AUTOGENOUS SECONDARY ALVEOLAR BONE GRAFTING

IN THE TREATMENT

OF

CLEFT LIP AND PALATE

by

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INTRODUCTION

Cleft palate, with or without cleft lip, is one of the more common congenital malformations in man. Fogh-Anderson (1942) (cited in Fogh-Anderson 1961) reported a birth incidence of 1.45 per thousand live births for a Danish population. He suggested that this was a minimum value, since some cases may have escaped notice or registration during the 10 days the infants remained at the maternity clinics. The incidence of this condition appears to be fairly uniform in various parts of the world, although there are some exceptions, such as the Negro, in whom it appears to be significantly lower than for the Caucasian. Grace (1943) reported an incidence of 1/800 (1.25 per thousand) in Pennsylvania. Negroes accounted for only 3.6 percent of the cleft infants although they did represent 5.5 percent of total births. Ingalls, Taube and Klingberg (1964) reported an incidence of approximately 1/800 (1.25 per thousand) for "whites" and 1/1500 (0.67 per thousand) for "non-whites", also in a U.S. study. MacMahon and McKeown (1953) in an English population, found an incidence of 1.30 per thousand total births in the period 1940-1950. Rank and Thomson (1960) reported a relatively high incidence of 1.66 per thousand, in Tasmania during the period 1945-1957. Chi and Godfrey (1970) reported an incidence of 1.21 per thousand live births\(^1\) in New South Wales for the period 1964-1966.

Fogh-Anderson (1961) reported on the changing incidence of cleft lip and palate in Denmark from 1939 to 1957. Dividing this period into four five-year intervals, he found a progressive increase in the number of cleft lip and palate cases per one thousand live births, from 1.45 in 1938-1942, to between 1.7 and 1.8 in the period 1953-1957. Further, reductions in post-natal mortality were also resulting in an increasing number of infants with this condition presenting for corrective surgery. Jensen, Kreiborg, Dahl and Fogh-Anderson (1988) reported a further increase in incidence, to 1.89 per thousand live births for the period 1976-1981. Fogh-Anderson felt that the rise was too

\(^1\) 1.31 per thousand for total births.
great to be attributed solely to errors in the earlier estimates of incidence. He proposed several factors which may be responsible, such as decreased neonatal mortality, drugs, as well as increased frequency of inter-marriage and childbirth in people with the cleft condition, due to better social acceptability and fertility. Although many factors have been associated with the cleft lip and/or palate condition, from a practical preventive aspect, the aetiology remains undetermined.

As Ferguson (1981) so rightly points out, the statistics do not convey the full magnitude of human suffering and the health-care costs related to this condition, until it is realised that between fifteen thousand and eighteen thousand new cleft patients are born each year in the USA alone! In the absence of any probability of actively reducing the incidence of cleft lip and palate in the foreseeable future, the need for corrective therapy is greater than ever.

Modern plastic surgery has achieved wonders of repair and rehabilitation, particularly in relation to the soft tissue defects of lip and soft palate. However, correction of the combined lip and palate deformity is still a major feat and the results are far from perfect. Bridging the bony defect of hard palate and alveolus and achieving a normal arch form without jeopardising maxillary growth, have posed a challenge to surgical and orthodontic ingenuity over the last thirty years.

The stimulus for the incorporation of bone grafting in cleft protocol appears to have been the universal observation that the unsolved problems of maxillary configuration and instability of the bony segments led directly to malocclusion of the dental arches, poor speech and an unsatisfactory cosmetic appearance (Horton et al 1964).
Pickrell et al (1968) stated that bone grafting is based on the concept that the cleft defect involved not only a lack of closure in the lip, alveolus, maxilla and palate, but that there appear to be deficiencies in substance of both bone and soft tissue. It seemed logical therefore, to attempt to eliminate the bony defect by placement of a bone graft under the repair of the underdeveloped soft tissues, especially of the lip.

Schultz (1964) stated the aims of bone grafting of the hard palate more specifically:

a. To provide an improved, stable dental arch.

b. To provide stability to the lateral maxillary segments.

c. To provide more normal palatal resonance for speech.

d. To provide a bony framework for surgical improvement in facial features (specifically the typical depressed alar deformity and associated apparent mald development of portions of the middle third of the face).

Isolated attempts at bone grafting in cleft patients had occurred as early as Lexer (1908) and Drachter (1914). However, Axhausen (1952) is credited with heralding the "osteoplastic era" (Koberg 1973). Axhausen had written that all attempts to induce bony healing through excision of mucosa in the area of narrow clefts and "freshening" the underlying bone surface had proved futile. He had therefore proclaimed that induction of bony healing between the premaxilla and lateral fragments presented the "final problem" in cleft treatment.

Bone grafting at the time of primary palate repair was initially thought to hold great promise for definitive correction of the bony defect. However, an increasing number of retrospective studies in the mid to late 1960's showed significant growth retardation of the maxilla with this technique. Consequently this approach has been abandoned at most centres, in favour of a delayed "secondary" bone grafting of the alveolar/palatal defect.
It is the aim of this treatise to investigate the hypothesis that secondary alveolar bone grafting is effective in achieving and maintaining the following objectives:

a. Formation of a continuous alveolar ridge in the maxilla with provision of bony support for unerupted teeth in the line of the cleft as well as teeth adjacent to the cleft.

b. Stabilise the maxillary alveolar segments after alignment by orthopaedic/orthodontic means into a normal arch form.

c. Improve the skeletal support of the alar base, which in turn would facilitate soft-tissue surgery to correct nasal asymmetry and facial contour.

d. Eliminate vestibular mucosal recesses and residual oronasal fistulae in the maxilla.

The terminology proposed by Boyne and Sands (1976) will be used to classify the grafting procedures:

a. Primary bone grafting
   - bone grafting performed in children less than 2 years old.

b. Early secondary bone grafting
   - bone grafting performed in children between 2 and 5 years old.

c. Secondary bone grafting
   - bone grafting performed in patients between 6 and 15 years old.

d. Late secondary bone grafting
   - construction performed in physically mature adults.
In order to achieve the aim of the treatise, it will be necessary to:

a. Describe the classification and anatomy of the various configurations of the cleft lip and palate condition.

b. Provide an overview of cleft lip and palate treatment. This will include a review of its evolution as well as a description of modern concepts of treatment.

c. Review the histology of bone grafts. This will include human and animal studies investigating the relative efficacy of a variety of grafting techniques, as well as bone from different sources.

d. Review the evolution of bone grafting in cleft lip and palate treatment.

e. A detailed analysis of the literature relating to the various techniques of secondary bone grafting and their results.

If the hypothesis is to be accepted as valid, evidence must exist in the literature that secondary alveolar bone grafting can achieve and maintain its stated objectives without any significant complications, either in terms of operative morbidity and mortality or in terms of the normal growth and function of the facial structures.
CHAPTER ONE

THE CLEFT LIP AND PALATE CONDITION

1.1 The anatomy of the facial skeleton in cleft lip and palate

Pruzansky (1953) presented one of the most detailed and systematic descriptions of the cleft condition, based on the study of more than 350 cases. The present description will draw heavily from the work of Pruzansky, as well as that of Latham (1977).

1.1.1 Clefts of the lip and alveolus

A cleft of the lip and alveolus may vary from a minimal defect involving only the vermilion border, to a complete defect extending from the vermilion border to the floor of the nose and clefting the alveolus. The defect may also exist as a submucous cleft in the muscle band of the upper lip, bridged only by mucous membrane, skin and fibrous connective tissue.

The nasal alar cartilage on the side of the cleft is displaced and flattened to a greater or lesser degree, depending on the extent and width of the cleft. The tip of the nose tends to deviate to the non-cleft side.

The cleft may be unilateral or bilateral and the latter may be symmetrical or asymmetrical in the extent of clefting. In bilateral clefts, the median portion of the lip contains the philtrum and is attached to the columella and the premaxilla. In bilateral complete clefts of the lip and alveolus, the premaxilla protrudes considerably forward of the facial profile. It is attached to a stalk-like vomer and the nasal septum. The columella appears deficient and the alar cartilages are flattened on both sides (Fig.1).

\[1\] In man, except for a brief period in the embryo, the "premaxilla" does not exist as a separate entity. However the term is retained with reference to cleft lip and palate conditions due to its descriptive convenience and homology with experimental animals. In man, the term "premaxilla" refers to that part of the maxilla anterior to the incisive foramen and mesial to the canine teeth. (From Latham 1976).
Pruzansky (1953) reported two cases of submucous cleft of the alveolus in the absence of a lip cleft. However, he described both cases as "bizarre", with associated multiple anomalies elsewhere in the body.

Fig. 1  A. Frontal view of unoperated complete bilateral cleft of lip and alveolus. (from Pruzansky 1953).

Fig. 1  B. Profile view of unoperated complete bilateral cleft of lip and alveolus. (from Pruzansky 1953).
1.1.2 Cleft lip and cleft palate

Clefts of the lip and palate may be unilateral or bilateral, complete or incomplete.

1.1.2.1 Unilateral cleft lip and palate

Varying degrees of clefting in the lip and palate can exist, in a wide range of combinations. In a complete unilateral cleft of the lip and palate, a direct communication exists between the oral and nasal cavities on the cleft side. There can be substantial variation in the degree of palatal shelf separation. There is an associated skeletal deformity, of which the prominent features are lateral displacement of the non-cleft maxilla, malformation of the nose and lateral distortion of the nasal septum (Latham 1969, 1977) (Fig.2).

i. Premaxillary segment and nasal septum:

The premaxillary segment, in the frontal view, tilts upward into the cleft.\(^1\) The cartilagenous nasal septum is also bent in the same direction. The nostril on the non-cleft side is constricted and may be functionally occluded. The constriction is due to a combination of deviation of the nasal septum and approximation of alar base and columnella. The alar of the cleft side is usually stretched and flattened (Fig.2).

ii. The lip and columnella:

In the complete unilateral cleft lip, the anatomy of the orbicularis oris muscle is disrupted. The muscle fibres proceed horizontally from the corner of the mouth toward the midline and turn upward along the margins of the cleft. The muscle fibres terminate beneath the alar base in the lateral

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\(^1\) If this is left uncorrected, the incisor teeth in the premaxillary segment erupt with their occlusal plane tilting up into the cleft in the same manner.
segment and beneath the base of the columella in the medial segment. Most fibres attach to the periosteum of the maxilla, but a few fibres blend into the sub-epithelium. Where the cleft is less than two-thirds of the lip height, the muscle fibres above the level of the cleft remain intact. A protrusion of excess muscle may be seen and palpated on the lateral aspect of the cleft, due to heaping up of the disrupted fibres. However, the medial segment tends to be under-developed (Fára 1977).

**Fig. 2** A. Unoperated complete unilateral cleft of the lip and palate on the right side. (from Pruzansky 1953).
Fig. 2  B. Varieties of unoperated complete clefts of the lip and palate. By classification and general description, each one of these clefts is alike. Clinically significant differences are apparent in the width of the cleft and in the spatial relationship of the palatal processes. (from Pruzansky 1953).
iii. Vomer and palatal process:

The palatal segment on the cleft side is often tilted medially and superiorly into the cleft. The vomer is deviated laterally at its line of attachment to the palatal process of the non-cleft side. This deviation may be so severe in some cases that the vomer assumes a nearly horizontal position at its inferior margin.

1.1.2.2 **Bilateral cleft lip and palate**

The incomplete bilateral cleft lip and palate can be either symmetrical or asymmetrical with regard to extent of clefting. In the complete bilateral cleft lip and palate, both nasal chambers are in direct communication with the oral cavity. The palatal processes are divided into two equal parts and the turbinates are clearly visible within both nasal cavities. The nasal septum forms a midline structure that is firmly attached to the base of the skull but is fairly mobile anteriorly, where it supports the premaxilla and columella (Fig.3).

i. Premaxilla:

There is a malformation of the premaxilla characterised by its protrusion relative to the nasal septum. The columella is non-existent, with the lip attaching directly to the nasal tip (Fig.4 A, C). The basal bone of the premaxilla articulates with the cartilagenous nasal septum superiorly and vomer posteriorly. In normal structure the alveolar process of the premaxilla is inferior to the basal component. However, in the bilateral cleft condition the alveolar component is anterior to the basal component, in horizontal arrangement (Fig.4 B, D).
ii. The lip and columella:

The lip moiety in the medial segment contains only collagenous connective tissue (Fára 1977). It is therefore grossly deficient in bulk and lacks the features (e.g. philtrum) normally produced by muscle.

Although the columella would appear to be absent clinically, in anatomic terms it may be present in that the medial crura of the alar cartilages appear to occupy a normal position relative to the tip of the nose and the nasal septum (Latham 1977). There is, however, a deficiency of columellar skin which complicates the re-establishment of normal anatomical relations during treatment.

Fig.3  A variety of unoperated bilateral clefts of the lip and palate. Note variation in size, shape and position of the premaxilla. (from Pruzansky 1953).
A. Profile of normal 1 month old male infant.
B. Diagram of skeletal and soft tissue relations of normal neonatal infant.
   Medial orula (MC) of alar cartilages (AC) support the columnella and nose (N). The lip lies inferior to
   the exposed anteroinferior angle of the nasal septum. The anterior nasal spine (ANS) and
   alveolar process (AP) are posterior to the septal angle. Cut surface of palatal bone (Pal); vomer
   (V).
C. Profile of 2 month old male infant with complete bilateral cleft lip and palate.
D. Diagram of skeletal and soft tissue relations of neonatal infant with bilateral cleft lip and palate.
   Alveolar process (AP) is superimposed upon medial orula (MC) with lip (L) displaced anteriorly.
   Anterior nasal spine (ANS) is in fork of MC and adapted to the anterior border of the nasal
   septum (NS).
E. Lateral radiograph of infant in C. showing the premaxillary deformity in bilateral cleft lip and
   palate.
iii. Nasal septum and vomer:

The cartilagenous nasal septum is reinforced inferiorly by a stem of bone which is the anterior part of the vomer. Postero-superiorly the vomer articulates with the sphenoid bone.

iv. Maxillary segments:

The gum pads of the maxillary segments of the infant with bilateral cleft are covered with gingival mucosa. They are demarcated from the palatal mucosa on their medial aspect by a groove corresponding with the position of the palatal alveolar process, to which the oral epithelium has fibrous connections. The developing teeth are situated lateral to the above-mentioned groove; the area medial to it corresponds to the palatal process of the maxilla and the palatine bones, which are covered by thick palatal mucosa.

The arch form generally appears normal at birth, but medial collapse of the maxillary segments occurs soon after. The medial aspect of the palatal processes are often tilted superiorly into the cleft.

1.1.3 Cleft palate

The cleft may involve only the soft palate, both soft and hard palates, but almost never the hard palate alone. The lip and alveolar process are not involved. The cleft may extend forward from the uvula to a varying degree; from a bifid uvula to a V-shaped cleft extending throughout the hard palate to the incisive foramen (Fig.5). In the latter instance the uvular processes are shortened and distorted in an anterior direction. In extensive clefts of the hard palate, the nasal chambers communicate directly with the oral cavity. Usually the cleft occurs on both sides of the nasal septum. However, occasionally unilateral clefting may occur.
Deficiency of mucosa and bone is the main feature of clefts of the hard palate. In the soft palate, deficiency of mucosa is combined with shortening of the velar musculature, which has abnormal sites of insertion. Submucous clefts of both soft and hard palate, of varying degree, may also occur. In these cases the mucosa is intact, but deficiency of bone and/or muscle exists.

Fig. 5  A variety of unoperated cleft palates. The cleft may extend, in varying degrees, as far forward as the nasopalatine foramen. In its lateral dimensions, the cleft may be wide or narrow, V-shaped or pear-shaped. (from Pruzansky 1953)
1.1.3.1 Maxillary arch dimensions

Harvold (1954) studied a group of 67 children with cleft lip and palate, 44 of whom were between 7 and 14 years of age. Using postero-anterior cephalometric radiographs, he reported that the position of the zygomatic bones was normal in spite of the severe malformation in the maxilla and palatine bones. The maxillary sinuses were found to develop normally in a lateral direction even though nasal stenosis on the cleft side was a frequent finding. Abnormal asymmetries in the sinuses were found only in those parts extending into the alveolar and palatine processes. Harvold felt that the size of the sinuses indicated that the corpus maxillae were not abnormal to any significant degree. He concluded that the deformity of the facial skeleton is localised mainly to the alveolar, palatal and premaxillary processes, as well as the nasal septum. Critical analysis of Harvold's findings is difficult however, due to the confusing manner in which the results were presented.

Subtelny (1955) also found no difference in bizygomatic width between a cleft and non-cleft sample. However, the cleft subjects (with the exception of those restricted to the premaxilla) did have a small but significant increase in nasopharyngeal width compared to the normal group. Subtelny tentatively suggested that this may be caused by unbalanced pterygoid pull, due to an aberrant tensor palatini function.

Coupe and Subtelny (1960) performed a cephalometric laminograph study of 127 cleft palate subjects, 3 years of age and younger, compared to 50 non-cleft control subjects. They found no significant difference in either mean palatal dimensions or width of nasal cavity between the controls and subjects with clefts of the lip and/or alveolus. There was a small but significant deficiency in tissue of the hard palate in the subjects with cleft lip and palate and with isolated palatal clefts. The deficiency was greatest in bilateral clefts. The lateral displacement of segments was greatest in the unilateral clefts. However, the relatively high standard deviation indicated a large variation in individual cases. Furthermore, in the unilateral clefts, the decrease in palatal dimensions were no longer significant after 1 year of age. There were no significant differences between the cleft and non-
cleft samples at the level of the zygomatic arch. Peyton (1934), in a comparison between 49 cleft and 91 non-cleft children, had also reported slight but significant reduction in hard palate tissue in the cleft subjects (cited in Coupe and Subtelny 1960, Huddart et al 1961).

Huddart et al (1961) used "highly accurate graphic reproductions" of models of unilateral cleft palates in the transverse and sagittal dimensions. A comparison between 30 cleft and 30 normal new-born subjects led to the conclusion that the posterior arch width is significantly (albeit slightly) greater in the cleft palates. More than half the width of the cleft was attributed to lateral displacement of segments, about 29 percent to tissue deficiency and the remainder to an increase in the slope of the palatal shelves. Huddart et al felt that the major factor in the lateral displacement of the palatal segments was the unopposed pull of the pterygoid muscles, in the absence of a functional tensor palatini. The action of the tongue in the cleft was also recognised as a significant factor.

1.1.4 Dental anomalies

Agenesis, supernumerary teeth, concurrent agenesis and supernumerary teeth and anomalous morphology of teeth are seen in individuals with cleft lip and alveolus, both with and without the involvement of the palate (Bohn 1963, Zilberman 1973). The supernumeraries are customarily termed "fissural teeth". They may occur either in the medial segment or the lateral segment and on rare occasions, in both segments.

