and re-attach themselves to the bone. Note the length of these flaps. From Wardill, (1937).
Figure 28. Wardill's V-Y palatoplasty. The mucoperiosteum of the hard palate is completely severed by an oblique incision. From Wardill, (1937).

Slaughter and Pruzansky (1954) recommended soft palate closure as a primary procedure to correct the displacement of structures in the nasopharynx caused by aberrant muscle forces. Just as the lip repair had a profound and usually beneficial influence on the anterior segment of the cleft, they reasoned that soft palate repair would have similar results in the nasopharyngeal area and prevent movement of the tongue into the nasal cavity. Whilst this procedure would probably produce a supple soft palate of sufficient length to enable good speech quality, one would conjecture that a major disadvantage may be collapse of the anterior segments prior to repair of the anterior palate. This would
require subsequent orthodontic expansion, perhaps resulting in large palatal fistulae which would be difficult to close surgically.

Figure 29. The use of the Vomer flap for palate repair as described by Dunn, (1952). An incision is made at the junction of the palatal and vomer mucous membranes and the tissues are elevated from the bony portion of the vomer. The edge of the opposite side of the cleft is incised and freed for about 2 mm. The Vomer flap is then sutured to the palatal edge with mattress sutures. The V-shaped defect is obliterated in about 2 months by granulation tissue giving a smooth palatal arch.
Other authors have also been concerned with the effect of scar tissue contracture on facial growth. Wynn (1979) advocates closure of the secondary palate with an osteotomy technique, avoiding the use of mucoperiosteal flaps which are suspected of being responsible for subsequent maxillary arch constriction. This is performed at the age of nine months to take advantage of "better" osteoblastic activity and to lessen the risk of damage to the blood supply of the hard and soft palates. However, as yet very few cases have been presented by Wynn to substantiate his claim of normal dento-maxillary complex growth following his procedure.

Figure 30. Vomer flap covered with an autogenous skin graft for closure of the hard palate. From Jonsson et al, (1980).

Fig. 1. A schematic representation of the palatal surgery performed on group VS. The vomer flap is turned and approximated to the lateral segment (a) and is then covered with a skin graft (b).
Blockmra et al (1975) have also adopted a conservative approach avoiding use of mucoperiosteal flaps. Soft palate repair is carried out at age two years using a modified Von Langenbeck without pushback, exposure or interruption of the posterior palatine vessels. Surgical closure of the hard palate is delayed until after five years of age, when a simple turnover vomer flap is used. Surprisingly, these authors report no more speech problems with this technique when compared with the palate lengthening, mucoperiosteal flap techniques previously used. However, they do mention the possible need for Teflon paste injections into the posterior pharyngeal wall or a pharyngeal flap operation as a secondary procedure to facilitate palato-pharyngeal closure, improving soft palate function for speech purposes. Jonsson, Stenström and Thilander (1980) suggested a vomer flap covered with an autogenous skin graft for closure of the hard palate to minimize scar contraction, with soft palate closure by medial displacement of soft tissue.

Occasionally, no matter how carefully the primary repair has been carried out, residual fistulae will be present in the hard and soft palate. These need to be assessed carefully as to whether they warrant further surgical intervention for closure, as they may be asymptomatic with little clinical significance. Occasionally, however, the fistulae may cause speech problems or allow nasal secretions to enter the mouth, or vice versa, the embarrassing exit of oral fluids via the nose. Larger fistulae require lateral release incisions with undermining between them and the fistula margins, if successful healing is to be main-
tained. Some large fistulae require local rotation flaps utilizing a turnover flap from one side to obtain nasal lining and a direct flap from the other side to obtain oral lining.

Persistent hypernasal speech which does not respond to adequate speech therapy will require a revisionary surgical procedure to assist in providing the correct anatomical relationship of the structures to enable them to function correctly. There are three basic forms of this procedure:

1) secondary pushback palatoplasty
2) pharyngoplasty
3) palato-pharyngoplasty.

In the final analysis these procedures must all be evaluated in the light of our original aims of treatment which are easily stated yet not so easily achieved, namely, to achieve normal appearance, normal speech and normal dental occlusion. Whilst these requirements are to some extent dependent on each other, the achievement of one may often lessen the chances of achieving another. Perhaps the best that can be hoped for is a compromise in which sacrifices must be made in some areas to achieve a result which is in accord with patient's greatest need.
CHAPTER 8: Effects of Cleft Palate Surgery

As Graber (1949) so succinctly stated, "In congenital defects such as cleft palate and cleft lip, the surgeon starts with an abnormal pattern of obscure aetiology, unknown growth and development, and questionable prognosis. Surgical management labours under a further handicap because in spite of the fact that therapy is instituted quite early, any appraisal of the results must await nature's fullest contribution conferred by maturity."