Bohn (1963) reported on an extensive study of dental anomalies in lip, alveolar and palatal clefts.
A. 281 patients with cleft lip and alveolus, with or without cleft palate, were examined with reference to the incidence of fissural teeth.  

i. A medial fissural tooth developed in 47.0 percent of the clefts in the primary dentition and in 25.6 percent in the secondary dentition.  

ii. A distal fissural tooth developed in 75.6 percent of the cases in the primary dentition and in 44.3 percent in the secondary dentition.  

iii. Agenesis of the fissural teeth was found in 14.3 percent in the primary dentition and 45.5 percent in the secondary dentition.  

iv. Both medial and distal fissural teeth were present in 36.9 percent in the primary dentition and 14.3 percent in the secondary dentition.  

v. The number of fissural teeth usually decreased with increasing size of cleft. However, this reversed in complete bilateral clefts, where there was an increase.  

vi. Sex of the subject and side on which cleft occurred, did not significantly influence the pattern of tooth incidence.  

B. 37 patients with cleft lip, without corresponding alveolar cleft, were studied with reference to the number of teeth adjacent to the cleft area.  

i. In the primary dentition, a supernumerary tooth was found in 60 percent of cases and a single lateral incisor in 40 percent.  

ii. In the secondary dentition, the relative frequencies of occurrence were reversed, with 40 percent showing a supernumerary tooth and 50 percent a single tooth.  

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1 Bohn (1963) defined "fissural teeth" as the dental formations which occur instead of the upper lateral incisor in the cleft area.
iii. Agenesis of the lateral incisor was seen only in the secondary dentition and then in about 10 percent of patients.

iv. The number of supernumerary teeth in the primary dentition increases with radiographically demonstrable signs of a bone defect just above the alveolar process.

C. Three groups of children were examined for evidence of oligodontia outside the cleft area in the secondary dentition.

i. In a group of 22 patients with either cleft lip or cleft lip and alveolus, 1 child (4.5 percent) had oligodontia; a prevalence similar to the general population.

ii. In a group of 176 children with cleft lip and palate, 42.6 percent showed oligodontia.

iii. In a group of 31 children with palatal clefts, 32.3 percent had oligodontia.

iv. In most instances the number of teeth missing were either one or two.

v. The most frequently missing teeth were the upper second premolar, lower second premolar and the upper lateral incisor on the non-cleft side.

D. The fissural teeth showed the following characteristics:

i. The medial fissural tooth was generally shaped like an incisor. It is often malformed or hypoplastic.

ii. The distal fissural tooth showed a mainly conical shape in the secondary dentition, whereas in the primary dentition it can be either shape. Hypoplasia and malformation was not as common as in the medial segment.

iii. Fissural teeth in the primary dentition were often larger than the lateral incisor on the non-cleft side, whereas the opposite occurred in the secondary dentition.
Fishman (1970) studied a group of 68 cleft subjects. He found that unilateral complete clefts had a higher incidence of missing teeth than any other cleft group. In a significant majority of cases the lateral incisor and any supernumerary teeth were located in the lateral alveolar segment. The lateral incisor was also the most commonly missing tooth. Delayed eruption was found to be a common occurrence, even in the posterior cleft subjects.

Olin (1964) found almost four times the frequency of missing bicuspid teeth in a group of clefts than previous estimates for a normal population.

Dixon (1968) studied the nature of enamel hypoplasia in a group of 100 subjects with clefts of the lip and/or palate. He concluded that the association between pattern of enamel hypoplasia and nature of cleft indicated the surgical repair as a significant factor.

Foster and Lavelle (1971) reported the dimensions of the permanent teeth in cleft subjects to be significantly smaller than for a normal population.

Zilberman (1973) however, reported that in a group of adults with clefts of the lip and alveolus, the dental anomalies were highly localised in the area of the cleft defect.

Ranta (1986), in a review of the literature, reported the conflicting views on the association between lateral incisor anomalies and possible microforms of the cleft lip and palate.

The literature in general appeared to concur with Bohn's findings on dental anomalies in clefts.

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1 However, there were 33 unilateral clefts, compared to 14 bilateral, 13 premaxillary and just 7 posterior clefts.
1.1.5 Analysis of the defect

Pruzansky (1953) listed four factors which must be considered for the accurate analysis and consequently, adequate restoration, of the cleft defect:

i. Adequacy of parts: Does the cleft represent a non-union of parts that are intrinsically adequate or are the parts per se inadequate or even in excess?

ii. Distortion of parts: Are the segments in any way distorted?

iii. The spatial relationship of the anatomic segments: The aberrant vectors of muscle pull from affected muscles as well as adjacent musculature such as tongue, buccinator and pharynx, may contribute to the spatial derangement of the bony segments.

iv. What is the relationship of the palate to contiguous anatomic structures?

v. The fourth dimension of Time: What are the modifications induced by the processes of growth and development to the final nature of the defect?

1.2 The pathogenesis of the skeletal deformity

The classic theory of facial development, which persists to the present, is based primarily on the observations in rabbit, pig, chick and deer, of His (1874) and Dursey (1869) (from Stark 1954, 1958). This theory holds that the face develops from five major "processes" or peninsular masses of ectoderm and mesoderm which develop around the rim of the invaginated oral cavity. These are the paired maxillary processes laterally and the single frontonasal process medially, which is in turn subdivided into pincer-shaped nasolateral and nasomedian processes. The processes grow, meet and fuse in a manner similar to the healing of a wound. Any inhibition of this succession of events causes a persistence of the pre-existing cleft.
The nose was thought to form by the fusion of the paired nasolateral processes with the nasomedian process at six weeks in utero, forming an umbilicated nasal pit where clefts existed previously, thus shutting off the two nasal pits from the mouth. The lips are formed by the two maxillary processes growing medially and uniting with the nasomedian process, which is now thickened and is called the globular process. The philtrum remains as the permanent furrow between the convexities which were once the nasomedian processes.

The area between the deepening nasal and oral cavities is the future premaxilla. The maxillary processes, triangular in shape in the coronal plane, are blocked medially by the tongue, which fills the oral cavity, its dorsal surface resting against the base of the skull. In time however, the tongue drops caudally and maxillary processes grow upward obliquely until they arch over the tongue and fuse in the midline. Superiorly, they fuse with the nasal septum at nine weeks, in the region that is to be the hard palate. The fusion occurs anteriorly first, just behind the premaxilla. At the junction with the premaxilla, no fusion occurs in the midline. An incisive foramen remains as the canal of Stenson. The maxillary processes fuse posteriorly last, to form the soft palate and the uvula. The nose is now separated from the oral pit by the secondary palate as far posteriorly as the future pharynx.

The work of Pohiman (1910) and Yeau (1937) and his own study of six cleft embryos, led Stark (1954, 1958) to conclude that the classic theory of facial processes did not apply to the central part of the face i.e. the nasal, labial and premaxillary region. The facial furrows seen in embryo were thought to be, not “processes”, but grooves between mesodermal masses which lie under the surface ectoderm. The definitive facial features are formed by differential growth of mesenchyme, not by fusion of processes. The hard and soft palate, mandible and external nares however are still believed to be formed by fusion of processes, as described in the classic theory of His.

According to the “mesodermal penetration” theory, during normal embryonal growth, the ectoderm between the nasal pits and the oral cavity (i.e. the “pre-palate” anlage of upper lip, lower
nose and pre-maxilla) thickens, forming an "epithelial wall". Mesodermal infiltration of this wall follows and is completed, usually by the end of the seventh week. The infiltration into the epithelium proceeds from both sides and from a medially placed mass of mesoderm.

Yeau (1937) and Hoepke and Maurer (1938) (cited in Stark 1954) had different interpretations of the mechanism of cleft formation in the pre-palate anlage. Yeau had envisaged that a cleft formed when the mesodermal projections failed to penetrate the "epithelial wall" completely. The "unsupported" ectoderm degenerates and pulls apart, either completely or incompletely, giving rise to the cleft (Fig. 6). A "web-like" band of epithelium (Simonart's band), which has been described in the floor of the cleft nares and may bridge the cleft, was felt by Yeau to confirm his theory of incomplete mesodermal penetration. Hoepke and Maurer, on the other hand, felt this same phenomenon confirmed their belief that the cleft occurred due to a failure of fusion, with the body making an attempt to "heal the cleft" by mesodermal penetration. They felt that the mitotic activity in the epithelial band and the orientation of the mesoderm towards it indicated the role of the Simonart's band as the scaffolding along which the mesodermal penetration must take place.

Stark's own study of six cleft embryos, to date the largest such sample, led him to the following findings:

i. In bilateral clefts, the total volumes of mesoderm on either side of the clefts were in the ratio 1:1.

ii. In the three embryos with unilateral cleft lip and palate, the mesoderm on either side of the cleft was also in a ratio of 1:1 at the level of the hard and soft palate. However at the level of the pre-palate region, there appeared to be a deficiency of mesoderm on one or the other side of the cleft.

iii. There was evidence of bands of epithelium, which were orientated across the cleft and appeared to attach to the premaxilla medially and be continuous with the ectoderm laterally. These
were too far anteriorly placed to be remnants of the oro-pharyngeal membrane. The increased mitoses and streaming of mesenchyme described by Maurer and Hoepke were not observed.

Iv. In two of the embryos, fusion had failed to occur in spite of intimate approximation of the epithelial surfaces. There was complete lack of mesoderm on the side of the cleft in the area of the premaxilla and lip in all but one of the embryos.

While he acknowledged that these findings did not prove a cause and effect relationship, Stark considered them to lend credence to Yeau's hypothesis.

**Fig. 6** The pathogenesis of harelip is presented diagrammatically. An epithelial wall exists initially as the anlage of the upper lip, pre-maxilla and upper incisor teeth (primary palate). Three mesodermal volumes are located in it, one in the midline and two laterally. These grow and fuse, forming the normal upper lip, premaxilla and four upper incisor teeth. If one volume of mesoderm is absent, the epithelial wall will rupture and a cleft will occur in that area. The absence of a lateral volume will result in a unilateral harelip. Absence of the two lateral volumes will cause a bilateral harelip to occur. (from Stark 1954).
Stark (1958) described the secondary palate (hard and soft) as developing bilaterally from the palatal processes of the maxillary bones. These processes become prominent during the sixth and seventh week in utero, after the primary palate has formed. The processes extend vertically or obliquely downward on either side of the tongue. Between the eighth and ninth week, the palatal processes begin to arch over the tongue and lie in the horizontal plane. This movement occurs in a wave-like fashion from a posterior to an anterior direction. Fusion of the palatal processes occurs in an anterior to posterior direction, with each other and the nasal septum. The division between the primary and secondary palate is signified by the incisive foramen. Numerous theories have been suggested to explain the mechanism of palatal shelf elevation, the delay or failure of which, along with the failure of fusion are considered significant factors in the development of the cleft palate. However, this area of study is beyond the scope of the present treatise.

Latham (1969) described a "clearly defined ligament" coursing from the anterior border of the nasal septum to the anterior nasal spine and median suture of the premaxillary region. He called this the "anterior septopremaxillary ligament". Latham claimed this ligament is important in the context of cleft lip and palate for two reasons:

1. It represents the means whereby the force of the forward growing nasal septum may be transmitted to the developing maxillae, pulling them forward to ensure proper spatial adjustment during the rapid growth of the embryonic face.

2. For some distance posterior to the attachment of the ligament there is no apparent connection between the nasal septum and the premaxillae, a relationship which, in the event of a cleft, may be regarded as unstable.

Latham studied six embryos with unilateral cleft lip and palate and reviewed five other embryos described in the literature, representing the period from 6 weeks in utero to 2 months
postnatal. He concluded that the youngest known cleft embryos\(^1\) tended to show that at the time of initial cleft formation (at 33–35 days in utero) the primordial face is symmetrical. The deformities, evidently caused by a condition of unbalanced development, arise in the period immediately following, when the skeletal structures of the face begin to appear. The skeletal deformity is relatively well differentiated by 12 weeks.

Latham concluded that the primacy of nasal septal growth in the embryonic period appeared to be the major cause of the skeletal deformity rather than disruption of the facial musculature as proposed by Pruzansky (1953). He contended that musculature would not be significantly developed until later in foetal life. It is the attachment of the anterior septopremaxillary ligament which dictates the nature of the septal deviation. The deformity was thought to develop in two distinct phases. The initial phase is characterised by the development of the septal deformity. The final phase is characterised by the reversal of the initial premaxillary rotation, with tilting of the premaxillary segment into the cleft due to interference of the deviated septum with the normal downward and forward growth of the maxilla (Fig. 7).

Latham (1973) studied seven embryos with bilateral cleft lip and palate covering a development period from 10 weeks in utero to 6 weeks post-natal life. The cleft proportions appeared to have been established by 10 weeks in utero. A large part of the premaxillary protrusion appeared to be due to the anterior position of the alveolar process. Latham attributed this pattern of growth to the break in continuity antero–posteriorly, of the muco–gingival pad. The premaxillary segment was thought not to be bodily rotated labially, contrary to its clinical appearance, since the teeth maintain their normal upright posture. (Fig. 8). Latham also extrapolated from his observation of the initial septal deviation in unilateral clefts, that there would be some initial under–development of the maxillae in bilateral cleft conditions, due to the absence of the normal growth–stimulation by the nasal septum.

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\(^1\) The 41–day embryos of Hochstetter (1936) and Maurer and Hoepke (1938).
Fig. 7  Diagrammatic representation of the development of the septal and premaxillary deformity in unilateral cleft lip and palate. (from Latham 1969).

A. Phase 1: Top, normal relationship between nasal septum and upper jaw showing symmetrical septopremaxillary ligament. Bottom, unilateral restraint of non-cleft side pulls growing septum off course.

B. Phase 2: The vertical deformity is compounded by interference with downward displacement of upper jaw as maxillary growth on orbital and posterior free surfaces provides the motive force. The non-cleft maxilla rotates medially, tilting premaxillary region upwards.
Fig.8 Sagittal section through the premaxillary segment of a 6 weeks old specimen with bilateral cleft lip and palate. Basal premaxillary bone has attachment to nasal septum by septopremaxillary ligament on to anterior nasal spine (ANS) and in fibrous septopremaxillary joint generally. Growth is rapid in vomer premaxillary suture (VPS); and deciduous incisor tooth lies completely anterior to nasal septal cartilage. Note protrusive labial alveolar bone (AB) and permanent incisor organ (P). (from Latham 1973).

Atherton (1967), from a study of fifteen unilateral cleft specimens also reported early establishment of the form of the cleft deformity. When compared with a group of normal controls in a cross-sectional growth study the palatal length and width measurements were very similar. This indicated that the pattern of the deformity, once established, is quite stable and differs from the controls in form only, rather than any failure of development.

Atherton also reported a relative reduction in size of the maxilla on the cleft side (approximately 8 percent), palatal shelves on both sides and the premaxilla on both sides (severe on the cleft side), when the skeletal deformity is first established. He also made the interesting observation, that the effect of the cleft on the two maxillae is remarkably similar when the premaxillary segment is excluded.
1.3 Classification of the cleft lip and palate deformity

The description and classification of the cleft lip and palate condition had begun as the individual enterprise of surgeons who maintained records for comparison and review of the defects and their treatment. These early classifications were based on morphology and surgical requirements and tended to vary quite significantly from each other. While many classifications have been proposed, only a few found wide acceptance.

Davis and Ritchie (1922) (cited in Millard 1976) had classified congenital clefts into three groups (with numbered sub-groups) according to the position of the cleft in relation to the alveolar process:

Group I - Prealveolar clefts, unilateral (1), median (2) or bilateral (3).

Group II - Postalveolar clefts involving the soft palate only (1), the soft and hard palates or a submucous cleft (2). These two sub-groups were further divided by extent - 1/3, 2/3, 3/3.

Group III - Alveolar clefts, unilateral (1), median (2) or bilateral (3).

Yeat (1931) (cited in Millard 1976) had suggested a classification divided into four types:

Group I - Clefts of the soft palate.

Group II - Clefts of the soft and hard palate.

Group III - Unilateral complete clefts of the alveolus, hard and soft palate.

Group IV - Bilateral complete clefts of the alveolus, hard and soft palate.

Table 1. shows a comparison of some of the classifications introduced in the 1920's and 1930's, which persisted well into the 1950's.
Table 1. A comparison of previous classifications of cleft lip and cleft palate. (from Kernahan and Stark 1958).

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1.3.1 Kernahan and Stark classification

The multidisciplinary nature of the modern cleft palate team created the need for a simple standardised method of classification. Kernahan and Stark (1958) were the first to present a classification based on embryology. The incisive foramen is the dividing point between the different groups of deformities. Three distinct groups were proposed:

Firstly, clefts lying anterior to the incisive foramen i.e. clefts occurring in the "primary palate" as a result of failure of proper mesodermal penetration. This group would include from minor cleft lip to whole premaxilla.

Secondly, clefts lying posterior to the incisive foramen i.e. those due to a failure of the fusion of the two palatal processes to form the secondary palate.

Thirdly, clefts which combine these two embryological events, i.e. failure of normal development of both the "primary" and the secondary palate.

The complete classification is shown diagrammatically in Appendix 1.

1.3.2 American Cleft Palate Association classification

Harkins, Berlin, Harding, Longacre and Snodgrasse (1962) presented the classification adopted by the American cleft palate association (ACPA), which was based on the same embryological principles as described by Kernahan and Stark (1958). The maxilla was divided into "Pre-palate" and Palate with the incisive foramen as the point of separation:

Pre-palate
  - Lip; Alveolar process (to incisive foramen)
Pate
- Soft palate; Hard palate (to incisive foramen)

Under each of the sub-headings, provisions were made for location (left, right, median), extent (1/3, 2/3, 3/3) and width of cleft, with specific modification such as palatal attachment of the vomer, rotation and protrusion of the pre-palate, sub-mucous clefts and congenital scar of the lip. All hard palate clefts are viewed as midline, with provision for specifying the side of vomer attachment. A grouping for “facial clefts other than palatal or pre-palatal” was also provided for completeness. Appendix 2. shows this classification diagrammatically.

1.3.3 Spina's modification of the ACPA classification

Spina (1974) presented a simplified version of the above classification for adoption by the International Society of Plastic and Reconstructive Surgery:

Group 1. Pre-incisive foramen clefts (clefts lying anterior to the incisive foramen).
Clefts of the lip with or without an alveolar cleft.
   a. Unilateral
      Right/left - total when they reach the alveolar arcade.
                   - partial

   b. Bilateral
      i. Total
      ii. Partial - on one or both sides.

   c. Median
      i. Total
      ii. Partial
Group II. Trans-incisive foramen clefts (Clefts of the lip, alveolus and palate).
   a. Unilateral
      - right
      - left
   b. Bilateral

Group III. Post-incisive foramen clefts (clefts lying posterior to the incisive foramen).
   i. Total
   ii. Partial

Group IV. Rare facial clefts.

1.3.4 International Confederation of Plastic and Reconstructive Surgery classification

The classification adopted by the International Confederation for Plastic and Reconstructive Surgery in 1967 (cited by Millard 1976) is as follows:

Group 1. Clefts of the anterior (primary) palate
   a. Lip: right and/or left
   b. Alveolus: right and/or left

Group 2. Clefts of anterior and posterior (primary and secondary) palate
   a. Lip: right and/or left
   b. Alveolus: right and/or left
   c. Hard palate: right and/or left
Group 3. Clefts of posterior (secondary) palate

a. Hard palate: right and/or left

b. Soft palate: medial

The above divisions can be further sub-divided into "total" and "partial" categories.