The problem is to determine the effect of surgery on the potential for growth. (Slaughter and Pruzansky, 1954). The surgery can either aid in directing the natural growth processes into proper channels through establishment of muscle balance across the defect, or it can grossly interfere with the normal developmental changes by hindering growth through interference with blood supply, introduction of scar tissue, or destruction of growth centres. According to Slaughter and Pruzansky, several interacting muscle groups require consideration:

1) the tongue which acts to displace the palatal process

2) the lateral and medial pterygoid muscles which exert an unopposed lateral pull on the pterygoid plates during mandibular function, and
3) the levators and tensors of the soft palate which are unopposed in cleft palate patients and hence contribute to the divergence of the pterygoid plates.

According to Coupe and Subtelny (1960) all palatal clefts exhibit some tissue deficiency. Bilateral and posterior cleft types are worse in this regard and also have considerable lateral displacement of the maxillary bones of the oronasal area. This is an important factor in evaluation of the outcome of surgery.

Figure 31. The postoperative scar tissue which extends more or less continuously from the pterygoid plates to the incisor region, affecting the anterior adjustment of the buccal teeth as well as the incisors. From Ross and Johnston, (1972).
In assessing these outcomes, we are concerned with facial growth measurement, usually determined by cephalometric analysis, and occlusion. The majority of workers cite the following variables which may affect the outcome:

1. Type of surgery performed
2. Skill of the surgeon

Several workers have carried out animal experiments to determine the effects of surgical repair on the secondary palate. As part of a continuing series of rabbit experiments, Bardach et al (1979) tested the assumption that palatal repair, per se, is an inhibitor of facial growth, especially in the transverse direction. A group of rabbits with surgically created clefts of the lip, alveolus and palate, was subjected to direct cephalometry following palatal repair, using a two flap palatoplasty procedure. By leaving the cleft lip unrepaired, the influence of the lip repair on mid-facial growth was eliminated. The results were compared with an unrepaired group and a normal group. As no significant differences were found between the two groups with surgically created clefts, the authors were unable to conclude that cleft palate repair either adds to, or counteracts, the inhibition of anteroposterior growth caused by surgical creation of the cleft. Bardach et al (1980) tested the influence of simultaneous lip and palate repair, based on the assumption that an interaction between both operations would cause more severe
growth inhibition than either procedure performed separately. The main effect on the experimental group was a significantly less weight gain than animals in the other groups, possibly as a result of such a traumatic operation causing long term feeding problems.

Caution must be exercised in drawing too many conclusions based on the results of these animal experiments as the clefts were surgically created and the animals were killed twenty weeks postoperatively, at age twenty six weeks, when according to Engdahl (1972) the growth of the rabbit's facial skeleton is completed.

Meijer and Prahl (1978) compared two different surgical techniques in dogs with artificially created clefts of the palate using implants inserted into the maxilla before surgery in the method described by Bjork (1955). The types of surgery compared were the common periosteal flap technique of Langenbeck and the bone flap technique of Dieffenbach in which the mucoperiosteum was left intact except along the fracture line. Theoretically this should lead to less scar contracture. The results suggested the bone flap operation had the most detrimental effect on the growth of the dento-maxillary complex, but these cases showed a slight tendency toward catch-up growth following the initial surgery. Kremanak et al (1978a) studied the effects of V-Y-type palatoplasties on maxillary growth in beagles, as a follow-up to
previous work related to contraction during healing of standard palatal excision wounds and their connection to subsequent aberrations of maxillary growth. Their surgery involved excision of midline mucoperiosteum (but not bone) and medial advancement of flaps. Their findings confirmed the hypothesis that contraction during healing of open palatal wounds is critical to the onset of some maxillary growth disturbances. Later Kremanak et al. (1978b) studied the effects of vomer surgery on anteroposterior growth in the mid-face. The findings suggested that trauma to the area of the vomer, or absence of the vomer, were more important determinants of anteroposterior growth effects than palatal scarring.

Jonsson and Stenström (1978) followed on with a further animal experiment to test the effect of covering the raw nasal mucoperiosteum on one side of a surgically created cleft with an autogenous full-thickness skin graft. Their aim was to reduce the area of denuded palatal bone which had been identified by Kremanak (1978b) as the source of maxillary constriction. A comparison of the two sides of the defect revealed a small but consistent tendency for a more pronounced overall growth on the grafted side.

Several human studies have been concerned with the connection between facial growth and type of repair carried out. Palmer, Hamlen, Ross and Lindsay (1969) compared the results obtained using the simple closure (von Langenbeck) technique with those from the "pushback" or palate lengthening procedure. Because the patients in both groups were too young to assess all facets of
maxillary growth, they attempted assessment based on crossbite incidence. Both the incisor crossbite and buccal segment crossbite were significantly less in the simple closure group, which also had a higher incidence of anterior palatal fistulae. Wynn (1979) tested the influence of wedge osteotomy on the dento-maxillary complex. However, very few cases were presented to verify the excellent results Wynn claims for this technique.

Friede and Johanson (1977) carried out a follow-up study of patients treated with a vomer flap as part of a three stage soft tissue surgical procedure according to the method described by Pichler (1934). Although the authors lacked control cases which had not been treated with a vomer flap, the cephalometric results showed severe maxillary retrognathia, especially in patients who had been treated with vomer flaps accompanied by bone grafting. These results were sufficiently disappointing to convince them to exclude the use of vomer flaps in future surgery.

Since Graber's edict of 1949 in which early traumatic surgery was cited as the cause of growth deficiency and palate closure was advocated at the age of four to five years, other authors have also been concerned with the timing of surgery. Unfortunately, the easiest technical variable to alter is the age at surgery rather than a traumatic technique. One could conjecture that traumatic surgery performed at any time during childhood could cause facial growth inhibition. Kaplan (1981) stated the four recommended approaches:
1) delayed complete palate repair (12 to 24 months)

2) late complete palate repair (2 to 5 years)

3) early complete palate repair (3 to 9 months)

4) early soft palate repair (3 to 9 months) and delayed hard palate repair (5 to 15 years)

The controversy which exists regarding timing of repair seems to revolve around the relative importance of better speech versus maxillo-facial growth. Jolleys (1954), Robertson and Jolleys (1974), and Evans and Renfrew (1974) all demonstrate a direct correlation between early palate repair and improved speech quality regardless of surgical technique. Kaplan (1981) attributes this fact to the existence of an optimal time for speech development which requires a functional palate when palate-related sounds are first learned. As the repaired palate, like other tissues, has limited mobility due to postoperative oedema and scarring for three to six months, Kaplan advocates surgery many months prior to normal speech onset to allow for proper healing and scar maturation.

Perko (1979) also uses a two stage palatal closure with soft palate repair at eighteen months using Widmaier's technique of 1959, which is designed to spare mucoperiosteal mobilization and hard palate repair between five and eight years. The spontaneous narrowing of the hard palatal cleft following velar closure is
said to enable hard palate closure with less tissue mobilization. Good speech results, with little requirement for additional palate lengthening, are also claimed. Unfortunately, final results of hard palate closure are not presented.

Since 1975, due to dissatisfaction with their earlier results, Friede and Johanson (1980) have also employed a delayed closure of the hard palate until the stage of the permanent dentition eruption, notwithstanding that poor speech from nasal "air escape" may necessitate earlier intervention with need for possible artificial obturation. They advocate an atraumatic technique, with lateral excisions of the soft palate and denudation of bone being kept to an absolute minimum. The soft palate muscles are united in the midline after careful dissection of fibres from their abnormal insertions, resulting in soft palate lengthening without pushback. No significant cephalometric difference was found in the anteroposterior position of the maxilla or its length development in three year olds of this group when compared with those of the abandoned method of primary closure of the hard palate with a vomer flap and soft palate repair with pushback technique. Cast analysis showed significant occlusal improvement in the experimental group. See figure 32. Hotz and Gnoinski (1979) rely on analogous intrinsic potential of maxillary development present in all individuals to justify their delayed closure technique, in order to postpone the effect of any extrinsic influence on this "normal" growth.
Figure 32. Maxillary casts of one patient with complete bilateral cleft lip and palate at three different ages (7 months; 1 year 8 months; and 3 years.) Note the gradual decrease in size of the cleft in the hard palate after velar repair at the age of 6 1/2 months.

This case is from the series operated upon by the delayed hard palate closure technique described by Friede and Johanson (1980). Along with the gradual decrease in the size of the cleft of the hard palate, note the position of the anterior segment in relation to the maxillary segments. It could be argued that this so-called beneficial narrowing has made satisfactory repair of the anterior palate inordinately difficult and has also increased the risk of resultant residual fistulae which may be impossible to close.

Kaplan (1981) cites increased surgical morbidity and anaesthetic difficulties as the main reasons the earlier surgeons opted for delayed complete palate repair. As these problems have been largely overcome, he prefers combined early soft and hard palate repair since he feels the claim that early palate surgery causes greater facial deformity has not been verified.

One of the problems associated with delay of hard palate repair is persistent middle ear infection or "glue ear" due to Eustachian tube orifice malfunction tied to abnormal tensor palati attachments. Hagerty and Mylin (1981) have overcome this by using a pin-retained expandable palate prosthesis to simulate the
intact palate as well as to prevent maxillary arch collapse following lip repair. This is inserted approximately one month after birth and is worn continuously until hard palate repair at six years of age. Several cases showing satisfactory dental results are presented by these authors. However, it would seem that cooperation from both parents and child would be a difficulty with this approach, especially if further orthodontic treatment should be required during adolescence.