Rare facial clefts:

a. Median clefts of upper lip with or without hypoplasia or aplasia of the premaxilla.

b. Oblique clefts (oro-orbital).

c. Transverse clefts (oro-auricular).

d. Clefts of lower lip, nose and other very rare clefts.

Although numerous classifications still exist with reference to cleft lip and palate treatment, the above described are the most widely adopted in the literature.
CHAPTER TWO

AN OVERVIEW OF CLEFT LIP AND PALATE TREATMENT.

The profound influence of the nature of initial surgery and to a lesser extent, orthodontic treatment, on the subsequent development of the facial structures, is well documented in the literature. Any corrective procedures affecting the growth of the facial structures, must necessarily influence the nature and timing of subsequent procedures. Hence an appreciation of the overall management of the cleft condition is essential for objective assessment of the role of secondary bone grafting in cleft repair. Such an appreciation should include an overview of the evolution of cleft treatment, as well as current principles and protocols of management.

2.1 A historical perspective of cleft treatment

The earliest description of cleft therapy appears to have been an account of a lip repair in Ancient China, in the year 390 A.D. [Boo Chai (1966)]\(^1\). Millard (1976) mentions the Saxon surgeons of pre-Norman Britain as describing a method for cleft-lip repair around 950 A.D. Cautery had been a feature of Arabian surgery, whereas the Greeks and Romans had preferred the scalpel.

The Flemish surgeon Yperman (1295-1351)\(^1\) appears to have been the first to fully document a description of the cleft lip and its surgical repair. He had closed the freshened borders of the cleft lip with waxed suture. The closure was reinforced with a long needle passed through the lip on either side of the cleft and held in place with a wrap-around figure-of-eight thread. Franco (1556)\(^1\) had noted the nasal escape and related speech problems with cleft palate. Paré (1564)\(^1\) had advocated occlusion of palatal defects with a gold or silver plate, which he termed an "obturateur". Desault and

\(^1\) Cited in Millard (1976) and Converse (1977).
Bichat (1798)\(^{1}\) are credited with using a head-bandage to realign the protruding premaxilla to a more favourable position for lip closure.

Mirault (1844) had introduced the cross-flap technique on which modern lip-closure is based. Since that time nearly every conceivable type of flap—triangular, rectangular or curvilinear—has been tried. Hagedorn’s (1884) rectangular flap modification to prevent linear scar contracture, eventually led to the Le Mesurier (1949) technique. Various types of Z-plasty techniques were tried in this period in an attempt to further reduce scar contracture. This had led to the low triangular flap technique of Tennison (1952) and the rotation-advancement flap of Millard (1958).\(^{2,3}\)

Converse (1977) traces the origins of modern techniques of cleft palate surgery to the work of Von Graefe (1816) and Roux (1819), who had closed clefts of the soft palate with interrupted twine sutures. Roux had documented an immediate, dramatic improvement in the patient’s voice.

Direct closure of the hard palate had been recorded first by Dieffenbach (1826)\(^{2}\), who had suggested separating the soft tissue from the underlying bone. Fergusson (1844) and von Langenbeck (1862) had greatly advanced this method of undermining and soft tissue midline closure of the palatal defect. Flaps from tongue, neck, upper arm, forehead and even implantation of the little finger in the defect, were all tried during the latter half of the nineteenth century.\(^{3}\) In a notable set-back, Brophy (1923)\(^{3}\) had for many years influenced other American surgeons to compress the palatal segments and mechanically reduce the cleft until suture could be achieved without tension. This unfortunately led to gross deformities, with breakdown of the palatal tissues as growth proceeded.

Passavant (1861)\(^{2}\) is credited with being among the first to consider surgical methods directly designed to assist speech. Speech therapy began to be incorporated in the treatment regime around the early part of the twentieth century.

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1 Cited in Converse (1977).
2 Cited in Converse (1977) and Watson (1980).
3 Millard (1976)
Gillies and Kelsey Fry (1921) had attributed the narrowing and collapse of the maxilla noted postoperatively, to the various methods of hard palate closure then practised. They suggested closure of the soft palate only, with the palatal defect closed with a dental obturator. Gillies argued that in unoperated clefts of the hard palate, the dental occlusion was usually normal and that most of these patients needed to wear a denture whether the hard palate had been repaired or not. He further suggested that the lip operation be performed as soon after birth as possible and the soft palate repaired before the development of speech.

Wardill and Whillis, in the period 1930–1936, had demonstrated that fibres from Passavant’s ridge in the posterior pharyngeal wall extended to the tissues of the soft palate, forming a sphincter. They also described the normal function of the soft palate during speech.

With the increasing appreciation of the anatomical and physiological requirements for normal speech, the 1930’s and 1940’s saw a number of related surgical procedures. Veau had introduced a push-back technique to increase the length of the soft palate as well as suturing the palate in its three separate tissue layers to reduce scar contracture. Wardill devised the pharyngoplasty, which he coupled with the palate lengthening procedure. Both Veau and Wardill advocated lip repair as early as possible since they found this helped to mould the dental arch of the maxilla to a more normal shape prior to the palate repair. This period signified the transition from empirical selection of treatment modalities to organised research on aetiology and pathogenesis of the cleft condition and objective analysis of various procedures and their results, especially long-term effects on growth and development. This period also marked the advent of the team approach to cleft management with dental specialists and speech pathologists becoming increasingly involved as the treatment protocol became more refined.

Early dental treatment in cleft lip and palate management was mainly prosthodontic, especially involving obturation of palatal defects. Fauchard (1786) had made valuable contributions in this area. Kingsley (1897) and Case (1921) were among the first to document orthodontic
treatment regimes for the cleft lip and palate deformity.\footnote{Cited by Coccaro (1977).} However, it was from the 1930’s onwards that orthopaedic/orthodontic and related growth effects became a major area of concern in cleft management. The development of cephalometric radiography was a significant factor in the advancement of growth and speech related study. McNeil (1956) introduced the concept of pre-surgical orthopaedics, to obtain normal alignment of the segments prior to surgery. It was about this time that bonegrafting of the re-aligned clefts—in an attempt to maintain their normal relation and promote normal growth—started to gain widespread interest.

2.2 Current concepts of cleft management.

There is a vast body of literature relating to this subject. There appear to be as many variations in technique, sequencing and timing of treatment, as there have been cleft management teams. A detailed review of the literature is beyond the scope of this treatise. Hence the following description is but a very brief overview of the more commonly accepted principles and techniques. The surgical and dental protocol will be considered together to keep the coordination of procedures in perspective.

2.2.1 Aims of cleft treatment

Hotz (1973) stated the goal of cleft lip and palate treatment quite succinctly, as the restoration of anatomical continuity and normal form, with the least possible damage to growth and development through surgical intervention.

Peat (1970) divided the treatment objectives into three categories:

1. Normal speech
A long mobile palate capable of completely closing off the oropharynx from the nasopharynx is of fundamental importance.

2. Normal appearance

Modern surgical techniques are needed to produce a full upper lip with a symmetrical Cupid's bow and reconstruction of the columellar and alar architecture of the nose. Musgrave and Garrett (1977) point out that the normal appearance should be restored in infancy and persist through childhood, adolescence and adulthood.

3. Normal dental occlusion

This would involve coordination of surgery, orthodontics and prosthodontics.

2.2.2 Surgical management

For ease of description, the surgical management will be considered in three sections:

i. Repair of the unilateral cleft lip

ii. Repair of the bilateral cleft lip

iii. Repair of the alveolar and palatal cleft

Management of submucous cleft palate will not be considered since lip, nose or alveolar deformities are not involved and the clinical problems relate mainly to speech.

2.2.2.1 Repair of unilateral cleft lip

Steffensen (1953) listed five criteria for satisfactory lip repair:

i. Accurate skin, muscle and mucous membrane union.

ii. Symmetrical floor of the nostrils.
iii. Symmetrical vermilion border.

iv. Slight eversion of the lip.

v. Minimal scar, to avoid distortion from its contracture.

To this list, Musgrave (1963) added two further criteria:

vi. Preservation of the Cupid's bow and recognising the significance of the vermilion-cutaneous ridge in reproducing a normal lower border of the philtrum.

vii. Production of symmetrical nostrils as well as the nostril floors.

Watson (1980) stated that one of the most significant current advances in repair of the cleft lip has been the recognition that the muscle of the cleft lip runs parallel to the free margins of the cleft and is abnormally attached to the edge of the piriform fossa. Unless this muscle is freed and rotated across the cleft during the repair, the lip, which may look excellent at rest, will be noticeably abnormal during movement and the muscle bundles will bulge unnaturally on each side.

Musgrave (1963) and Musgrave and Garrett (1977) noted two important features of most current lip-repair techniques: most tend to discard a minimum of tissue and most introduce lateral tissue medially, in an effort to save as much of the Cupid's bow as possible. Although straight-line repair techniques such as the modified Rose-Thompson repair may be used in the correction of the most minor unilateral cleft lips¹, the more extensive clefts are repaired with a variety of Z-plasty techniques. These may have planned flaps such as the Le Mesurier (quadrilateral) or Tennison (triangular) repair or adopt a "cut as you go" principle such as the very popular Millard rotation-advancement repairs. The 'zigzag' scar tends to avoid the tight contracting line extending from nostril floor to free margin of the lip, which is a major disadvantage of straight-line repair. Musgrave and Garrett felt that the rotation-advancement repairs are much more amenable to secondary revision in

¹ These consist of a groove in the upper lip with depression of the skin and a lack of continuity of the lateral and medial muscle mass; there is a minimal associated deformity of the nostril and Cupid's bow.
the event of a less-than-perfect initial result, then are the rectangular- and triangular-flap techniques.

In the case of very extensive clefts, some surgeons may consider a preliminary lip-adhesion operation, such as the Randall-Graham method (Millard 1976, Watson 1980), where a rectangular flap is raised from the medial and lateral segments and interdigitated to allow muscle-union. This allows a period of moulding of the underlying maxillary segments and "stretching - out " of the attenuated and displaced soft tissues to provide a better base for the definitive lip repair. Musgrave and Garrett point out that one of the few disadvantages of this procedure is that the resulting scar may be a hindrance in the highly tissue-conservative methods such as the Millard repair.

Two sources of controversy in cleft management protocol relate to the extent and the timing of the primary repair procedure.

2.2.2.1.1 Timing of primary repair

Two factors appear to be significant in the timing of the primary repair:

Firstly, the ability of the infant to withstand the trauma of anaesthesia and surgery.

Secondly, the adequacy of soft tissue to satisfactorily repair the defect.

Wilhelmson and Musgrave (1966) proposed the "rule of 10", i.e. weight 10 pounds, haemoglobin 10 gms. and white cell count 10,000 per millilitre, as the minimum requirement prior to contemplation of surgery. Millard (1976) stated that most modern surgeons would follow this general rule and undertake the primary repair between 10 - 12 weeks of life. In some centres where pre-maxillary orthopaedic alignment is favoured in the case of complete clefts (Hotz and Gnoinski 1983),

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1 According to Wilhelmson and Musgrave, these parameters were met at about 10 - 12 weeks of life for most babies. They claimed that in a series of 585 cases, strict adherence to the "rule of 10" resulted in a five-times reduction in complications—although improvements in operative care and the introduction of endotracheal anaesthesia within the study period may well have significantly affected the results.
1976), primary lip repair may be delayed until 5–6 months of life.

The advocates of early surgery i.e., within the first week of life where the child's general condition allows, argue that early surgery has three major advantages:

i. Reduces the psychological trauma to the parents.

ii. Reduces feeding difficulties and allows the baby to thrive.

iii. Avoids the increase in deformity which would occur with initial rapid growth and is hence less difficult than a delayed procedure.

However, the opponents of early surgery feel that the feeding and psychological advantages are overemphasised and in fact parents who have lived for a few weeks with a cleft child are much better prepared for the vicissitudes of cleft rehabilitation. Furthermore, the general condition of the baby can be much better assessed after the first few weeks. The increase in size of the lip structures during this period would also enhance the ease and by implication, the quality of the repair (Musgrave and Garrett 1977, Watson 1980, Millard 1976).

2.2.2.2. Repair of Bilateral cleft lip

A bilaterally cleft lip tends to be usually associated with a complete cleft of the palate, though sometimes the cleft may be restricted to the premaxillary segment.

Several factors are considered critical in the correction of the bilateral cleft (Cronin 1977):

i. The size and position of the premaxillary segment and prolabium. The lack of muscle fibres in the prolabium is an added complication.

ii. The length of the columnella - which is frequently very short.

iii. The position of the lateral maxillary segments - which can often collapse medially behind the premaxilla.
The absence of muscle in the prolabium, lack of any vestige of the Cupid's bow or philtrum, the greatly reduced or absent columella and the generally greater tissue deficiency makes the surgical repair of the lip much more difficult than in the unilateral case (Watson 1980).

The timing of primary lip repair is closely coordinated with pre-surgical orthopaedic alignment of the premaxillary and lateral segments and the criteria as well as the controversy are similar to those for the unilateral cleft (Millard 1976). In the less protrusive premaxillary segment, the surgeon may elect to use the lip adhesion technique instead of an orthopaedic appliance, however this has the disadvantage of not being able to expand the lateral segments. Although some surgeons still describe the surgical repositioning of the premaxillary segment (Cronin 1977), the subsequent severe growth disruption in many cases has led to its general demise (Watson 1980).

There is a wide range of techniques of primary lip repair, most of which have two stages. Some, such as the Millard and the Cronin repair consist of a first stage where both sides of the lip defect are corrected at the one operation, followed by a second stage at which the columella is lengthened. Cronin performs the second stage within the first year but Millard tends to vary the timing of his second stage from one year for incomplete clefts to just before school-age for the more extensive defects (Cronin 1977, Millard 1976). Other techniques such as the Bilateral Tennison repair, the repair of Bauer, Trusler and Tondra and the repair of Skoog, although varying in design of their flaps, basically consist of unilateral repair with columella lengthening at one stage followed by repair of the opposite side 2-3 months later (Cronin 1977). The Millard rotation-advancement repair has also been used as a bilateral two-stage procedure in incomplete clefts (Millard 1976). Hotz and Gnoinski (1976) (also mentioned in Millard 1976) reported the closure of bilateral clefts by the Ciesniki repair, where Stage I consists of approximation of the premaxillary and lateral segments by symmetrical closure of the nostrils and nasal floor followed by a second period of orthopaedic alignment. Complete lip closure is obtained at Stage II via a Yvau or Manchester repair.
2.2.2.3 Repair of the palatal and alveolar cleft

Stark (1977) listed the following goals of cleft palate repair:

i. Construction of an air- and water-tight velopharyngeal valve, which is essential for normal speech. The velopharyngeal valve has two components, the velum (soft palate) and the pharynx.

ii. Preservation of hearing. Closure of the velum to restore the function of the tensor palatini and consequently the opening/closing function of the eustachian tubes.

iii. Preservation of facial growth.

iv. Functional occlusion and aesthetic dentition.

The timing and extent of palatal surgery is one of the major and continuing controversies in cleft management (Millard 1976; Stark 1977, Watson 1980).

Gillies and Kelsey Fry in the 1920's had pointed out that early hard palate closure led to severe maxillary deformity. They had initially advocated closure of lip and soft palate only, preferring to use an obturator for the cleft hard palate (Gillies and Kelsey Fry 1921). However they had later modified their stance to delaying hard palate closure until at least 4-5 years of age (Watson 1980).

Schweckendiek (1944)(cited in Millard 1976) had advocated closure of the soft palate at age 7 months, followed by lip closure 3 weeks later. The hard palate closure was delayed as late as 12-15 years of age. Closure of the bony cleft from 15mm. to 2-3mm. during this time was also reported. Schweckendiek (1978) presented the 25-year results of this protocol. He claimed that normal maxillary and cranial growth occurred with 60-70 percent of the palatal clefts narrowing and alveolar segments approximating in 95 percent of cases. Only about 5-10 percent of patients required a pharyngoplasty later, due to a short or immobile velum. However, speech difficulties were
encountered in the 2–5 year age period. The intelligence and temperament of the child were found to be important modifying factors in determining the degree of difficulty experienced. Normal speech occurred in 57 percent and residual minor speech problems occurred in 37 percent of cases.

Slaughter and Pruzensky (1954) also advocated the two stage palate repair in the case of wide palatal clefts. They argued that with the hard palate closure delayed, the repaired velar musculature would mould the pharyngeal and posterior palatal anatomy. The reduced hard palate defect and consequent reduction in surgical trauma would lessen adverse effects on the growth of the maxilla.

The advocates of early surgery contend that speech in cases of early closure (i.e. prior to development of speech) is significantly better than speech in delayed-closure cases. They also argue that early studies showing severe maxillary growth deformity were also related to drastic and highly traumatic surgical techniques and do not apply to modern surgical technique. Some, like Manchester (1970) maintain that the importance of attaining good speech far outweighs the importance of dental structures and to a lesser extent, even the patients' appearance.

The opponents of early surgery, however, argue that any speech problems due to delayed palate closure are easier to correct than the maxillary deformity due to early surgery (Hotz et al 1978). Hotz reported that even patients with early complete palate closure tended to show severe hypernasality between 3–5 years of age. 50 percent of the delayed-closure group required "speech plates" but their hypernasality disappeared after palatal closure and speech therapy.

Jolleys (1954) found significantly better speech results in patients who had undergone palate repair before the age of 2 years compared to those in whom the operation had been delayed beyond 3 years of age. However, these were all cases of one-stage complete palate closure with either Wardill push-back or von Langenbeck repair. Jolleys acknowledged the findings of Graber (1949), showing greater maxillary deformity with earlier surgery. While pointing out that these findings would also have been due to the traumatic nature of the early surgical techniques, he nevertheless speculated that a logical alternative would be a two-stage closure. The soft palate would be repaired soon after birth to
allow natural development of the musculature and improve the bony morphology at the posterior edge of the hard palate. A prosthesis could be used at about 3 years of age to close the hard palate defect. Hard palate repair could be performed between 5–10 years of age. The timing of the hard palate repair was based on the suggestion by Graber (1949) that most lateral maxillary growth would have occurred by 5 years of age.

Dorf and Curtin (1982) stated that errors in speech due to congenital structural deficiency, such as in unrepaired clefts of the palate, are predominantly errors in the place of production and thus distinguishable from hypernasality and nasal air emission. They cited the findings of Trost (1981) that these "compensatory articulations" can exist in children who have undergone satisfactory correction of the structures related to speech. These were thought to persist into adolescence and adulthood in spite of extensive speech therapy.

Dorf and Curtin also cited the work of Smith and Oller (1981), showing consonant–vowel sequences to emerge as early as 6–9 months of age in non–cleft infants. In their own study of 80 cleft palate children whose palates had been repaired by either von Langenbeck or Wardill push-back method; children who had palate repair prior to 12 months of age showed significantly less frequency of compensatory articulation when compared with those who had palate repair at a later stage. Interestingly, no comparison was made between the two methods of closure. Dorf and Curtin concluded that articulation age rather than chronological age should be used for timing of palatal surgery.

Evans and Renfrew (1974) also found that a group of children who had undergone palatal surgery before 8 months of age, did show better speech. However, they did not make any firm conclusions in view of the small sample size.

Herfert (1958) showed in a study on beagle dogs with normal palatal structure, that raising and repositioning of mucoperiosteal flaps led to significant transverse constriction of the maxilla.

1 Speech assessment had commenced at age 6 months, or earlier if palatal closure was to be carried out before this time.
Herfert attributed this to the ligation of the palatine artery during the flap procedure. However, Kremenak, Huffman and Olin (1967, 1970a, 1970b) repeated Herfert’s study with better controls and statistical analysis and concluded that the critical factor in maxillary growth retardation appeared to be the extent of denudation of the palatal shelves during the repositioning of palatal flaps. Temporary elevation of mucoperiosteum per se and ligation of the neurovascular bundle did not appear to significantly affect maxillary growth.

A report by Loatham and Burston (1966) indicated that lateral expansion at the midpalatal suture is greatly reduced by age 2 years. This finding had also led to the belief that palatal surgery after 18 months of age would significantly reduce the severity of transverse maxillary growth retardation (Melsels 1966).

Robertson and Jolley (1974) reported a study comparing early versus delayed hard palate closure in a group of 40 unilateral cleft lip and palate cases. The “early palate closure” group had undergone presurgical orthopaedics for arch alignment followed by lip, alveolus and soft palate closure at 3 months. A retainer plate was then worn and the hard palate was closed at 12–15 months. The authors noted that there had been a marked reduction in the width of the palatal cleft in the interim, so that only 1cm. mucoperiosteal flaps were raised on either side of the cleft. The “delayed closure” group had also undergone a course of presurgical orthopaedics, but only the lip and soft palate were closed at 3 months. The hard palate closure was delayed until 4.5–5 years of age. When the two groups were compared both clinically and cephalometrically at this time there were no significant differences between them. Robertson and Jolley concluded that although the long term effects on growth could not be discounted, these findings indicated that early palate closure with limited operations did not interfere with the blood supply or cause fibrosis sufficient to materially interfere with growth.
2.2.2.3.1. **Techniques of palate closure.**

The methods of palatal closure as mentioned previously are either one-stage procedures for repair of both hard and soft palate or two-stage procedures with delayed closure of the hard palate:

a. One-stage closure.

i. von Langenbeck repair-

A releasing incision is made in the palatal mucoperiosteum extending from the canine region to the hamulus bilaterally and placed just medial to the alveolar process. The mucoperiosteum is elevated, the soft palate relaxed, the cleft edges are pared and the two segments of palatal soft tissue are slid medially towards each other and sutured in the midline (Fig.9). Watson (1980) lists two major criticisms of this repair. Firstly, in a complete cleft, an oronasal fistula is left in the primary palate. Secondly, the procedure does nothing to lengthen the palate, which is almost always short, with the resulting problem of nasal escape during speech.

![Fig.9 The Langenbeck operation for closure of the secondary palate.](image)

(a) The margins of the cleft have been pared and the release incisions have been made medial to the alveolar processes, curving behind the maxillary tuberosities. (b) Mucoperiosteum stripped from the oral surface of the hard palate. The arrow on the right of the diagram indicates how the mucoperiosteum is raised as a bipedicled flap. The nasal mucoza on the upper surface of the hard palate and if necessary, the septum, is mobilised far enough to allow it to meet across the cleft. (c) Appearance after suturing the nasal layer. (d) Closure of the cleft completed. Raw areas are left on each side, exposing the bare bone of the hard palate anteriorly. (from Watson 1980).
11. Push-back repairs-

This type of repair was developed by Veau (1927) to increase the length of the palate. Consequently, speech results greatly improved in comparison to those he obtained with the von Langenbeck procedure. The original procedure has been modified many times, most notably by Wardill and Kilner (1937) and Braithwaite (1968) (from Watson 1980). The same release incisions are made as for the von Langenbeck repair and in addition two oblique incisions, medially and posteriorly towards the cleft from the canine ends. This allows the soft tissue to be positioned not only medially, but posteriorly as well, thus increasing the palatal length. Thus, in the isolated posterior cleft a three-flap V-Y closure occurs (Fig.10), whereas in the complete uni- and bilateral clefts, a four-flap closure occurs (Fig.11). In the modern procedure, full mobilisation of the soft palate is carried out with release of the nasal mucoperiosteum. Abnormal muscle attachments are released and most importantly, rotated medially prior to suturing, in order to reform the muscle slings. The nasal side of the cleft is generally closed with a superiorly based vomer flap of nasal mucoperiosteum. In an attempt to reduce tension in the nasal mucoperiosteum and maximise palatal lengthening, numerous techniques such as the island flap of Millard and pharyngeal flap of Höning have been devised.

b. Two-stage repair.

The technique of soft palate closure is often by simple paring of the cleft and suturing. Widmaier (1964) had described a technique with full mobilisation of the muscles and their transposition to form a functional muscle sling. The cleft of the hard palate is significantly smaller at the time of delayed closure and may often be closed with minimal medial displacement of the palatal flaps.
Fig. 10 The three-flap Wardill-Kilner push-back operation, modified after Braithwaite. (a) The margins of the soft palate cleft have been pared and the cleft of the hard palate incised along the junction of oral and nasal mucoperiosteum. The lateral incision has been made inside the alveolar ridge from opposite the canine anteriorly to a point just behind the hamulus posteriorly. An oblique incision joins the anterior end of the lateral incision to the cleft margin. (b) The mucoperiosteal flap has been elevated from the hard palate. The point of a pair of forceps is fracturing the pterygoid hamulus by pushing it medially. Deeper blunt dissection at this site strips mucoperiosteum from the side wall of the nose anteriorly and frees the attachment of the superior constrictor to the medial pterygoid plate. (c) The oral mucoperiosteal flap has been turned back showing the greater palatine vessels passing from the greater palatine foramen to the flap. The muscles of the soft palate are seen attached to the back of the hard palate. The mucosa has been elevated from the septum (the vomer) which is attached to the margin of the palate on this side. (d) The muscles have been detached from the back of the hard palate. The nasal mucosa is intact and is being dissected free from the upper surface of the hard palate. The dissector is behind the posterior nasal spine. The soft palate is falling away from the hard palate and becoming elongated. This is the "push-back". (e) The muscle has been freed from the overlying mucosa. (f) The muscle has been freed laterally and has been rotated medially. (g) The two muscle bundles have been overlapped and sutured under slight tension to construct the muscle sling. The nasal mucosa has been sutured. (h) Suturing of the oral layer completed. The tips of the oral mucoperiosteal flaps are sutured to the apex of the anterior flap, indicating the degree of palatal lengthening. (from Watson 1960).
Fig. 11  A. The four-flap modification of the Wardill-Kilner operation for closure of unilateral cleft of palate and alveolus. (from Watson 1980).

B. The four-flap modification of the Wardill-Kilner operation for closure of bilateral clefts of palate and alveolus. (from Watson 1980).
2.2.3.2. **Closure of the alveolar cleft**

Alveolar clefts, as with the nasal side of the anterior hard palate, have been in general closed with a mucoperiosteal flap turned off the vomer (Fig.12) (Millard 1976). Yeau (1938) had described a posteriorly based palatal mucoperiosteal flap which he swung across the cleft to reinforce the nasal closure behind the alveolus. However this flap did not reach the critical area of the alveolar cleft so that the alveolar closure was still a one-layer nasal mucoperiosteal repair (Fig.13) (Watson 1980).

Muir (1966) suggested that the one-layer alveolar closure is not appropriate in some situations. He pointed out that in some babies with unilateral clefts, the premaxillary portion of the medial segment is in good alignment at birth; hence the medial movement of the lateral segment which takes place after lip, alveolus and anterior palate repair results in an end-to-end abutment of the alveolar segments (Fig.14A). The arch is of good shape and later the dental occlusion is acceptable, if not ideal. However, in other babies the medial segment is rotated away from the cleft, so that medial movement of the lateral segment leads to its collapse behind the premaxilla (Fig.14B). The resulting dental occlusion would be significantly deranged. Muir attributed the arch collapse to healing by secondary intention and subsequent scar contracture due to the then-common one-layer closure. Burian (1957) (cited in Muir 1966, Millard 1976) had already reported the use of a transposed buccal mucosal flap to obtain two-layer closure to avoid the perforation of the anterior palate repair, which he found a frequent occurrence with the Yeau closure. Muir devised a similar two-layer procedure using a flap of lip mucosa which would otherwise have been discarded in the lip repair (Fig.15 A-F). Follow-up had shown no instances of fistula formation; medial collapse of the lateral segment was delayed with the alveolar gap still present at 1 year, allowing the premaxillary segment to come back into alignment (Fig.16). The alveolar gap had closed by the time of the full deciduous dentition with "good arch form".
Fig. 12. Single-layer closure of cleft alveolus and nasal floor. Incisions are made at the junction of oral and nasal mucosa on each side of the cleft. Nasal mucosal flaps are then elevated from the septum (cartilage anteriorly and vomer posteriorly) and lateral wall of nose and sutured together. (from Watson 1980).

Fig. 13. Veau mucoperiosteal flap for closure of oral aspect of anterior palate cleft. Shown in association with nasal mucosal closure of alveolar cleft. Incisions are made and the mucoperiosteal flap elevated and transported across the cleft. (from Watson 1980).
Fig. 14  (from Muir 1966).

A. Shows a dental arch of good shape at the time of repair. The stippled area represents raw surface left when the cleft through the alveolus is repaired by a nasal layer only.

B. At the age of 1 year, the alveolar gap has closed by movement of the lesser segment, but the shape of the arch is good.

Fig. 15  (from Muir 1966).

A. Shows a dental arch of poor shape at the time of repair.

B. Inward movement of the lesser segment after repair has resulted in deformity of the arch.
Fig. 16  (from Muir 1966).

A. Shows a cleft repaired by a nasal layer covered on the buccal aspect by a mucosal flap from the edge of the lip. The condition immediately after repair at the age of 3 months.

B. At the age of 1 year the distorted premaxilla has come back into line, but the alveolar gap has not closed and the general shape of the arch is good.

Fig. 17  (from Muir 1966).

A. A complete cleft lip and palate.

B. Shows the raw areas left in a standard type of repair. A Veau flap will not reach forward between the alveolar ends.
Muir (1966) noted that the maintenance of the alveolar gap at 1 year tended to make the closure of the hard palate more difficult with the conventional four-flap method than it is in the collapsed arch. Consequently it had been necessary to close the secondary palate as a two-stage procedure, with the hard palate closed later using a Veau flap. Furthermore, in some cases a substantial deficit of bone existed in the alveolar cleft. Muir proposed primary alveolar rib-grafts in these cases.

Dey (1974) also reported a variation of the Burian flap. A bulky soft tissue flap from the inner surface of the lip was carried through the alveolar gap at the primary operation. This flap consisted of muscle tissue and red lining which was mobilised from its attachment to the alar cartilage at its base. Not only did this reinforce the closure of the nasal floor, but it had appeared to successfully prevent narrowing of the alveolar gap post-operatively. This was attributed to both reduction of secondary healing due to the two-layer closure, as well as the bulk of the flap. Dey claimed that the upper incisors in particular maintained their correct relationship with the mandibular incisors. Chan (1988) found that the antero-posterior relation of the incisors appeared to be better in those cases originally subject to the Dey flap. However, this improvement was not statistically significant.

Although primary and early secondary alveolar bone grafting was popular in the 1950’s and 1960’s, long-term studies showing severe maxillary growth retardation led to its demise in most centres (Koberg 1973). However, at least two centres (Rosenstein et al 1982, Nordin et al 1983) report good results and continued adherence to early bone-grafting regimes.

In Hotz and Gnoinski’s (1976) study sample, both unilateral and bilateral clefts where Burian flaps had been used for two-layer alveolar repair at the time of primary lip closure, resulted in canine crossbite. Hence this particular variation of the procedure had been abandoned.
Too great a width of the cleft in some cases (before the advent of pre-surgical orthopaedics) and the possibility of alveolar arch collapse with one-layer closure meant that in the past, the alveolus tended to be ignored at the time of primary lip repair. Once the pull of the repaired lip brought the alveolar cleft margins close together, the alveolar closure became very difficult. Consequently, the patient was left with an oronasal fistula between the repaired lip and the repaired secondary palate which can be a great nuisance for the patient, allowing particles of food and drink to escape into the nose and interfering with speech.

Some surgeons still elect to leave a fistula as deliberate policy to avoid interfering with bony structures during the growth period. The fistula is closed as a secondary procedure¹ once growth is complete (Millard 1976, Watson 1980).

2.2.3. **Dental management.**

Hellquist (1970) listed the goals of orthodontic/orthopaedic treatment as:

i. Establish the best possible occlusion with good masticatory function.

ii. Develop a more normal maxillary architecture for better speech function.

iii. Provide better support for the upper lip and reduce any tendency toward depression of the middle third of the face.

Coccoaro (1970) also suggested that constriction and impaction of the palatal shelves would tend to affect the normal growth expression at the bony margins of the cleft.

Clefts restricted to the primary palate do not require orthodontic treatment until the permanent dentition (Peet 1970). The dental problems encountered relate to supernumerary, rotated and malformed teeth near the line of the cleft.

¹ A supplementary bone graft is now commonly placed at this secondary procedure.
In the case of isolated clefts of the hard palate, orthodontic treatment is delayed until the eruption of the premolar teeth (Peat 1970). The typical malocclusion is a constriction across the second premolar region, opposite the band of scar tissue resulting from the flap surgery used to "push back" the palate. Treatment consists of correction of the buccal crossbite.

The orthodontic management of complete lip and palate clefts can be divided into four stages (Peat 1970):

i. Pre-surgical orthopaedics.


iii. Permanent incisor correction.

iv. Full permanent dentition correction.

Coccaro (1970) made the important observation that in children with cleft lip and palate, the growth and development of skeletal and dental structures of the maxilla and the extent of maxillary involvement in the orofacial defect are problems of an ongoing nature. This makes it difficult to achieve a permanent result by instituting a definitive treatment at a set stage. Rather, the final result is a cumulative effect of the various procedures performed at critical periods of development.

2.2.3.1. Pre-surgical orthopaedics

Pre-surgical orthopaedic treatment was introduced by McNeill (1956) as part of a concept of non-surgical closure of cleft palate by "mechanical stimulation of bone". It was thought that achieving a normal alignment of the maxillary segments would firstly, influence development towards normality and secondly, provide a sound foundation for the lip repair. Treatment was started 6 weeks prior to primary lip repair. Hotz (1973) listed the objectives as:

i. Normalise function (feeding, swallowing, tongue posture).
ii. Permit and guide physiological growth under normalised conditions.

iii. Create or maintain regular intermaxillary relationship before and after primary operations.

Peat (1970) and Hotz and Gnoinski (1976) advocated up to 5 months of orthopaedic treatment prior to the primary lip repair.

Huddart (1971) found no significant difference in arch-form and anterior crossbite at age 5 years between a group of unilateral cleft lip and palate cases which had undergone 6 months of orthopaedic treatment prior to primary lip repair and a group which had not. He concluded that pre-surgical orthopaedic treatment in unilateral cleft lip and palate does not result in a better arch form or occlusion in the long term, but it is still of value in other areas, such as facilitation of lip repair and functional corrections.

Peat (1982) reported on two groups of bilateral cleft lip and palate which were "identical" with regard to surgeon, surgical technique and timing of surgery, but only one of the groups had undergone pre-surgical orthopaedics. The cases which had undergone pre-surgical orthopaedic treatment showed significantly lower incidence of incisor crossbite at the deciduous dentition (4 percent versus 46 percent) and mixed dentition (21 percent versus 73 percent) stage of development. Cephalometric analysis however, showed no significant difference between the two groups at either stage.¹

2.2.3.2. Deciduous dentition treatment

Helquist (1970) considered collapse of maxillary arch, buccal and anterior crossbite as indications for early orthodontic/orthopaedic intervention in the deciduous dentition, from both an

¹Comparisons between different studies and sometimes within a study are difficult, if not invalid, due to the likelihood of individual differences in severity of defect and surgical technique. Hence, throughout the treatise, comparative findings should be treated with caution.
anatomical and functional (tongue posture and speech habits) standpoint. Subtelny (1966) felt that force exerted by the reconstructed lip led to the medial collapse of one or both maxillary segments. He stated that both normal development of the maxillary segments and normal tongue function during speech would be aided by expansion of the segments into better alignment. He emphasised that retention of the repositioned maxillary segments is essential to maintain them in the desired position.

Pruzansky (1955) also mentioned improvement of the nasal airway and removal of any unnecessary supernumerary teeth if they blocked the alignment of normal dental units as important aspects of early treatment.

Rygh and Tindlund (1982) advocated the early protraction of the maxilla with a face-mask according to the principles of Delaire, in patients who display under-development of the midface in the mixed dentition. The maxillary protraction is preceded by a period of slow maxillary lateral expansion with a Quad-helix.

2.2.3.3. **Permanent incisor correction**

After eruption of permanent incisors, if they were in crossbite, a simple upper fixed appliance is used to correct the incisor position. This is only carried out if the overbite is adequate for retention; if not, the incisor relation is accepted until the full permanent dentition correction. Hellquist (1970) and Hotz and Gnoinski (1976) conducted this stage of treatment between 7–9 years of age. Subtelny (1966) noted that the incisor teeth adjacent to the cleft were often severely rotated and had unfavourable inclination.

2.2.3.4. **Full permanent dentition correction**

Following the eruption of all permanent teeth, full orthodontic appliances are placed to correct the occlusion, with a cobalt-chrome denture or either a conventional or Maryland-type bridge, which
acts as both prosthesis and permanent retainer. Subtelny (1966) pointed out that whereas the earlier repositioning involved movement of bony segments, this last stage involves individual tooth movement.

Secondary and late-secondary bone grafting is performed, respectively before and after the final phase of orthodontic treatment.

The literature tends to show that although the general aims of cleft management are the same throughout the world, the protocols for achieving these aims are not. In fact, there appear to be as many variations in cleft management protocol, as there are cleft treatment centres. This observation applies particularly to the timing and nature of primary lip and palate closure, as well as presurgical orthopaedics. In contrast, later orthodontic treatment and secondary grafting procedures appear to be more standardised.

These differences in primary corrective protocol and consequently the type and severity of secondary defects tend to occur, not only between different treatment centres, but for different patients within the same centre as well. This is to be expected, since the type and severity of the cleft defect and its attendant problems relating to speech, hearing and general condition of the infant, varies from patient to patient. Even the one surgical technique, when performed by different surgeons, or the same surgeon on different occasions, may give quite different results, due to variations in the nature of the cleft defect, flaps raised and extent of post-operative scarring.