Subtelny and Nieto (1978) examined the effects of pharyngeal flap surgery on maxillary growth in cleft palate individuals. Analysis of longitudinal cephalometric radiographs were obtained on three separate populations. The pharyngeal flap group showed significant reduction in maxillary forward growth, but showed normal vertical maxillary dimensions with growth; whereas, the non-flap cleft group exhibited some reduction in vertical growth of the maxillary complex.

It seems clear that chronologic age in itself is a meaningless index. What really matters is the original dimensions of the defect, the quality and quantity of adjacent tissues available to repair it, and the size and shape of the pharyngeal port in which the repaired velum must function. The "quality of surgery" and possibly, the type and site of surgery, seem to be additional factors, according to a number of authors. Review of the literature reveals little attempt to correlate the outcomes of surgery with the original condition.
CHAPTER 9: Occlusion in Cleft Lip and Palate Patients

As Angle (1907) wrote, "Normal occlusion of the teeth is maintained first by harmony in the sizes of the dental arches through the interdependence and mutual support of the occlusal inclined planes of the teeth and second, by the influences of the muscles labially, buccally and lingually." There is general agreement that it is erroneous to discuss each of these factors separately and that rather we should be mindful of the interdependence of several forces. From what has been learned about the growth of the face and eruption of teeth, it is clear that the forces of occlusion are in a constant state of flux in the growing child and, further, that the growth potential is in itself an important determinant of arch form and occlusion.

Eruption of the incisor teeth and development of the supporting alveolar bone contribute to the anterior growth of the maxilla. It is of vital importance to the occlusion that eruption occurs freely in response to the musculature and the position of the mandibular teeth. Many techniques of palatoplasty leave an area of denuded bone close to the alveolar process and incisor teeth. This area rapidly heals by the formation of scar tissue. According to Ross and Johnston (1972) many of the supporting fibres of the teeth including part of the periodontal ligament extend for some distance into the surrounding mucosa and are caught up in this scar tissue. Thus, the initial retraction during healing results in a mild retrusion of the incisor segment, particularly in the unsupported region adjacent to the cleft, which may result
in a crossbite of one tooth or more. Subsequent tooth eruption and vertical development of the alveolar process are resisted by the scar tissue, and a posterior deflection of the dento-alveolar structures is induced. Ross and Johnston (1972). See figure 33.

Figure 33. Some of the variations in incisor teeth relations are illustrated. In the normal occlusion the incisors contact on closure and establish a stable vertical relationship. If the teeth are in crossbite, as in B, incisors will continue to erupt more than is normal, producing a deep overbite (excess vertical relationship). If, however, the tongue intrudes, as in C, the maxillary incisors will be prevented from overeruption, and a normal overbite results. If the maxillary incisors are linguually inclined, as in D, they do not come into proper contact, and both maxillary and mandibular incisors will overerupt, producing a deep overbite. Protrusion of the maxillary teeth (E) may also permit overeruption of the incisors, unless the tongue prevents mandibular incisor eruption and the lower lip prevents maxillary incisor eruption. Note that in these diagrams the mandible is in rest position and will close 3–5 mm into occlusal contact. From Ross and Johnston, (1972).
At the same time the repaired lip is exerting more than normal pressure against the teeth. A third factor which may act to inhibit normal anterior development is tongue posture. Unless the tongue can produce an adequate forward pressure on the incisor teeth and alveolar bone, in rest position as well as during function, compensatory anterior growth will not occur.

A final factor in anterior alveolar growth is the support provided by the mandibular incisors when a normal incisor overlap is present. When there is inadequate vertical development of the maxillary incisors or if they are posterior to the mandibular incisors, such an overlap does not occur. A retrusive tendency is liable to be expressed in such cases.

9.1 The Evaluation of Arch Form and Occlusion

Arch form has become one of the major considerations in the orthodontic treatment of the complete cleft lip and palate patient. The controversies regarding early orthopedic procedures, early bone grafting, and many operative techniques originated mainly from aspects related to the alignment of the segments of the upper jaw. However, only a few methods are available for analysis of arch form in cleft lip and palate infants. Isolated measurements of the intertuberosity width and measurements of the posterior and/or alveolar cleft width do not portray arch form.
Descriptive criteria such as "approximation without contact", "approximation with contact" and "overlap" are much too vague for comparative assessments. Evaluation of occlusion does not reveal the reaction of the upper arch during the early stages of treatment.

Stockli (1971) attempted to design a measurement system which would be suitable for use in longitudinal and comparative studies. His primary concern was to express, in numerical terms, the spatial relationship of the two maxillary segments in the frontal and sagittal planes, particularly in the region of the alveolar cleft. Stockli's method involved conversion of marks on study models into a two-dimensional system by using a photocopying machine similar to the method used by Huddart (1967). The measuring system was designed in relation to three anatomical landmarks (the interincisal point and the two bilateral tuberosity points) which were most reliably identified on the casts. Stockli found that the exact value of each measurement may not be of ultimate interest, but rather the range of values served as a basis for the establishment of a more refined grouping when large numbers of cases were evaluated.
Figure 34. Photocopy of maxillary cast. Prior to photocopy, crest of ridge, mesial and distal border of the cleft, tuberosities, canines, and incisive points are marked on casts.