This wide variation in both treatment protocol and results of primary corrective procedures must be taken into account when assessing the success of secondary grafting procedures.
CHAPTER THREE

THE TRANSPLANTATION OF BONE

Bone grafts are used clinically for 3 main purposes (Burwell 1965):

i. To cause union in fractures.

ii. To stiffen joints.

iii. To overcome defects in bone.

In grafting bone, Burwell makes two demands:

i. The method of grafting must be mechanically sound.

ii. The graft is incorporated into the host skeleton within a reasonable period of time, without complications.

Burwell stated that in their attempts to fulfil the latter demand, most authorities had arrived at the conclusion that the patients' own live bone i.e. autogenous bone, provides the best material for grafting. Virtually all grafting in the management of cleft lip and palate involves the use of autogenous bone. Hence the discussion on the histology of bone grafting for the purposes of this treatise will be limited to a consideration of autogenous grafts.

3.1 Histology of transplantation of bone.

The basic concepts of bone graft healing were formulated at the turn of the century by Barth (1895) and Axhausen (1908) (from Albrektsson 1980, Burchardt and Enneking 1978). Barth had determined that the greater part of a bone transplant underwent necrosis and that a successful repair of the transplant depended on the intimate contact of the graft with vascular tissue of the host. Axhausen had further illustrated that empty lacunae were an indication of necrosis; that cellular
proliferation occurred beneath the periosteum; that bone formation occurred from transplanted bone marrow whenever the latter was in contact with living tissue and that necrotic bone was replaced by new tissue moving along channels made by invasive blood vessels. Axhausen had called this last phenomenon "creeping substitution". Axhausen had also suggested that transplant revascularisation involves the infiltration of host osteocytes into the empty transplant lacunae.

3.1.1 Theories of graft repair.

Axhausen (1956) stated that historically, two conflicting theories of bone regeneration in grafts had existed:

i. The "Osteoblastic" Theory

Ollier (1867) had found that free bone transplants with periosteum intact could survive and grow in response to functional stress. Axhausen (1909) had confirmed this work by showing that while transplanted bony elements became necrotic, the periosteum survived and deposited bone. This "osteoblastic theory" maintained that in graft regeneration, bone is produced by distinctive cells of the periosteum, bone marrow and Haversian canals, which were destined to become osteoblasts.

ii. The "Inductive" Theory

Barth, however, had found that in transplants of calvarium without periosteum, the transplants became necrotic and were replaced by bone arising from the osteogenic tissue of the graft bed. Baschkirzew, Petrow (1912) and Wereschinski (1925) had found bone formation to occur after transplantation of bone fragments devoid of periosteum into the soft tissues. Martin (1927) had noted the absence of "resting osteoblasts" in adults and supported the concept of osteogenesis by metaplasia of the cells of the recipient beds. This concept had received further support from the work of Orell (1934) who found bone formation occurring after 100 days, around transplants which had been prepared by boiling. Engström and Orell (1943) noted similar results after 53 days with transplants.
which had been prepared by repeated freezing at −190° centigrade. This had led to the "inductive hypothesis", i.e. the recipient bed, when stimulated by an inductor substance in the transplant, is able to elaborate bone.

Axhausen (1956), in his own series of experiments found that active bone formation occurred as early as four days following transplantation of free autogenous grafts into the soft tissue of adult dogs, provided that periosteum or marrow cells were part of the transplant. When periosteum or marrow was absent, new bone formation was reported to be minimal and greatly delayed. Where autogenous periosteum alone was transplanted into adult dog muscle, the periosteum shrunk and underwent necrosis. When the periosteum was maintained in a stretched position, the contracture was avoided but osteogenesis still did not occur. However, implantation of cortical bone fragments near the stretched periosteum resulted in typical periosteal new bone formation in some cases. In a series of seven grafts frozen at −35° centigrade and maintained at −15° centigrade for 24 hours prior to implantation in the soft tissue of dogs:

Grafts removed between the tenth and fourteenth day after transplantation were completely necrotic. Vascular buds of the host bed infiltrated the bone canals and periosteum but no new bone formation was visible. Grafts removed between the sixteenth and thirtieth day showed resorption of the bone, with mononuclear and multinuclear osteoclasts, but no new bone formation. The two graft-specimens which were implanted for longer than thirty days however, showed widened bone canals which contained not only osteoclasts but osteoblasts, with newly-formed seams of bone at the periphery of the canals. While lacunar resorption by osteoclasts was actively proceeding deep in the resorption area, the more superficial parts of that area disclosed osteoblasts forming lining layers of osteoid. In eight fresh cortical transplants of rib or tibia with periosteum on the other hand, the bone deposition was characterised by patterns of young, plexiform bone tissue spread on the surface of the graft. In ten experiments with bone grafts where the organic component had been denatured by alcohol, an osteoclastic resorption could be demonstrated as early as four weeks. However there was no trace of bone regeneration in these grafts even at twelve weeks after transplantation.
Axhausen concluded from these findings that non-specific pluripotential connective tissue cells of the host develop into osteogenic tissue under the influence of the necrotic transplanted bone, but only in the presence of adequate extent and/or duration of osteoclastic resorption. The determining factor appeared to be the composition of the organic matrix. Axhausen stated that the osteoblastic and induction theories could be unified in a single concept of osteogenesis:

Bone regeneration occurs in two phases. The first and physiologically most important phase originates in the pre-existing specific cells and begins in the first few days after transplantation. The second phase occurs after several weeks and originates within the non-specific host connective tissue of the graft bed.

Goldhaber (1961), Ray and Sabet (1963) and Sabet et al (1964) demonstrated quite strongly, the osteogenic inductive capacity of embryonic and neonatal bone grafts. The latter two studies showed that in in-bred rat neonatal isografts (and by implication, autografts), both the graft (by proliferation) and the host (by induction), contributed to new bone formation.

Burwell (1965) felt that some of the early confusion regarding the inductive capacity of adult bone was due, at least in part, to the type of models used. These had involved the implantation of devitalised bone in tissues of low osteogenic potential, e.g. subcutaneous tissue, skeletal muscle, kidney and anterior chamber of the eye. In contrast, Burwell used a composite homograft-allograft bone and marrow model in the rat (Burwell 1964a, 1964b) to show that iliac bone which has been frozen or freeze-dried induced osteogenesis in marrow, whereas deproteinised bone inhibited osteogenesis. Bone which had been boiled or merthiolated before impregnation with autologous marrow lay between these two extremes. Burwell felt that although these studies did not illustrate the mechanism of osteogenic induction, they nevertheless strengthened the view that adult cancellous bone, after transplantation, will induce osteogenesis in other connective tissues; moreover, that this inductive potential can be lowered by certain physical and chemical treatments.
Extrapolating from studies on dermal cell function, Burwell (1965) speculated on the triggering mechanisms for osteogenic activity by the graft tissue, such as loss of contact between bone cells and alteration in the diffusion of substances between bone cells. He further speculated on possible factors regulating size of the repaired graft such as contact-inhibition of mitosis and diffusion of tissue-specific mitotic inhibitors.

Burchardt and Enneking (1978) noted that it had not yet been determined whether transplant or host cells play the primary role in transplant-host wound repair, although both are considered to play a role in creeping substitution.

Theories of graft repair have thus progressed from the apparent conflict of the "osteoblastic" and "inductive" hypotheses to the current understanding that the osteogenic potential of the graft, as well as the inductive response of the host are both important and intimately involved.

Continuing investigation of the relationship between graft bone and host inductive response is vital to the success of non-autogenous bone grafting procedures. Although the inductive capacity of transplanted bone is well documented, it appears that the actual mechanism of induction and the complex relationship between bone, periosteum and host-tissue osteogenicity is not fully understood.

In fresh autogenous bone grafting (which is the method of choice in cleft repair) the host response is important. However, it is the osteogenic potential of the graft and its growth compatibility with the maxillary segments, which appear to be the factors critical for success. Hence an understanding of histological differences between cortical and cancellous bone as well as the difference in graft repair between cortical and marrow/cancellous bone grafts is essential, to adequately evaluate the secondary bone grafting procedures.
3.1.2 Cell function in bone grafting

Bassett (1972) stated that cell action is required to unite a bone graft with the host, to increase or decrease the mass of the graft and to precipitate rejection. He felt that much of the confusion regarding osteogenesis following bone transplantation had resulted from a failure to differentiate between cortical and cancellous bone and between mature and embryonic grafts. Many early investigators had doubted that cells in the graft survived even after autotransplantation. However, more recent studies (Bassett and Rüedi-Lindecker, 1964; Burwell, 1964a) had demonstrated that surface cells in free, mature, cancellous bone grafts can survive transplantation, if properly handled and will participate in the total early osteogenic response around the graft.

Burwell (1965) stated that in the cancellous graft, osteoblasts proliferate, not only on the trabecular surfaces, but periosteally as well. The marrow also survives in the outermost parts of the graft and rapidly invades the deeper necrotic marrow, accompanied by the ingrowth of vascular and cellular components of the graft-bed. Burwell (1964a), using a composite homograft/allo graft bone/marrow model in the rat, demonstrated that the new bone formed by a fresh iliac autograft is derived from two sources, osteoblasts from the bone and endothelial cells from the marrow. Bassett, on the other hand, doubted whether significant numbers of osteocytes persist in a graft of mature bone, whether cortical or cancellous. Even if they did survive, he considered it unlikely that they could free themselves from their lacunar entrapment to join other cells in the “pool” surrounding the graft.

The nonvital extracellular bony matrix of the graft could have several functions:

i. It could serve a mechanical function to occupy space and provide surfaces for cell migration.

ii. It could impose a micro-anatomic form to guide cells during reorganisation.
iii. It could serve structurally in resisting applied forces and thereby generate electric potentials.

Bassett pointed out that blood vessels could invade an osseous graft only when shearing motion between the graft and the host tissue is largely eliminated. Hence it is important to establish the largest possible mass of callous in the shortest possible time in order to "glue" the tissue and graft firmly together. Although the major source of these cells is in the more vascular regions of both soft and hard tissues at the recipient site, an appreciable number of osteogenic cells can be transplanted successfully on properly prepared cancellous grafts. The vascular invasion is primarily a product of the host and is responsible for an influx of cells which will sweep toward the center of the graft. The invading host perivascular cells and vessels set the stage for the sequential removal of pre-existing bone matrix and its replacement by new bone.

Bassett listed three basic requirements for bone formation:

1. A "proper" cell.

2. "Proper" nutrition.

3. A "proper" stimulus.

If these three factors do not co-exist, osteogenesis cannot occur.

The "proper" cell is a mesenchymal cell which, in the adult, generally arises from perivascular cells associated with proliferating blood vessels, reticular elements of bone marrow, trabecular surfaces and the cambial layer of the periosteum.

In addition to the properties of the cell itself, some of the more important factors determining the availability of nutrients are:

1. The nutritional status of the host.

2. The distance of the cell from its source of supply in the adjacent vessel.

3. The diffusion rate in the extracellular space.
iv. The interposition of barriers to diffusion (e.g. other cells) between the cell and its source of nutrition.

v. The propulsion of extracellular fluid by cyclic deformation of tissues.

vi. The electro-phoretic and electro-osmotic "pumping" action arising from stress-generated electric potentials.

Experimental transplants have also shown that mechanical force factors affect the differentiation of mesenchymal cells at the graft site. Tensile forces tend to lead to formation of osteoclasts and fibroplasia while compression is associated with specialisation of osteoblasts.

Bassett also emphasised the importance of correct handling of graft at operation to maintain the viability of surface cells and promotion of effective diffusional nutrition of the graft after placement.

The viability of the graft surface cells may be reduced by a variety of factors en route from the donor site to the host site:

i. Prolonged exposure to air.

ii. Contamination with cold sterilising agents at the operation site.

iii. Elevation of temperature from direct exposure to the operating-room lights.

iv. Cytotoxic effects of certain antibiotics.

Puranen (1966), using a tetracycline-labelling technique, showed that fresh autografts have a significantly higher osteogenic activity than those exposed to air for one hour or maintained in saline for three hours. In a similar experiment, Bohr et al (1968) demonstrated no significant difference in osteogenic capacity of fresh autografts or autografts maintained in saline for one hour. However both these types of grafts showed significantly greater osteogenic activity in the first ten days after transplantation than in autografts exposed to air for one hour. Bassett suggested that in the interim between harvesting and placement of graft, the bone should be wrapped in a moistened blood-soaked
sponge and placed in a container, the top of which is also covered with saline-moistened sponges and kept in a cool spot. He felt that cell-viability could be maintained for up to 4-6 hours in this way.

The results of Marx et al (1979), on relative rates of cellular survival of human iliac crest marrow and cancellous bone grafts in various media, contradicted Bessett's recommendation. Using dye-exclusion and tritiated thymidine uptake methods of measurement, Marx et al found that the best storage medium was normal saline, which showed 100 percent cell survival at 30 minutes and 95 percent at 4 hours after harvesting of graft. 5 percent dextrose in water (D5W) and Tissue Culture Medium No. 199 were also very good storage media, with about 98 percent cell survival at 30 minutes and 94 percent at 4 hours post-harvest. Autologous serum, in contrast, showed 90 percent cell survival at 30 minutes and a low 79 percent survival at 4 hours post-harvest. These authors suggested that although the normal environment of marrow cells is a filtration product of blood, clotted blood, from which the serum was obtained, may have cytotoxic products released by the clotting mechanism.

Several critical steps to ensure effective nutrition of the graft in situ include (Bessett 1972):

i. Prevention of interposition of dead-space, haematoma or necrotic tissue between the graft and its bed.

ii. Placement of cancellous surfaces of the graft next to the cancellous bone in the bed.

iii. Limit the thickness of cancellous grafts to a maximum of 5mm., if a central zone of necrosis is to be avoided.¹

iv. Survival of fresh autologous cancellous grafts can be improved by placing them in a

¹Stringa (1957) found that autogenous cancellous grafts had vascular penetration to 3mm. by 7 days, but 5mm penetration took at least thirty five days.
bed which is already actively producing new vessels and bone (Siffert and Barash 1961).}

3.1.3. Comparison of cortical and cancellous bone grafts

Cancellous transplant material is used primarily as a filler of small defects, whereas segments of cortical bone are used primarily as supportive struts (Burchardt and Enneking 1978).

Both types of transplants have similar histological features during the first two weeks post-grafting (Abbott et al. 1947, Delieu and Trueta 1965, Enneking et al. 1975):

At first, areas of coagulated blood are evident in the graft bed as a means of preventing blood loss. During the first week, the graft is the focus of an inflammatory response characterised by vascular buds infiltrating the graft bed, with exposed surfaces of the graft bathed in nutrients. There is an abundance of inflammatory cells. By the second week, there is a reduction in the inflammatory cell response and the proportion of fibrous granulation tissue in the graft bed increases, as does osteoclastic activity. While surface cells of the graft are maintained by diffusion of nutrients from the host tissue, necrosis and osteoclasts occur within the graft.

The cancellous bone grafts differ from the cortical grafts by the rate of revascularisation, mechanism of creeping substitution and completeness of repair (Stringa 1957, Burchardt and Enneking 1978, Albrektsson 1980).

Albrektsson (1980) reported that in a vital microscopic study of transplants and re-implants in adult hares, a number grafts showed incomplete revascularisation at 50–80 days, which was partly attributed to surgical trauma at the time of graft.

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1Siffert and Barash (1961) showed that delaying transplantation until the graft-bed haematoma had been largely replaced by a vascular undifferentiated tissue (at about 7 days in the rabbit and 10 days in the dog) caused the cellular elements arising from the host bone and periosteum to make more rapid contact with the transplant and to invade it more quickly.
3.1.3.1. **Secondary histologic features of cancellous bone transplants**

Although the primary method of revascularisation is considered to be by gradual host ingrowth into the marrow spaces, several studies (Abbott et al 1947, Deleu and Trueta 1965) have suggested that this process may commence within hours after transplantation as the result of end-to-end anastomoses of host and graft blood vessels. In Deleu and Trueta’s study in rats, cancellous grafts were entirely covered with vessels in two days and revascularisation was complete within two weeks. This was more rapid than previous studies, e.g. Stringa (1957), in rabbits. The authors commented that in animal studies, the rate of revascularisation appeared to vary between the different animal models used.

As the vascularisation of the graft proceeds, primitive mesenchymal cells from both host and graft differentiate into osteogenic cells. These differentiate into osteoblasts that line the edges of necrotic trabeculae and deposit a seam of osteoid which eventually surrounds the central “core” of necrotic bone. The osteoid is gradually remodelled and replaced by lamellar bone.

The entrapped cores of necrotic bone are gradually resorbed by osteoclast activity. Haematopoietic elements gradually accumulate within the graft and eventually the necrotic bone is completely replaced by viable new bone (Urist and McLean 1952).

3.1.3.2. **Secondary histologic features of cortical bone transplants**

The most obvious histologic difference between cortical and cancellous graft repair, according to Burchardt and Enneking (1978), is their relative rate of revascularisation. Generally, the cortical graft is not penetrated by blood vessels until the sixth day, with complete revascularisation occurring

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1 Albrektsson (1980) emphasised that this may occur only under very specific conditions.
2 The origins of the osteogenic precursor cells is still controversial (Burchardt and Enneking 1978).
within one or two months (Stringa 1957, Delau and Trueta 1965). They attribute the delayed revascularisation to the difference in bone structure, since vascular penetration of cortical bone is primarily through peripheral osteoclastic activity and vascular infiltration of Volkmann's and Haversian canals (Abbott et al 1947, Delue and Trueta 1965, Enneking et al 1975).

Cortical graft repair also varies from that of the cancellous grafts in that it is initiated by osteoclastic rather than osteoblastic activity.

Enneking et al (1975) in a controlled study of cortical bone transplants in dogs, stated that qualitatively, the pattern of repair resembled that described by Axhausen (1909) and confirmed by later investigators. An autogenous graft is repaired principally by creeping substitution of living for dead bone. This is externally reinforced by a callous, most likely produced by a combination of surrounding host cells induced by the presence of the graft and surviving surface elements of the graft itself.

The initial reparative resorption proceeded from without inwards during the first six weeks. Old necrotic Haversian canals were enlarged by osteoclasts which accompanied the invading buds of vascular granulation tissue and produced the early porosity. Albrektsson (1980) reported a maximal vascular ingrowth rate of .15- .30mm./day for cortical grafts, where pre-existing canals were being revascularised, compared to at least .2-.4mm./day for cancellous grafts. Strikingly, the resorption consistently involved only the osteonal systems and did not remove the interstitial lamellae. During the first six weeks appositional activity was less in the transplants than in the controls, even though revascularisation and resorption were well advanced. By eight weeks, about one-third of the transplant had been replaced with new bone, one sixth consisted of resorption cavities and the remainder was necrotic matrix. After the twelfth week, the rate of new-bone apposition in the graft and control specimens appeared to be the same. At forty-eight weeks, about sixty percent of the transplant was composed of new bone. The remaining necrotic matrix was almost exclusively interstitial lamella. Burchardt and Enneking (1978) felt that this appearance characterised a further
significant difference between cancellous and cortical bone grafts in that, while the former tend to be completely repaired in time, the latter tend to remain as admixtures of necrotic and viable bone.

Engdahl (1972) carried out an extensive macroscopic and microscopic animal study in a large sample of rabbit sucklings. He examined the influence of periosteum on bone regeneration and the effect on growth of bone regeneration in maxillary defects filled with blood, bone marrow graft, solid bone graft or a synthetic haemostatic material (Surgical):

The periosteum appeared to be essential for host-bone regeneration in the absence of a graft. The more periosteum preserved at surgery, the more rapid and more complete was the restoration of normal anatomy. When the lateral periosteum was resected together with the bone but the medial wall of mucoperiosteum remained in place, bone formation was slower and less extensive. When both lateral and medial periosteum was resected, bone regeneration was minimal. However, the status of the host periosteum had no significant effect on maxillary growth.

In contrast, bone grafting with split-rib grafts led to rapid bony healing with a large callous, irrespective of the status of the host periosteum, although the rate of incorporation and amount of bone formed was greatest when the periosteum was intact. Microscopic study showed that new bone formation started within a few days. In many grafts a small central portion was not resorbed but incorporated into the new bone. However, with growth, all the bone grafted animals showed severe distortion of the maxilla towards the grafted side. Engdahl observed that the large bony callous appeared to "lock" the margins of the maxillary defect together and thereby hinder normal growth. Stenström and Thilander (1967) had found similar growth effects in the guinea pig model, when iliac cortical grafts were used.

Blood and bone marrow aspirate both produced rapid and good bone formation. The reparative bone was equal to or thinner than that resected. Most importantly, the bone thus produced appeared to grow in harmony with the facial skeleton irrespective of the amount regenerated.

At face-value, the results appeared to indicate that the periosteal covering of the maxillary defect might be more important for bone regeneration than the use of bone marrow aspirate. It was only where the lateral periosteum had been resected, that the bone marrow aspirate led to relatively greater bone regeneration. Engdahl however, suggested that damage to the marrow structure by aspiration may have reduced its osteogenic capacity. Good results by Richter et al (1968), using

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1 The lateral maxillary wall was resected, i. totally subperiosteally, ii. including the lateral periosteum, iii. including periosteum of both lateral and medial walls, iv. including resection of the lateral periosteum and chemical cauterisation of the medial periosteum.
larger particulate marrow grafts supported this view. Richter and Boyne (1969) also queried the possibility of reduction of osteogenic potential when marrow is aspirated.

Engdahl commented that although larger particles of graft-bone may give more rapid and better bone regeneration, this would have to be further investigated in the light of growth retardation associated with block grafts.

Surgical delayed the formation of bone and the closure of the defect was seldom complete, although there was little interference with growth of the maxilla. This was contrary to the findings of Thilander and Stenström (1970), who found that Surgical stimulated bone formation. However, Engdahl explained this conflict as being due to differences in experimental technique and the animal model used.

Further notable findings were that local infection at the graft-site and cauterising the medial mucoperiosteum were also strongly associated with growth retardation.

Richter and Boyne (1969) suggested that the grafting technique should be related to the bony anatomy of the host site. Hence, in areas with extensive endosteal spaces, inlay grafts perform better than onlay grafts. However, in areas with reduced marrow spaces, e.g. ascending ramus or neck of condyle, onlay grafts would have the better chance of success.

The review of research into bone graft repair, if nothing else, highlights the complexity and fragility of this process. A large number of factors, ranging from type and maturity of the graft bone and the nature of intra-operative handling, to the preparation of the recipient site and the extent and quality of periosteal covering, have been shown to be critical for successful transplantation.

The differences between cancellous and cortical bone, not only with regard to speed and extent of graft "take", but also in terms of growth compatibility with the surrounding host skeleton (as highlighted by Engdahl (1972)), is of particular significance in the grafting of cleft defects. Virtually all the reports of primary bone grafting and many of the early secondary grafting studies involved the use of cortical bone from the rib. The use of cortical grafts in cleft repair (and not always as "struts") was still being reported in the early 1970's, even though the superior osteogenicity of
marrow/cancellous grafts had been well documented by this time. It should be noted when evaluating the studies of secondary grafting with cortical bone, that the criteria for success were often limited to a radiographically demonstrable bony union between the maxillary segments. With a few exceptions, effects on the dental arch either in terms of reconstruction or changes with continued growth, were not reported.

3.2 Evolution of bone grafting in cleft lip and palate repair.

A detailed review of the origins of bone grafting in cleft surgery and the vast array of literature on primary and early secondary bone grafting is beyond the scope of this treatise. However, a brief overview of the modern era of bone grafting would help to maintain the advent of secondary grafting procedures in perspective.

Schmid, in 1952 (Koberg 1973), had started the systematic application of bone grafting in cleft lip and palate repair. Nordin and Johanson (1955) (cited in Koberg 1973) had transplanted grafts from the iliac crest into the cleft alveolus, after orthodontic treatment in the mixed dentition. Nordin (1957), Johanson (1958) and Schruder and Stallmach (1959) (cited in Stallmach 1963), had introduced the concept of primary bone grafting at the time of initial cleft repair. The proposed advantages were that maxillary collapse could be avoided and more normal maxillofacial development could be achieved. Nordin (1957) showed that migration of teeth into the grafted area could occur.

During the early 1960's, bone grafting in clefts of the lip, alveolus and palate became routine practice at most cleft palate centres (Koberg 1973). Although late secondary grafting was being performed in some patients (Johanson and Ghisson 1961, Stenström and Thilander 1963, Matthews and Grossman 1964) the focus of grafting research was on the primary procedure.

The sources of bone used for grafting were cortical block and/or cancellous chips from rib, iliac crest or tibia (Giorgiade et al 1964). Skoog (1965) introduced the concept of "boneless bone
grafting" or periosteoplasty. He used a two-layer periosteal closure, employing nasal mucoperiosteum for the nasal layer and a rotation flap of mucoperiosteum from the anterior maxilla for the oral layer. Bone formation across the alveolar cleft was demonstrated at 3–6 months post-operatively.

The literature pertaining to bone transplant research (3.1) also indicates that during this period, from the mid-1950's to the late-1960's, the significant differences in osteogenic potential and subsequent growth and remodelling between cortical and marrow-cancellous grafts were not fully understood. Consequently, most if not all of the grafts placed, were solid-block or particulate corticocancellous bone. Boyne and Sands (1972) appear to be the first to report the use of particulate marrow-cancellous grafts in cleft repair.

A number of studies in the late 1960's and early 1970's, such as Kling (1964) (cited in Koberg 1973), Pickrell et al (1968), Robertson and Jolleys (1968, 1983) and Jolleys and Robertson (1972), Rehrman et al (1970) and Fiede and Johanson (1974)), reported significant maxillary growth retardation in both sagittal and coronal planes following primary and early secondary bone grafting. This led to its gradual abandonment. Several groups (Nylen et al 1974, Nordin et al 1983, Rosenstein et al 1982) claimed that poor results and growth retardation were due to technique of treatment rather than timing of the grafting procedure. However, Ross (1987), in a multi-centre study, confirmed that alveolar bone grafting in infancy retarded maxillary growth. He also pointed out that hard palate repair appeared to be the major influence on maxillary growth. He suggested that the delicacy of the surgery, rather than timing or technique of repair, is the critical factor in minimising growth retardation.

Koberg (1973) in his comprehensive review of bone grafting procedures concluded that, severe maxillary deformity occurred as a late result of primary bone grafting and that secondary grafting remained the only justifiable form of bone transplantation in cleft surgery.
CHAPTER FOUR

SECONDARY ALVEOLAR BONE GRAFTING IN CLEFT LIP AND PALATE

In the 1970's, studies appeared suggesting that, if bone graft repair of alveolar clefts was delayed until the age of mixed dentition, good function would result and there would be much less effect on growth and development.

Epker and Wolford (1980) suggested that when properly timed and performed, alveolar grafting provides "relatively normal alveolar bone continuity, good stabilisation for mobilised or expanded maxillary dento-osseous segments and supporting bone for adjacent teeth to erupt into or be orthodontically moved into and can improve the periodontal health and longevity of teeth adjacent to the cleft."

4.1 Indications for secondary bone graft

Clifford et al (1972), conducted a psychological survey of 98 'completed' cleft lip and palate patients. They found that although overall body-image scores were high, when various items of the body-satisfaction scale were ranked according to the mean satisfaction levels, those associated with clefts rated low. The item with the lowest satisfaction level was teeth, closely followed by speech. Talking, nose and lips had followed in ascending order of satisfaction. It is of interest that 65 percent of the sample had received no orthodontic treatment and 76 percent had reported no previous speech therapy.

Waite and Kirsten (1980) stated that, apart from any psychosocial disturbances, the following sequelae may arise from the unrepaired residual alveolar and palatal defect:

1. Malposition of one or more teeth in the anterior maxillary region.
2. Insufficient periodontal bone support for teeth adjacent to the bony cleft with consequent chance of early loss of these teeth.

3. Less favourable hygiene conditions caused by the malposition of the teeth and the presence of an oronasal communication. Consequently caries and periodontal inflammation may occur.

4. In relation to one or other movable maxillary segments with unfavourable conditions for prosthetics, insufficient retention for full or partial dentures and root fractures of bridge-abutment teeth.

5. Adverse effects of speech because of irregular tooth position, a tapered dental arch and escape of air via the oronasal communication.

6. Facial asymmetry, not only because of a difference in form of the nasal alae and columnellar deviation, but also due to lack of bony support for the alar base.

7. Nasal crusting caused by a lack of separation of oral and nasal secretions.

Witsenberg (1985), in his review of the literature, listed some additional problems:

8. Retention of food particles in mucosal recesses and/or narrow fistulae.

9. Lack of a firm bony base for support of a denture if required.

10. Unfavourable conditions for surgical repair of the posterior palate, since primary or early secondary replacement of the bone in the hard palate would have put these patients on par with those in whom only the soft palate is cleft.

11. Insufficient bone to enable orthodontic movement of teeth into the best position and occlusion. In this situation rather extensive prosthetic treatment cannot be avoided.

12. Relapse after orthodontic expansion of collapsed maxillary segments.

13. Long artificial teeth or excess of labial acrylic in cases where fixed or removable partial dentures are worn, because of lack of sufficient alveolar process at the site of the cleft. There
is a consequent retention of food particles, mucosal inflammation and development of calculus in relation to the denture.

14. Obstruction of air passage through the nose caused by the altered anatomy and a hypertrophic inferior turbinate.

15. Poor aesthetics of the lips because of lack of natural support from the alveolus and the teeth.

4.1.1. Residual oronasal fistulae

Jackson (1972) stated that many cleft lip and palate cases referred for secondary surgery have anterior fistulae of varying proportions. In the unilateral case these extend from the buccal sulcus through the alveolus into the hard palate, occasionally involving its whole length into the velum. In some instances there can be two or three fistulae along the line of the initial cleft repair. The width of the fistula may be slight where there has been simple parting of the old repair, or very considerable where the anterior flaps of the Wardill-Kilner four-flap closure have necrosed. In bilateral cases there may be unilateral or bilateral fistulae, sometimes complicated by previous excision of the premaxilla with subsequent flap necrosis. According to Jackson, it is in these latter cases that the very widest of fistulae are seen.

Bowers and Gruber (1973) pointed out that in the past, some surgeons made little effort to prevent fistula formation in the anterior palate when performing cleft palate repair. This practice had led to small, slit-like anterior palatal fistulae which were in general asymptomatic. However with the advent of maxillary orthopaedics and better dental arch alignment, these fistulae become wider and increasingly symptomatic (also mentioned in Åbyholm et al 1979).

In addition to fistulae deliberately left unrepaird at the time of primary closure, Reid (1962) listed several other causes of fistula formation:
i. Tension at the site of closure. This is often due to difficulty in sufficiently mobilising the nasal mucosal flaps.

ii. Necrosis of the anterior part of the mucoperiosteal flap. This is more likely to occur where the palatine arteries have been ligated.

iii. Haemorrhage and infection are less common causes.

iv. General post-operative anaemia: this occurs most often due to either operating too early, before adequate haemoglobin level has been achieved, or inadequate replacement of operative blood loss.

Reid (1962) also classified fistulae into three types:

i. Simple slit. This is a minimal midline defect such as would arise from a giving way of a suture line over the hard palate.

ii. Small hole—less than 2cm. in diameter. This again is typically midline and has probably resulted from tension on the suture line with localised disruption. There is also the possibility of associated sequestration following exposure of the underlying bone.

iii. Large hole—more than 2cm. diameter. This type may result from the necrosis of the anterior end of a mucoperiosteal flap. There is likely to be a greater sequestration of underlying bone.

Peer et al (1954) reported an incidence of post-operative fistula formation of approximately 15 percent in a sample of 134 patients. Drillien, Ingram and Wilkinson (1966) reported that fistulae occurred more frequently in patients with cleft lip and palate (55 percent) than in those with

1 In addition to these types, fistulae may result from extraction of fissural incisors as well. (Godfrey 1989).

2 Although Bowers and Gruber (1973) pointed out that in many cases the slit-like appearance is due to collapse of the alveolar segments; re-establishment of the maxillary arch dimensions by orthopaedic expansion shows the true size of the fistula and often substantial bony defect associated with it.
cleft palate only (20 percent). However, a significant proportion of the former were also fistulae at the junction of the hard and soft palates and would thus not be affected by alveolar grafting procedures.

After a von Langenbeck procedure in 66 patients operated upon for unilateral and bilateral complete clefts, Lindsay (1971) reported 16 percent and 23 percent fistulae, respectively; after a push-back procedure in 45 patients with the same cleft types the corresponding percentages of fistulae were 10 and 21 percent. All the fistulae were either alveolar or post-alveolar in position.

Johanson et al (1974), reported a pre-secondary treatment sample of 125 patients where the majority were in the age range 16-49 years. The primary treatment had consisted of lip repair and anterior palate closure at approximately 2 months of age followed by posterior palatal closure at around 24 months of age. A number of supplementary operations on the nose and palate had also been performed. All but 11 of the patients had penetrating vestibulopalatal fistulae.

A retrospective study of 845 cases of cleft lip and palate by Åbyholm et al (1979) found an overall prevalence of fistulae of 18 percent. Fistulae were recorded in 11.3 percent of primary palate and 36.1 percent of all complete clefts. In cases of cleft palate only, fistulae were found in 3.5 percent of incomplete and 20 percent of complete clefts. In primary surgery, lips were closed using a modified Le Mesurier or a Millard procedure at 3-9 months of age. In cases of complete clefts, Yeu's vomer flap operation was applied to close the anterior part of the palate simultaneously with the lip. The posterior part of the palate was closed at a later stage. The patients' age at palatal closure varied between 18 months and 48 years but 92 percent of the cases were between 18 months and 4 years. In 614 cases palatal closure was performed with von Langenbeck's procedure. In 164 cases, the palatal neurovascular bundles were ligated and divided to increase flap mobility.

In patients with bilateral complete clefts, closure of both sides of the lip and anterior palate in one operation appears to have greatly increased the risk of fistula formation (26 out of 60, or 43

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1 53 cases had complete clefts of the primary palate; 327 had complete clefts of the lip and palate; 465 had clefts of the secondary palate.
percent) when compared to two-stage closure (3 out of 26, or 11.5 percent). Ligation of neurovascular bundles was also associated with a higher incidence of fistula formation. 70.5 percent of the fistulae involved the alveolar area and corresponded to the anterior border of the vomer flap. 113 of the 152 patients with fistulae had symptoms that required surgical intervention.

Turvey et al (1984) also found that in a review of cleft lip and palate patients treated without bone grafting procedures, persistent oronasal fistulae were commonly present in spite of multiple surgical attempts to close them by rotation of soft-tissue flaps.

Enemark et al (1985) reported a study of 62 patients with complete unilateral cleft lip and palate. All patients had primary lip repair at 2 months of age, by the same surgeon, using a Tennison procedure. The hard palate was also closed at this time with double vomerine flaps. The soft palate was closed at 22 months using a Wardill palatoplasty. This method had often resulted in a residual fistula between the buccal sulcus and the anterior hard palate. When reviewed at age 12 years, 89 percent of the patients had a slit-like fistula of the alveolus and hard palate which measured 1-2mm. at its widest point. Interestingly, there was no correlation between size of the fistula and degree of patient discomfort reported. Initially, 52 percent of the patients complained of regurgitation of fluids into the nasal cavity. However, following transverse maxillary expansion for dental arch alignment, a further 37 percent of the patients exhibited symptoms.

Lehman et al (1978) stated that the nasal mucosa adjacent to the fistula undergoes hyperplasia and frequently results in an increased nasal discharge.

4.1.2. Residual dental arch deformity

In the study by Johanson et al (1974), the most common malocclusions were crossbites of varying types. The maxillary segment on the cleft side was often rotated medially, with a lateral crossbite. There was rotation and tilting of the teeth adjacent to the cleft with an associated frontal and
lateral open-bite in some cases. The premaxilla in many cases was found to be retroclined or retruded with a resulting anterior crossbite.

Subtelny (1966) found that symmetrical maxillary and mandibular midline relationship could not be attained in the ungrafted cleft cases since lateral movement was restricted for the central incisor adjacent to the cleft.

Eppliy et al (1986) and Bergland et al (1986) pointed out the mobility of the premaxilla in bilateral cleft lip and palate cases as a significant secondary problem.

Boyne and Sands (1976) described the residual alveolar cleft defect as being characterised by loss of the lateral incisor as a result of previous eruption of the tooth into the cleft. In addition, there is a paucity of osseous support around the central incisors and canines. The small amount of osseous tissue existing along the mesial surface of the erupting canine and along the distal surface of the central incisor gradually diminishes with increasing age of the individual so that, in the adult patient, the defect becomes a periodontal one, with increasing depth of periodontal pocket and increasing loss of bone next to the osseous margin of the cleft.

Enemark et al (1985) reported that “orthodontic indications for bone grafting” were found in 50 percent of the sample. These had consisted mainly of anterior and lateral crossbite as well as tipping of teeth and shifting of midlines.

4.1.3. Speech disturbance

Reidy (1961), reported that alveolar collapse following primary palate repair, particularly when there were gaps in the front teeth, was frequently associated with defects of articulation, lateral “s” hissing and indistinct articulation.
Reid (1962) noted that speech defects tended to occur in "the larger fistulae". These defects were mainly nasal escape and "a nasal quality to the voice".

Morley (1970) stated that even a small fistula may affect the development of normal articulation by preventing the build-up of the intra-oral air pressure for articulation anteriorly and particularly for the alveolar consonants such as [t], [d], [s] and [z]. Pressure for the bilabials [p] and [b] and the labio-dentals [f] and [v] may sometimes be obtained by closure of the fistula with the tongue, whilst air pressure for [k], [g] and [(ʃ)], produced posteriorly to the fistula, is not affected if the palato-pharyngeal sphincter is competent.

Hamlen (1978) noted that in addition to the weakening of the consonants, a "snorting friction of air in the nose" may be substituted for the [s] sound. Furthermore, covering the fistula with the tongue may cause the consonants to be lateralised (i.e. formed by lateral, rather than central, emission of air).