I-T' - Incisor to tuberosity length of the affected side was the sum of I-C' and C'-T'.

I-T - Incisor to tuberosity length of the unaffected side.
From Huddart, (1967).

Berkowitz and Pruzansky (1968) and Berkowitz, Krischner and Pruzansky (1974) saw limitations with this method as it allowed only a two-dimensional representation of what is a three-dimensional problem. These authors asserted that this deficiency could lead to incorrect conclusions being drawn without the benefit of consideration of shapes and areas of the segments.
For this purpose stereophotogrammetry was developed for use with cleft palate casts. The principle involved juxtaposition of two binocular photographs so that the left eye sees the left photograph and the right eye sees the right photograph in proper relationship. This improves the depth perception and the effect of relief is created. A series of standardized projections can thus be created and reproduced for consecutive casts. These authors tried to quantify the variation in the amount of tissue in each palatal segment for a single type of cleft, as well as the increments in the area of the palatal shelves as a result of growth. See figure 35.

Figure 35. Arrangement for stereophotographs. From Berkowitz and Pruzansky, (1968).
Figure 36. Stereophotogrammetric tracings of palatal casts of dizygotic twins, age three months. The male twin demonstrates an unoperated complete left unilateral cleft lip and palate. The sister's palate is normal. From Berkowitz and Pruzansky, (1968).

Recently Berkowitz, Gonzalez and Nghiem-Phu (1982) have described an optical profilometer, a new instrument for three-dimensional measurement of cleft palate casts which automatically creates contour data via a computer and eliminates the need for the expensive data extrapolation equipment required for conventional stereophotogrammetry. Huddart (1969) and Huddart et al (1971) have also proposed methods of studying maxillary arch dimensions using computers.
Berkowitz, Krischer and Pruzansky (1974) found four questions central to the quantitative analysis of cleft palate casts:

"1) Are the palatal shelves intrinsically deficient, adequate or excessive in mass?

2) To what extent does the geometric relationship of the palatal shelves in one cleft compare with that of another in the same type of cleft? With other types of clefts? With normal palates?

3) To what extent are the palatal shelves displaced in space?

4) How are these parameters altered as a consequence of growth and surgical reconstruction?"

The following measurements were selected to describe the cleft palate as a changing and variable three-dimensional form:

1) Degree of curvature which reflected on the form of the arch

2) Fitted relative lengths - the relative length each palatal segment would have if the anterior cleft space was to be eliminated

3) Surface areas - comparisons were made between two-dimensional and three-dimensional surface areas to determine the reliability of diagnostic interpretation based on two-dimensional drawings or photographs
4) Slopes - the requirement to determine which slope measurement more truly reflects on the spatial relationship of the palatal segments for interpretive analysis.

5) Time sequence analysis - to demonstrate the rate that palatal form changes relative to time.

See figure 37.

Figure 37. A parabolic curve fit for a complete unilateral cleft lip and palate. These are the points used in the geometrical analysis. From Berkowitz, Krischner and Pruzansky, (1974).
The dental occlusion has been used in a number of studies to evaluate the results of cleft palate treatment, the success or failure of treatment usually being related to the frequency with which crossbites appear in the sample studied. Pruzansky and Aduss (1967). Two examples of occlusal classifications used for this purpose are those of Pruzansky and Aduss (1964) and Mathews et al (1970).

Pruzansky and Aduss 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

In contrast, Mathews et al divided the occlusion into:

1. Class A - where all segments of the maxilla are in normal occlusion with the mandible
2. Class B - (1) the tooth bordering the cleft on the lesser segment is in lingual occlusion
3. Class B - (2) normal occlusion of the greater segment but lingual occlusion of the lesser segment
4. Class B - (3) perfect maxillary arch but it is too small

5. Class C - an overall Angle's class III occlusion of all segments of the maxilla and in addition, collapse of some part of the small maxillary arch.

Although both classifications describe the occlusion, the categories used are so dissimilar that effective comparison of results is extremely difficult. Nevertheless, such comparisons must be made if cleft lip and palate treatment is to be eventually rationalized. Even if all investigators used the same system in the interest of comparability, two further problems emerge:

1) What is the most satisfactory classification to use?

2) How reliable would this be in the hands of different observers who might find difficulty in establishing common assessment criteria?

Huddart and Bodenham (1972) investigated the reliability of occlusal assessments by different observers using the same classification and examined the consistency of the results obtained by each observer when making repeated assessments of the same case. The descriptive classification of Pruzansky was compared with a numerical classification developed by the authors. They found observer reliability and consistency were very similar no matter which classification was used, although the numerical classifica-
tion was found more suited for future studies due to its facility for ranking different malocclusions in order of severity and ease of statistical handling.