Åbyholm et al (1979) stated that a large fistula can cause nasal air escape with consequent nasality of the voice. Smaller fistulae can cause pronunciation defects, which usually affect the quality of the plosive consonants. This was in keeping with Morley's (1970) description of articulation defects.

In the study by Enemark et al (1985), 13 out of the 62 patients were deemed to have distortion of the 'S' sound related to the oronasal fistula, during a standard speech test which included samples of casual speech.

Robinson (1989) lists two further categories of articulation defect, in addition to the distortion mentioned above:

1. **Substitution**— More posterior sounds such as [k], [g] for [t], [d] and [k] for [s] may be substituted in an effort to prevent the audible anterior air-escape.
ii. Omission - The sound produced in the region of the fistula may be simply omitted.

Although these latter articulation defects are not a direct result of air-escape through the oronasal fistula, they can nevertheless be attributed to its presence.

4.2 Surgical management of the residual palato–alveolar defect.

The surgical management of the residual palato–alveolar defect has been performed by soft-tissue closure, with or without placement of a bone graft.

4.2.1. Methods of soft tissue closure

4.2.1.1. Two-layer closure

The general method of closure of residual palatal fistulae over the years appears to have been a two-layer closure. A "nasal" layer consisting of a mucoperiosteal flap elevated from one edge of the fistula is covered by the mobilisation of a second mucoperiosteal flap (Fig.18). Millard (1976) credits Krimer (1827) with describing this type of closure. Von Langenbeck (1864) had also described a two-layer closure of palatal fistula (Millard 1976). A turnover hinge flap was used for the nasal side. Lateral release incisions along the alveolar ridge allowed midline approximation of mucoperiosteal flaps to form the oral layer (Fig.19). Wassmund (1939) had described three variations of the two-layer closure, for fistulae in various locations (Millard 1976). In each method the nasal lining was turned from the edges of the fistula. The oral layer in one case was a flap similar to that of Krimer. In another modification two sliding palatal mucoperiosteal flaps were apposed in the palate. The third modification employed a labial sulcus flap (Fig. 20).
Holdsworth (1951) had also advocated two-layer closure of residual fistulae (cited in Millard 1976). However, instead of the hinge flap, Holdsworth had advocated closure of the nasal mucosa as a separate layer, followed by the oral mucoperiosteum.

Fig. 18. Krimer’s (1827) two-layer closure of the residual palatal fistula. (from Millard 1976).

Fig. 19. Von Langenbeck’s two-layer closure of palatal fistula. (from Millard 1976).
Fig. 20. Wassmund's modification of the two-layer secondary palatal fistula closure. (from Millard 1976).

Fig. 21. Gabka technique of two-layer closure of the palatal fistulae. (from Millard 1976).
Gabka (1964) (cited in Millard 1976) showed a further series of modifications for two-layer closure of fistulae. The nasal layer was formed using turnover hinge flaps and the oral layer was formed with either a V-Y (for anterior hole), sliding mucoperiosteal (for anterior split) or rotation (for posterior hole) flap (Fig.21).

Reid (1962) also described a local rotation or advancement flap for two layer closure of palatal fistulae. He advocates this method only where the fistula does not extend as far forwards as the alveolus. Extensive mobilisation of the nasal mucoperiosteum is required for the inner layer. Reid suggested this method be used only for small defects. He also found severe bleeding a problem, especially in the adult.

Oneal (1971) described a technique for an anterior vestibular-alveolar fistula, utilising a vomer flap and lateral nasal flap for the nasal layer and a labial gingival mucoperiosteal flap for the oral layer (Fig.22).

Rintaala (1971) described a method of two-layer closure using two hinge flaps at the edges of the fistula (Fig.23). One flap (a), barely the size of the fistula, is turned over to form the nasal layer. A larger flap (b), from the opposite side of the fistula, is drawn and denuded of epithelium before elevation and is subsequently sutured over flap (a) to form the oral layer. The oral surface is dressed with a Squibb "orahesive" bandage and the patient is kept on a liquid diet for two weeks. Re-epithelialisation of the oral surface occurs in 3–5 weeks. Haemorrhage from the raw surface of the flap as well as scar contracture are complications of this method. A palatal plate was used to counteract these complications in growing patients. Rintaala reported 30 successful fistula closures with this method, the largest being about 2cm. in diameter.
Fig. 22  Oneal's technique of two-layer closure of the palatal fistulae. (from Oneal 1971). A- Inferiorly based vomer flap, B- Superiorly based lateral nasal flap, C- Gingival mucoperiosteal flap.

Fig. 23  Rintala's technique of two-layer closure of the palatal fistulae. (from Rintala 1971).
In Åbyholm et al's (1979) series of 113 cases of symptomatic oronal fistulae, secondary repair was carried out by soft tissue closure only, using one of three methods of closure in each case. The vast majority of fistulae were small, according to the authors and did not warrant tongue flaps or more distant flaps for closure.

Fistulae in the anterior part of the palate were closed at the same time as secondary lip-nose correction. After excision of the lip scar, the lip and anterior nasal floor were opened to give wide exposure. The epithelium lining of this fistula was excised and it was often necessary to remove associated nasal polyps. Subperiosteal elevation of nasal lining, skin and mucosa usually allowed reconstruction of the nasal floor with evertting sutures (Fig.24). The oral layer was also closed in most cases by mobilising mucoperiosteal flaps from both sides of the hard palate, if necessary facilitated by undermining through short relaxing incisions along the alveolar ridges. In fistulae extending through the alveolar process, it was sometimes impossible to obtain sufficient mucosa for the anterior closure of the oral layer by this method. In such cases a mucosal flap from the buccal sulcus was rotated and sutured into position (Fig.25). The authors also noted that fistula closure in the alveolar region was very difficult from an oral-only approach. Opening the lip and the alveolar cleft area gave good access for reconstruction of the nasal layer from above. With simultaneous closure of the palatal layer from the oral side, a two-layer closure with some distance between the two epithelial surfaces was achieved. The authors felt that this relation between oral and nasal flaps reduced the risk of recurrence.

If there was no indication for a lip-nose correction and in cases where the fistulae were located posterior to the incisive foramen, an oral approach was used. A two-layer closure was achieved by means of a hinge flap for the nasal closure, which was then covered by a palatal mucoperiosteal flap (Fig.26). In some cases e.g. high palatal vault, where a hinge flap was thought to be unsuitable, the nasal layer was provided where possible by undermining and suturing of the nasal mucoperiosteum. The oral layer was achieved by lateral releasing incisions and approximation of the palatal
mucoperiosteum (Fig. 27a). However, in many cases complete nasal closure was impossible and a single layer closure by approximation was performed (Fig. 27b).

Fig. 24. Two-layer closure in combination with a tip-nose correction. (from Åbyholm et al 1979).

Fig. 25. Buccal sulcus flap and approximation of palatal mucoperiosteal flaps for closure of alveolar fistula. (from Åbyholm et al 1979).

Fig. 26. Closure in two layers with hinge flap and rotation and transposition flap. (from Åbyholm et al 1979).

Fig. 27a. Oral layer approximation. (from Åbyholm et al 1979).

Fig. 27b. Cross-sectional view of single-layer closure by approximation. (from Åbyholm et al 1979).
Åbyholm et al reported a success rate for the fistula closure varying from 85 percent for the two-layer approximation and 74 percent for the two-layer closure combined with lip-nose repair, to 60 percent for the one-layer closure (Table 2.).

In his review of the literature, Witsenburg (1985) pointed out that although numerous methods of fistula closure have been published, the differences between them are often quite small. However, he conceded that these small differences may be of significant importance to the overall results. Witsenburg listed the following as major differences between the various methods, in the evolution of secondary soft tissue closure of palatal fistulae:

i. The total or sub-total re-opening of the lip for easier approach to the fistula.

ii. Precise nasolabial muscle reconstruction.

iii. Possible scar revision.

iv. Simultaneous rhinoplasty.

v. Type of vestibular flap design.

vi. Resection of the inferior turbinate.

vii. Manner of nasal layer closure.

viii. Extent of palatal mucoperiosteal mobilisation.

ix. Application of a palatal pressure device.

4.2.1.2. Single-layer closure.

Reid (1962) described a hinge flap for one-layer closure of palatal fistulae. Reid claimed that this method is ideal for closure of slit-like fistulae as well as small holes i.e. up to 2cm. diameter.
Table 2. Results of secondary correction of fistulae related to the surgical method.
(from Åbyholm et al 1979).

<table>
<thead>
<tr>
<th></th>
<th>Fistula closed</th>
<th>Residual fistulae</th>
<th>End results Fistula closed</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No.</td>
<td>No. %</td>
<td>No.</td>
</tr>
<tr>
<td>Two-layer closure combined</td>
<td>35</td>
<td>26</td>
<td>74.3</td>
</tr>
<tr>
<td>with lip-nose repair</td>
<td>21</td>
<td>14</td>
<td>66.7</td>
</tr>
<tr>
<td>Approximation, two-layer</td>
<td>113</td>
<td>81</td>
<td>71.7</td>
</tr>
<tr>
<td>closure</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Local flaps, two-layer closure</td>
<td>27</td>
<td>14</td>
<td>66.7</td>
</tr>
<tr>
<td>One layer closure</td>
<td>30</td>
<td>18</td>
<td>60.0</td>
</tr>
<tr>
<td>Total</td>
<td>113</td>
<td>81</td>
<td>71.7</td>
</tr>
</tbody>
</table>

Fig.28. Bipedicle flap of Egyedi. (from Egyedi 1976).
The flap is based along one edge of the fistula and turned over the hole so that the raw surface is facing into the oral cavity. This raw surface epithelialises quickly. The need to control haemorrhage from the raw palatal surface, to facilitate rapid epithelialisation, was emphasised.

Egyedi (1976) described a bi-pedicled "bucket handle" flap for closure of fistulae in the premaxillary region, especially in bilateral clefts, where maintenance of vascularity is difficult. The flap is raised from labial alveolar mucoperiosteum and sutured into position as a single-layer oral closure (Fig. 28). Egyedi mentioned the shortening of the vestibular depth and presence of pedicles on the alveolus as two disadvantages of this technique.

4.2.1.3. Tongue Flaps

Guerrero-Santos and Altamirano (1966) reported on the use of tongue flaps for secondary closure of large palatal fistulae. Scar tissue was excised from around bony defect and a hinge flap raised to form the nasal layer. A wide, anteriorly based tongue flap was then raised to form the oral layer. Initially the tongue flap was sutured in place directly, but after detachment had occurred in two patients, they modified their technique to tether the pedicle flap to the dental arch by means of a wire suture.

Guerrero-Santos and Fernandez (1973) suggested an alternative two-stage method, which would reduce the risk of dislodgement. A unilateral anteriorly based tongue flap is first joined to the cheek on the same side. Once blood supply from the cheek has been established, the tongue attachment is divided and swung up to close the palatal fistula in two layers.

Jackson (1972) reported the use of an anteriorly based tongue flap to close the palatal part of wide palato-alveolar fistulae. The nasal layer closure was obtained with flaps raised from the vomer and the lateral walls of the fistula. The tongue flap was anchored at the anterior extremity of the palatal fistula, just behind the alveolus. The anterior oral layer of the closure was obtained with a
buccal mucosal flap (Fig.29). After 3 weeks, the tongue flap was elongated and divided and then inserted posteriorly into the remainder of the palatal defect.

Pigott et al (1984) felt that the tongue flap is a viable alternative in the closure of fistulae where viability of local tissue flaps is not feasible, either due to the size of the fistula or degree of scarring consequent to reduction in vascularity of the surrounding mucoperiosteum. In a sample of 20 patients¹ who had tongue flap repair of fistulae considered by the authors to be inoperable by means of local flaps², 11 patients had complete closure. Four patients required two attempts for successful closure; two required three attempts. A further two patients had small persistent fistulae after partial necrosis of the tongue flap but refused further surgery and one patient had a large persistent fistula due to total necrosis of the tongue flap. It is of interest that Pigott et al, in their discussion of their procedure, speculated that almost half of the cases may have been successfully treated by an island flap based on the palatine artery, as described by Henderson (1982).

4.2.1.4. Other pedicle flaps

Millard (1976) summed up the role of pedicle flaps in cleft surgery most succinctly: "There have been far more distant pedicles transferred to the palate than one would ever imagine. When primary surgery has been well executed or the cleft is standard, no such pedicle gymnastics are necessary, but severe clefts, poor surgery, failure in healing and multiple secondary procedures may use up the local tissue. As anywhere else in the body, if local tissue is not available, distant tissue must be brought in to fill the defect."

Esser (1918) (cited in Millard 1976), had designed a nasolabial skin flap based inferiorly, incorporating the angular artery. The flap was introduced into the mouth through an incision in the

¹ 3 were infants aged 13–27 months; 6 were under 10 years of age; 6 patients were between 10–20 years; 3 patients between 20–30 years and 2 patients were over 30 years old.
² 2 fistulae were less than 1cm. in diameter; 12 fistulae were 1–2cm. in diameter and 6 fistulae were more than 2cm. in diameter.
cheek with the skin surface pointed towards the tongue. The borders of the palatal defect were denuded and the flap had been attached with bronze sutures. The bite was held open until the pedicle was well healed. The pedicle was then divided and the cheek defect closed.

Fig. 29. Diagram to illustrate tongue flap closure of palatal fistula. A, Nasal closure using upturned flaps, tongue flap raised. B, Complete closure. (from Jackson 1972).
Wallace (1966) reported on three cases treated by Esser's skin flap. He outlined the indications for this procedure:

a. The fistula is too large to close with local mucoperiosteal flaps, or these have been tried and have failed.

b. The patient is edentulous, or has a gap between the teeth through which the skin flap can be introduced.

c. The upper alveolus has atrophied and the upper denture, in the absence of adequate suction, either will not stay up or slips about inside the mouth.

d. The beard area does not extend higher on the cheek than the level of the palate.

e. The patient is prepared to accept a scar in the nasolabial line.

Wallace further suggested that the flap could reach well beyond the midline and should usually be taken from the larger side of the face when, as is often the case with old clefts, it is asymmetrical. Provided the cheek was perforated by blunt dissection, no facial nerve damage need occur. Wallace also felt that the parotid gland could always be avoided, ectropion was not a problem and the facial scar was unobtrusive.

Georgiadis et al (1969) suggested that a superiorly-based flap could be wider than that of Esser without fear of ectropion. The flap was taken from the nasolabial fold closest to the defect and was made slightly larger than the defect. Mucosal or mucoperiosteal flaps were turned in to form a nasal layer. If this was not possible, a skin graft was used to line the flap.

O'Connor and McGregor (1972) reported the use of intraoral tube-pedicile flaps of cheek mucosa in the closure of a large secondary defect involving the hard and soft palates.
Millard (1976) credits Ganzer (1917) with first describing the method of closing a traumatic palatal defect by means of a tube-pedicle flap from the patient's inner hairless surface of the arm. Quick (1928) is credited with first using the tube-pedicle flap (from the skin of the neck) in the closure of a cleft fistula. Numerous authors have described as many variations of the tube-pedicle flap. Gillies (1957) used this method extensively in the secondary correction of cleft fistulae residual to the original Gillies-Fry repair. Many of the modifications related to overcoming the problem of trauma or dislodgement from the occlusion or tongue-action (Millard 1976). Schuchardt for example, had advocated a nasolabial path and Cupor had suggested a cervicopectoral or acromiopectoral skin-tube introduced through an incision at the lower border of the mandible (from Millard 1976).

The soft tissue-only closure of the fistula failed to address the problems of residual alveolar deformity and poor maxillary contour with the resulting limitations on nasal reconstruction.

4.2.2. Soft tissue closure with bone grafting.

Stenström and Thilander (1963) described their procedure as follows:

The defect in the maxilla was exposed up to the nostrils and posteriorly for an adequate distance into the palate. The dissected bone margins were roughened by careful chiselling and the cleft was packed with bone chips from the iliac crest. A considerable excess of bone was transplanted to take account of expected reduction during healing. The bone was packed particularly on the anterior aspect of the maxilla to compensate for underdevelopment on the side of the cleft.

Soft tissue covering was achieved on the labial aspect by means of a gingivolabial flap from one or both (for bilateral clefts) sides (Fig. 30a). By dividing the periosteum at its base,
Fig. 30. Diagrammatic representation of closure of palato-alveolar fistula.
(from Stenström and Thilander 1963).

(1). A diagram showing a gingivolabial flap mobilised over the alveolar cleft, which is filled to excess with bone chips. A- The chips build up the underdeveloped maxilla on the cleft side to give a normal contour and are responsible for the rounded bulging of the flap. B- The donor area of the flap is completely covered by the labial mucosa. In the diagram, the donor is left partly uncovered to show the origins of the flap.

(2). Median displacement of a palatal mucoperiosteal flap bridging the cleft. A- The flap bulges because of the underlying bone chips. B- The gingivolabial flap passes between the teeth and is sutured to the palatal flap.
the buccal flap was made mobile enough to pass through the defect between the teeth and reach the palatal flap. The latter was raised from one side and moved over the graft (Fig. 30b). The authors did not describe what, if any, measures were taken to provide a nasal-layer closure.

Jackson (1972) described a two-layer technique of fistula closure, applicable to narrow fistulae of any length and those of moderate width and length. This method involved re-opening the lip defect and thus obtained much better access to the anterior palatal fistula (Fig. 31a). The nasal layer of the closure is obtained by raising flaps from the vomer and the lateral walls of the fistula (Fig. 31a, 31b). On the labial side of the fistula, the floor is formed by in-turning of the adjacent buccal mucosa. Jackson felt it was possible to obtain nasal closure in all cases, even those bilateral ones where the premaxilla had been excised in its entirety at the primary repair with subsequent extensive fistula formation, since the main bulk of the fistula is between the ends of the alveolar segments, the palatal portion usually being narrow. In most cases Veau flaps are raised to close off the palatal portion of the fistula just behind the alveolus (Fig. 31c). There is, at this stage, a wedge-shaped cavity bordered above by the nasal layer and below by the palatal layer, with the base being open anteriorly (Fig. 31d). The labial aspect of the oral layer of closure is performed with a buccal mucosal flap from the same side as the cleft, as shown in Fig. 31e. Care is taken to leave sufficient mucosa lying free of the alveolus. This facilitates closure of the residual defect once the flap is raised and swung over to close the anterior alveolar defect (Fig. 31e). Prior to complete closure anteriorly, the bone chips are inserted to fill the wedge-shaped pocket and to pack the nasal floor; particular attention is paid to filling the area between the ends of the alveolar segments. Bone is also inserted under the alar base along the front of the hypoplastic maxilla and into the lateral wall of the pyriform fossa to aid in correction of the nasal deformity (Fig. 31f, 31g). The secondary defect resulting from the buccal mucosal flap is closed directly. Fig. 32 shows a similar method used by Johanson and Ohlsson (1961), in late-secondary bone grafting of the alveolar cleft.
Fig. 31. Diagrammatic representation of closure of palato-alveolar fistula. (from Lehman et al 1978).