Figure 38. Arch alignment. Diagrammatic illustration of good segmental alignment. There may or may not be actual contact of the greater and lesser segments. The lower diagram illustrates poor segmental alignment with a contracted arch form. There is overlapping and contact of the greater and lesser segments. From Huddart and Bodenham, (1972).
Figure 39. Numerical classification. Subdivision of the deciduous maxillary arch into three segments. Each buccal segment consists of the canine and first and second molars whilst the labial segment consists only of the two central incisors. From Huddart and Bodenham, (1972).

Figures 40, 41 and 42 (below). Numerical classification. Occlusal scoring of the deciduous maxillary arch.

Figure 40. Scoring of the labio-palatal relationship of the canines.
Figure 41. Scoring of the bucco-palatal relationship of the molars. Both the first and second deciduous molars are assessed for purposes of classification.

Figure 42. Scoring of the anteroposterior relationship of the central incisors. A large upper incisor overjet may warrant a score greater than +1.
Bergland (1967) found that improved surgical techniques instituted in the Oslo area around 1950 caused a marked reduction in subsequent orthodontic problems. He compared outcomes in terms of anterior and posterior crossbites with those obtained by Pruzansky and Aduss (1964) and Kling (1964) and in spite of tabulation differences which made these results not directly comparable, found in favour of the Oslo methods. Bergland recommended early initiation of orthodontic treatment as soon as the permanent incisor can be banded, with expansion and alignment with various retention appliances placed throughout up until the age of nineteen when a bridge can be inserted. It would seem that the "improved surgery" in the Oslo area may only save treatment time in the first five to seven years of life. The questions I would ask would relate to the significance of the orthodontic problems Bergland is correcting. Is a crossbite a significant enough problem to warrant orthodontic intervention at such an early age and, if so, what advantages are gained by correcting this crossbite and being faced with an indefinite retention period because an adequate natural occlusal locking relationship cannot be developed to hold the teeth in the correct position? Bergland also found no evidence that the minor cleft segment is underdeveloped and lags behind the non-cleft side because it has been disconnected from the nasal septum.

Friede and Johanson (1977) used similar methods to Huddart and Bodenham (1972) to study the occlusions of patients with complete
bilateral cleft lip and palate or with complete unilateral cleft lip and palate who had been treated using a vomer flap. Some of the group had undergone an additional bone graft. Generally, the sagittal occlusion reflected a maxillary retrognathia with a higher frequency of class III occlusions than the non-cleft population. The bone grafted cases were the most severe. The dental arch measurements also demonstrated inferior results for the vomer flap cases, as the molar width was greater while the canine width and arch length were smaller. The formation of a bony bridge across the former cleft seemed to have a deleterious effect on maxillary development in the bilateral cleft group, in which there was a more frequent occurrence of class III relationship than cases without this bone formation. It would be difficult to identify the vomer flap as the cause of these poor results since no control cases were presented by these authors. Also, there is always the possibility of some inherent unavoidable minor variation in treatment even within groups; a factor which may possibly have operated here.

Ponitz and Spyropoulos (1979) suggested that absence of posterior crossbite in non-treated cases is due to a compensatory effect of adjustment in the direction of dental eruption and alveolar apposition. This is caused by the unrestricted influence of tongue pressure, which makes up for any mild maxillary deficiency. These authors also warned against early palatal closure which may lead to maxillary collapse, although they presented very little evidence to support this.
Ross and Johnston (1972) suggested that the widespread dental abnormalities associated with cleft lip and palate are due to the deficiency of facial mesenchyme, which seems to play a major part in the formation of the primary and secondary palate, with the deficient mesenchyme providing poor support for each developing tooth germ. Foster and Lavelle (1971) found a generalized reduction of tooth size in both upper and lower jaws with cleft lip and palate.

Sofaer (1979) found abnormally high levels of tooth size asymmetry in cleft lip and palate which were most marked in the upper lateral incisor region, but were not restricted to the vicinity of the cleft nor the upper jaw. Sofaer postulated that this asymmetry results from a generally high level of developmental instability throughout cleft lip and palate conditions which may also have an effect on occlusal development.

Dahl and Hanusardottir (1979) compared the prevalence of malocclusion in the primary and early mixed dentition of Danish children born with complete cleft lip and palate with a non-cleft group. The unilateral group had undergone lip repair at the age of two months concurrently with hard palate repair with a two-layer palatovomeroplasty. In the bilateral group, the left side of the lip had been repaired using the Blair-Brown technique with simultaneous palatovomeroplasty on the same side at two months, followed by the right side repair at four months. In both cases the remainder of the palatal cleft had been closed by a Wardill pushback procedure at two years.
The cleft palate group was characterised by high frequencies of dental anomalies such as supernumerary teeth, aplasia, tooth malformations, and rotations and ectopic eruptions. No cleft child was found to be malocclusion free. There were high frequencies of mesial molar relationship and mandibular overjet, indicating a difference in the sagittal jaw relationship between the cleft group and normal group. The frequency of posterior lingual crossbite was so high that the authors decided to change from the two-layer palatovomeroplasty to a one-layer vomer flap for hard palate repair.

Subdivision of the cleft group by type of cleft gave a more detailed impression of the dental occlusion. The group of unilateral clefts was characterised by unilateral lingual crossbite, high frequency of midline deviation, mesial molar occlusion and mandibular overjet. Frequencies for extreme maxillary overjet and openbite were low. A tendency was also seen for a difference in the sagittal molar relationship between the cleft side and non-cleft side, with distal molar occlusion being more frequent on the cleft side. The authors explained this as a result of a primary difference in the sagittal position of the two segments of the maxilla and also, as a result of secondary changes, such as lateral shifting of the mandible. This was caused by lingual crossbite on the cleft side, medial rotation of the cleft segment subsequent to surgery, and tipping of the teeth towards the cleft on the affected side.
The patients with bilateral clefts were characterised by a pronounced variability of the incisor occlusion. In the sagittal direction this was demonstrated by the high incidence of both mandibular overjet and extreme maxillary overjet. The vertical incisor occlusion was also highly variable. These findings were interpreted to reflect the great variability in the primary position of the premaxillary segment in complete bilateral cases, as well as the marked changes in the position of this segment subsequent to the primary surgery. The frequency of midline deviation to the right and left side was equal, although the frequency of crossbite of the buccal teeth was higher on the side operated on first. In this type of cleft the midline registration primarily indicates the position of the "premaxilla" whereas in unilateral cleft lip and palate, it reflects more a dento-facial asymmetry.

Jonsson, Stenström and Thilander (1980) compared the increase in length and width of the maxillary arch, from the time for lip surgery to that of full eruption of the deciduous dentition, and the dental arch dimensions and occlusion at five years of age in children in whom palatal closure was made using the modified vomer flap covered with an autogenous skin graft with those children in whom the closure of the cleft in the hard and soft palates was made using a modified von Langenbeck procedure. Interestingly, these authors made an attempt to validate their comparisons by selecting cases so that the status preoperatively was as identical as possible between pairs of children one from
each group. The initial morphology of the cleft and the surrounding structures are essential factors in determining arch form (Pruzansky and Aduss 1967). In the children with crossbite occlusion, canine crossbite was more frequent in the group with the vomer flap, while buccal crossbite was more frequent in the group with the Langenbeck procedure. In this investigation all raw surfaces in the vomer flap group were covered by a well adapted skin transplant. Therefore, it is likely that a smaller amount of scar tissue developed in this group than would have developed if the raw surfaces had been left uncovered or covered with a palatal flap. The growth of the alveolar arches was similar in both groups, although there was a tendency to better occlusion in the vomer flap group than in the Langenbeck group. However, it must be remembered that the study only went as far as age five years. There is no idea of changes after that time as frequency of crossbite increases from the deciduous to the early mixed dentition. (Bergland and Sidhu, 1974). The results from Jonsson et al (1980) showed a less detrimental influence on arch dimensions and occlusion than operations involving pushbacks. However, considerable changes in the facial morphology and the occlusion may occur by the time these children reach adulthood. Therefore, further observation of these groups at a later stage is needed to evaluate the influence of this vomer flap procedure on the growth of the middle face.

Jonsson et al do not mention the speech results obtained with this technique. After all, one of the main reasons for the pushback procedure is improved speech brought about by palate
lengthening. These speech results cannot be divorced from the results on occlusion, although some authors neglect the importance of this "overall evaluation".

Dahl, Hanusardottir and Bergland (1981) made an inter-centre comparison of the types and degrees of malocclusion found in children with complete unilateral cleft lip and palate. The children were operated on at two Scandinavian cleft palate centres, where three different surgical procedures were used. The types of repair tested included:

1. Millard lip repair/Langenbeck palate repair (Norwegian cases)
2. Tennison lip repair/Wardill palate repair (Danish cases)
3. Le Mesurier lip repair/Langenbeck palate repair (Norwegian cases)

The transverse occlusion and sagittal incisor occlusion were documented by one person according to a standard method. The observed differences were related to:

1. Type of operation performed
2. Timing of operation
3. The number of operations (which was the same in all cases)
4. The individual approach of the surgeon
In the Danish group the transverse occlusion results were generally poorer than the other groups. This was thought to be related to the hard palate closure by a two-layer palatovomeroplasty at the same time as lip repair, which was associated with greater surgical trauma with more scarring. It has been shown that this procedure may have an adverse effect on the transverse growth of the maxilla. (Dahl, 1979). For these reasons, the surgical procedure in Denmark was changed some years ago to one-layer closure by a vomer flap. In the Norwegian groups, only a one-layer closure by a vomer flap had been used for repair of the anterior part of the hard palate. This procedure, which was assumed to be less traumatic, was also performed three to four months later than the surgical intervention in the Danish group. The different technique used for closure of the posterior part of the palatal cleft was thought to contribute to the difference in transverse occlusion. In all groups simultaneous closure of the hard palate and lip reduced the number of dissections needed at the final palatoplasty.

Palmer et al (1969) and Blockma et al (1975) have previously reported a higher risk for adverse effects on the dento-alveolar development from a pushback procedure than a simple closure. The Wardill procedure may have contributed to the higher frequency of lingual crossbite involving the posterior teeth on the cleft side in the Danish group. Posterior lingual crossbite in patients with complete clefts of lip and palate can be related to either
displacement of segments subsequent to the primary surgical closure, long-term inhibition of transverse maxillary growth, or a developing effect of poor lip function or contraction. At the early stage of occlusal development investigated in the present study, it can be assumed that displacement of segments had been the major reason for the observed changes in transverse occlusion. During future growth periods, the occlusions can be expected to deteriorate. (Bergland and Sidhu, 1974).

Differences were also found in the type of malocclusion in the incisor region between the Danish and Norwegian children. The authors explained the asymmetric localization of the anterior crossbite in the Danish group by the more extensive surgical procedures in the anterior part of the hard palate. Also, the different techniques used for lip repair might have influenced dento-alveolar development in a different way. The Millard procedure has been thought to produce a lip which is tighter in its lower third than is the case with either the Tennison or Le Mesurier techniques which move more tissue into the lower third of the lip. (Lindsay, 1972). This possible difference was not reflected in the incisor occlusion in this study. As both Norwegian groups had higher frequencies of mandibular overjet, and the crossbite relationship of incisors frequently extended across the midline to the non-cleft side, the authors concluded that postponement of lip repair for three to four months in the Norwegian groups had no obvious beneficial effect on incisor occlusion.
Bomba (1979) compared a variation in surgical procedure used by Dey (1974), after Muir (1966), with a standard method of repair used in unilateral cleft lip and cleft palate. The results of the treatment were evaluated by assessing the occlusion using the method of Huddart and Bodenham (1972). The control group involved a primary repair, using the Tennison procedure for the lip and medial advancement of the alar cartilage with z-plasty, and closure of the anterior palate with a septal flap tucked into the cleft margin. The second repair involved closure of the residual posterior cleft of the hard and soft palate, including palatal lengthening using the Wardill V-Y procedure. The study showed that mean incisor and buccal crossbite scores for the triangular flap group were all less than the corresponding scores for the standard treatment group, though the differences were not statistically significant. Further, the triangular flap group's incisor segment score compared favourably with studies utilizing periosteoplasty, bone grafting, orthopedic appliances, and the vomer flap technique.
Summary and Conclusions

The literature on orthodontic evaluation of outcomes of cleft lip and palate surgery has been reviewed. The problems and limitations of rehabilitation of children suffering from this complaint have been identified. The methods of evaluation of occlusal results have also been described. For the time being Huddart's method seems to be the most widely used and accepted, particularly in terms of measurement of crossbite relations and alignment.

It is clear that rapid progress has been made in recent years toward an understanding of the origins of facial clefts. Many new avenues of potentially fruitful investigation are now apparent, and hopes for eventual preventive measures are warranted. Unfortunately, it is unrealistic to assume that the clinical problems will be appreciably lessened in the immediate future. Increased efforts to improve the treatment of affected individuals are still needed.

Modern habilitative programmes are excellent, but there are some deficiencies. Not all children with cleft lip and palate participate in these programmes, and even those who do have many residual problems. If we accept that the aims of treatment are to produce normal appearance, normal speech, and normal dental occlusion, we are also forced to accept that the ultimate result will be a compromise solution. The development of present day methods of treatment has been largely empirical. From the clinicians' viewpoint these methods have been rather successful and,
by and large, fewer procedures are necessary. Improvements in treatment procedures have reduced dramatically the number of severely deformed individuals with un intelligible speech, common only a few decades ago. This review only serves to confirm the advantages of multidisciplinary clinics.

It must be admitted, however, that some cosmetic and speech results are less than satisfactory from the patient's point of view, particularly in relation to alar collapse and the cosmetic result of nasal repair. One could conjecture that this may be the result of unreasonable expectations on the part of the patient, or from failure to treat according to the patient's major needs.

It seems that cosmetic and speech results have gone ahead at a more rapid rate than dental results, although more attention is devoted to this aspect now. The absence of recognition of a mechanism for linking the outcome to the severity of the original defect may also be a factor which results in excessive therapeutic effort, with diminishing return for each attempt. As the point was so clearly made in the Jubilee Cleft Palate Clinic at the Royal Alexandra Hospital for Children in Sydney during 1981, "When is enough enough?"
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