- a. Exposure of maxillary defect.
- b. Elevation of nasal lining from the vomer.
- c. Elevation of palatal flaps.
- d. Wedge-shaped defect between nasal and oral closure.
- e. A buccal mucosal flap is raised to close the anterior opening.
- f. Cancellous bone-chips are placed into the cavity between the nasal and oral layers of closure and also under the alar base if needed.
Jackson et al (1982) suggested that the bone graft enhances the cleft-repair in two ways:

i. The graft acts as a splint which supports the nasal layer superiorly and prevents sagging, thus keeping the suture line in a good position. It also supports the oral layer and prevents it from being forced cranially by the tongue.

ii. The graft fills the large dead space produced by elevation of the nasal layer to its correct position to form the nasal floor and the horizontal position that results when Veau flaps are raised from the concave hard palate to be sutured in the midline. This prevents a large haematoma and consequently reduces the likelihood of infection and wound breakdown.

Lehman et al (1978) listed the “three essentials” of Jackson’s method as, accurate closure of the nasal and mucosal layers; cancellous bone grafts to fill the space between the mucosal layers; the use of a buccal flap for closure of the anterior defect. Excision of all hyperplastic mucosa adjacent to the cleft, especially anteriorly, was also considered important— a point reiterated by Jackson et al (1982). Lehman et al suggested that division of the lip is not essential for access (the anterior approach can also be made through the pyriform aperture) and should be performed only where revision of the original repair is indicated. Jackson et al (1982) however, maintained that most cases required lip revision with reconstruction of the orbicularis. Examination pre-operatively usually revealed the lip to be in continuity with the mucosa of the alveolus and the nose. The method of lip repair was always the Millard rotation-advancement technique. They felt that the critical feature of Jackson’s method is the re-creation in the adult of the original deformity of the primary cleft.

Jackson et al (1982) described a modification to the original procedure. As the vomerine and lateral nasal flaps are raised, the enlarged inferior turbinate as well as the deviated vomerine ridge and cartilagenous nasal septum are resected to eliminate the nasal obstruction which is a feature of these cases. This also facilitates closure of the nasal layer.
Fig. 32. Placement of late-secondary bone graft, with re-opening of lip defect. (a). Graft cavity prepared. (b). Graft cavity filled with cancellous bone. (from Johanson and Ohlsson 1961).
Boyne and Sands (1972) described a technique of secondary closure which varied somewhat from the original method of Stenström and Thilander (1963). An incision is made at the posterior limits of the labial portion of the alveolar cleft and extended down the bony rim to the cervical margin of the adjacent tooth. The incision is then carried distally at this level, behind the premolar teeth, with a relieving incision to the vestibular fold in the molar region and a large mucoperiosteal flap is thus raised and mobilised (Fig. 33). A similar flap is raised on the opposing maxillary segment. The nasal floor is formed from the mucoperiosteum of the walls of the cleft. A variety of palatal flaps are used. A common flap in this method involves incisions extending from the anterior border distally to the second molars, at about 2-3mm. below the gingival margin. A particulate marrow and cancellous bone graft from the iliac crest is packed into the defect (Fig. 34), with the nasal floor as a base. The mobilised labial, buccal and palatal flaps are extended over the graft area and sutured into place (Fig. 35).

Åbyholm et al. (1981) described a surgical technique similar to that of Boyne and Sands (1972). They listed three technical details, which they considered to be of importance in achieving adequate height of the inter-alveolar bone:

i. Complete filling of the alveolar cleft with purely cancellous bone.

ii. The provision of mucoperiosteal flaps large enough to cover the grafts without any tension.

iii. The flaps should be designed to ensure that attached gingiva is used to cover the inferior part of the alveolar defect.

Bergland et al. (1986) and Lilja et al. (1987) also adopted surgical techniques almost identical to that of Boyne and Sands. Several other authors (Bertz 1981, Troxell et al. 1982, Eppley et al. 1986 and Simonsen 1986) presented modifications of Boyne and Sands' technique. The modifications all
related to the nature of the labiobuccal and palatal flaps. The nasal layer was raised in the standard manner in each technique and all the authors adhered to the principle of a watertight two-layer soft tissue closure. None of these authors performed secondary lip-nose revision at the time of fistula closure, other than to pack a bone graft into the affected alar base region.

**Fig.33.** View of surgical site after elevation of labial and buccal flaps. Principal labial flap has been retracted superiorly (A). Periosteum (B) has been raised from bony walls of cleft and is now free to be inverted by suturing to form nasal floor for reception of bone graft. (from Boyne and Sands 1972).

**Fig.34.** Particulate autogenous marrow and cancellous bone graft is placed (arrow) into the cleft area. The graft is extended posteriorly into the palatal area to produce an osseous bridge there as well as in the alveolar region. (from Boyne and Sands 1972).
Fig. 35. Appearance after closure of (a) labial and (b) palatal flaps. (from Boyne and Sands 1972).

(a).

(b).
Bertz (1981) advocated a broad-based labial flap similar to Stenström and Thilander (1963). Palatal closure was achieved by local undermining and approximation; however in wide clefts releasing incisions were carried out along the palatal gingival margins of the teeth. Iliac crest cancellous bone chips were used for the graft.

Troxell et al. (1982) (Fig.36a-f) placed their incisions in the gingival sulcus both labiobuccally and palatally, thus including the interdental papillae in the flaps. Vertical releasing incisions palatally and buccally at the level of the second molars allowed the flaps to be mobilised and approximated anteriorly to affect the oral layer of closure. An iliac crest graft containing cancellous bone and marrow was placed.

Eppler et al. (1986) reported on the secondary closure of bilateral clefts. A variety of methods were advocated, depending on the extent of the residual defect. These included the standard method as described by Boyne and Sands; the use of rotated buccal mucosal flaps for labial closure when the anterior cleft is extensive (Fig.37a) and the bi-pedicled “bucket-handle” flap previously described (Fig.37b), in the more extensive palatal fistulae. In the last situation, Eppler et al. stated a preference for a tongue flap for palatal closure because of a higher incidence of complications due to flap necrosis and vestibular shortening with the bucket-handle flaps. Corticocancellous bone grafts from both iliac crest and calvarium were used.

Simonsen (1986) suggested that a separation of the palatal and labial soft tissue (during repair) into separate flaps, which are then stretched over the reconstructed alveolar ridge, may increase the risk of flap necrosis and rupture at the suture-line. Furthermore, he maintained that rotational labial flaps without attached gingiva compromise periodontal health and prosthetic therapy.
Fig. 36. Diagrammatic representation of closure of palato-alveolar fistula. (from Troxell et al 1982).

a. Pre-operative appearance of unilateral alveolar cleft.

b. Flap design for two-layer closure of defect and buccal and palatal flap advancement.

c. Buccal and palatal advancement flaps sutured in place with four-corner approximation.
d. Bilateral alveolar cleft with outline of palatal and labio-buccal incisions for advancement flaps.

![Diagram of alveolar cleft with incisions]

e. Flap design for two-layer closure and the advancement flaps.

![Diagram of flap design]

f. Placement of graft and four-cornered closure of flaps.

![Diagram of graft placement and closure]
Fig. 37. Diagrammatic representation of closure of palato-alveolar fistula. (from Eppley et al 1986).

a. Labial finger flaps with palatal mucoperiosteal advancement flaps.

b. Bucket-handle flap pedicled from upper lip mucosa.
Simonsen suggested an alternative flap design involving two palatal flaps, each continuous with the labial gingiva on its side of the cleft and a subsequent two-flap closure by approximation:

A horizontal labiobuccal incision is made from the first molar on the cleft side to the distal of the first tooth on the opposite side (Fig.38aA). An oblique relieving incision is then made distal to the first tooth on either side of the cleft and extended to the original cut (Fig.38aB). Palatal incisions are made, in the midline from the soft palate to the posterior border of the cleft (Fig.38bB) and laterally from the pterygoid hamulus forward (Fig.38bA), to join the labial relieving incisions previously made. Where the distance from the most anterior tooth to the cleft margin on each side is more than 1cm., both the labial incisions and the continuation palatally, are placed 2-3mm. medial to the adjacent tooth (Fig.38cA). The two palato-labial flaps are raised up to the posterior limit of the hard palate, preserving the palatine neurovascular bundle, to allow their advancement (Fig.38d). The medial margins of the flaps are cleaved to give a periosteal and a mucosal layer. The two layers are closed separately (Fig.38e). The nasal layer closure is achieved in the standard manner. The manner of bone grafting too, varies from the other techniques described in that an iliac crest bone-plate conforming with the anterior cleft morphology is made; a groove is cut in the lateral pyriform aperture, level with the nasal spine and the bone plate is wedged between this groove, the nasal spine and the upper limit of the cleft. The rest of the cleft including the alveolar process and the region beneath the alar base on the cleft-side is packed with cancellous bone.

Simonsen's technique varies slightly for bilateral clefts in that, firstly, the horizontal labial incision is not continuous in the midline (Fig.39aA) in order to allow vascularisation of the premaxillary soft tissue. The labial relieving incisions and the palatal incisions remain unchanged (Fig.39aB, 39b, 39c). Secondly, a triangular segment is resected from each palatal flap (Fig.39d stippled area) to allow the anterior junction of the flaps to be changed from points P to points Q in the correct V-Y approximation of the flaps (Fig.39e) when they are advanced prior to closure.
Fig. 38. Diagrammatic representation of closure of unilateral palato-alveolar fistula. (from Simonsen 1986).
Fig. 39. Diagrammatic representation of closure of the bilateral palato-alveolar fistula.
(from Simonsen 1986).

(a) 

(b) 

(c) 

(d) 

(e)
Simonsen claimed that post-operative vestibular depth and the attached gingival coverage of the reconstructed alveolar ridge is much improved with this technique. Velar insufficiency, as a sequel of palatal flap advancement, was not found.

Braun and Sotereanos (1981) suggested that in the case of bilateral clefts and large unilateral clefts, a staged reconstruction may reduce the likelihood of vascular compromise and create a better soft tissue bed for the alveolar graft. They suggested obtaining good palatal mucoperiosteal closure at the first stage. This was followed by elimination of fistula, closure of nasal layer and placement of bone graft via a labial intra-oral approach at the second stage.

Jackson (1972) described a technique of bone grafting in the case of wide fistulae where the palatal defect is closed with a tongue flap. The procedure for soft tissue repair was described in section 4.2.1.3. Since there is no palatal cover posteriorly at the time of initial closure, the bone graft is delayed until the second stage, 3 weeks later, when the pedicle is divided and inserted posteriorly. The bone graft is placed through a buccal incision. In cases where expansion of the alveolar segments is required, Jackson tended to do the bone grafting as a separate, third procedure. Alveolar expansion is undertaken after the tongue flap is fully healed. Earlier expansion, in Jackson’s opinion, would only relapse during the 3 weeks between the initial operation and division of the pedicle, since a retention appliance could not be worn.

A posteriorly-based, lateral tongue flap as described by Kinnebrew and Malloy (1983) allowed the placement of a bone graft at the time of initial soft tissue closure. The cleft site is prepared and nasal layer closure achieved in the standard manner. An iliac crest cortical bone strut is wired into position in the anterior part of the cleft. Corticocancellous bone from the ilium is used for the graft and is packed into the cleft defect and under the alar base on the cleft side as previously described. The posterior base of the tongue flap allows the posterior closure of the oral layer to be carried out (first), then the anterior tip of the flap is sutured into position, followed by closure of the oral layer.
Reichert (1970) reported on secondary closure, with bone grafting, in complete clefts of the secondary palate. He felt that the rigid bone graft would resist scar contracture and resulting maxillary arch constriction. A triangular piece of iliac crest bone was wedged in the bony defect, within the two-layer closure. Reichert found significant speech improvement and minimal lateral compression of the maxilla in 150 cases followed for more than 10 years.

4.2.3 Timing of secondary bone grafting

Boyne and Sands (1972) preferred to undertake the secondary bone grafting procedure when the patient is between 9–11 years old, just before the full eruption of the canine teeth. They followed 10 cases for 1–2 years post-operatively. This study led them to conclude that when the procedure is accomplished at this age, the unerupted canine may be moved into the grafted area by orthodontic means and excellent osseous support maintained around the root of the migrating tooth. However, in a later article, Boyne and Sands (1976) suggested that grafting should be carried out before 7 years age to avoid loss of the lateral incisor, which would otherwise erupt into the cleft and be lost through exfoliation. They felt that the particulate marrow and cancellous grafts from the iliac crest were viable and unlike the "nonviable" rib and solid one-piece iliac bone grafts, would not retard growth of the adjacent maxillary segments. Ten-year follow-up of 45 cases led Boyne (1985) to re-state his support for secondary bone grafting between 5 and 6 years of age.

Re-establishment of bony support on the mesial aspect of the canine was found even when the tooth was fully erupted prior to surgery. However, Boyne and Sands (1972) felt that there were problems in orthodontically moving these teeth into the graft in the older patient.

Ábyholm et al (1981) assessed the results of their bone grafting procedure in 69 clefts, mainly on the basis of: firstly, height and structure of bone in the alveolar cleft as observed on dental radiographs (fig.40) and secondly, clinically, by the response of the grafted area to spontaneous migration and orthodontic movement of adjacent teeth resulting in closure of the gap in the dental arch
(fig.41). Hence, for the purpose of analysing the success rate with these criteria, the timing of the surgery was categorised in terms of the stage of eruption of the canine tooth on the cleft side. Interalveolar septal height was divided into four categories for comparison:

1- Septum height approximately normal.

Fig.40. Semi-quantitative assessment of the interalveolar septum by means of radiographs. The arrow indicates crest of alveolus. Type I: Height approximately normal. Type II: Height at least 3/4 of normal height. Type III: Height less than 3/4 of normal height. Failure: No continuous bone bridge across the cleft. (from Åbyholm 1981).

<table>
<thead>
<tr>
<th>TYPE OF INTER-ALVEOLAR SEPTUM</th>
<th>I</th>
<th>II</th>
<th>III</th>
<th>FAILURE</th>
</tr>
</thead>
<tbody>
<tr>
<td>RADIOPHGRAPHS OF REPRESENTATIVE RESULTS</td>
<td></td>
<td></td>
<td></td>
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</tbody>
</table>

Fig.41. Patient with unilateral complete cleft subjected to bone grafting at the age of 13 years, after the eruption of the cleft-side canine. (a) 3 years prior to bone grafting. (b) Radiograph of cleft area prior to bone grafting. (c) 22 months after bone grafting. (d) Radiograph 22 months after bone grafting. (from Åbyholm et al 1981).
II– Septum height at least 3/4 of normal height.

III– Septum height less than 3/4 of normal height.

Failure– No continuous bony bridge spanning the cleft could be identified, even though significant bone formation may have occurred in the depths of the defect.

The results are shown in Table 3., figs. 42 and 43:

The presence of a normal interalveolar septum was found to be inversely related to the stage of eruption of the cleft-side canine at the time of bone grafting. All 13 clefts in which the bone grafts were inserted before eruption of the canine, achieved an acceptable septum. 10 of these were rated "normal". Bone grafts placed after full canine eruption, achieved an acceptable (3/4 of normal or more) septal height in 33 out of 37 clefts. However, in only 8 of these was the interalveolar septum found to be normal. Although complete arch alignment was achieved in all the secondary-graft cases\(^1\), in 5 of the late-secondary cases (Fig.43), complete space-closure could not be achieved. The authors indicated that this was not due to any difficulty in moving teeth through the graft. Rather, they felt an unusually wide initial cleft defect or excessive tooth loss had existed prior to grafting.

Ames et al (1981) also reported a significant difference in the success rate between patients younger than 14 years of age (96 percent) and older patients (64.3 percent).

Åbyholm et al cautioned that although normalisation of the dental arch was obtained when the grafting was done prior to eruption of the cleft-side canine and in most cases this tooth has erupted by 13–14 years of age, the upper age limit is not absolute, since good results were obtained in many older patients.

In a continuation of the above study, Bergland et al (1986) reported on a much larger sample of 292 cleft sites. Fig. 44 shows the results of this evaluation.

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\(^1\) The authors felt that the subsequent eruption of the canine tooth through the graft led to additional osteogenesis and reduced the difficulty of closing the gap orthodontically.
Table 3. The relationship between the stage of eruption of the cleft side canine (C) at the time of bone grafting and the radiographic height of the interalveolar septum in 61 cases (69 clefts) in which C was in final position at the time of evaluation. (from Åbyholm et al 1981).

<table>
<thead>
<tr>
<th>Stage of eruption of cleft side canine (C) at bone grafting</th>
<th>No. of patients</th>
<th>No. of clefts</th>
<th>Age in years at bone grafting Mean (Range)</th>
<th>Gap closed</th>
<th>Gap not closed</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group 1:</td>
<td></td>
<td></td>
<td></td>
<td>Total I</td>
<td>II</td>
</tr>
<tr>
<td>C fully erupted</td>
<td>38</td>
<td>43</td>
<td>13.8 (9.11–18.1)</td>
<td>32</td>
<td>8</td>
</tr>
<tr>
<td>Group 2:</td>
<td></td>
<td></td>
<td></td>
<td>10</td>
<td>6</td>
</tr>
<tr>
<td>C half erupted</td>
<td>10</td>
<td>12</td>
<td>12.5 (11.3–13.1)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Group 3:</td>
<td></td>
<td></td>
<td></td>
<td>13</td>
<td>10</td>
</tr>
<tr>
<td>C not erupted</td>
<td>13</td>
<td>14</td>
<td>11.6 (9.0–13.2)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

In the remaining part of the material (20 clefts), C was not erupted at bone grafting and had not reached final position at evaluation.

Fig.42. The relationship between the stage of eruption of the cleft side canine at bone grafting and the height of the interalveolar septum at evaluation (from Åbyholm et al 1981).

Fig.43. The relationship between the stage of eruption of the cleft side canine at bone grafting and the achievement of orthodontic closure of the dental gap (8 failures are not shown in this histogram). (from Åbyholm et al 1981).
Fig. 44. The relationship between the stage of eruption of the cleft side canine at bone grafting and the height of the interalveolar septum at evaluation. (from Bergland et al 1986).

Fig. 45. The relationship between the stage of eruption of the cleft side canine at bone grafting and the achievement of orthodontic closure of the dental gap. (from Bergland et al 1986).
Bergland used the same criteria for assessment as did Åbyholm. In the late-secondary grafting group, although 85 percent were acceptable, only 37 percent showed normal height of septum. In contrast the secondary grafting group showed 96 percent acceptability with 64 percent showing normal septal height. The cleft-related gap in the dental arch was successfully closed in 90 percent of secondary as opposed to 72 percent of late-secondary cases (Fig.45). There were no significant differences between unilateral and bilateral clefts in regard to the two criteria concerned. Incorporation of the bone graft was found to be consistently more rapid in the younger age group. These authors also concluded that the optimum age for secondary bone grafting is in the range 9–11 years. They added that although some authors had advocated grafting at 5–6 years of age to allow the lateral incisor to migrate into and erupt through the bone graft, the findings of Sillman (1964) and Björk and Skieller (1974, 1976), showing significant sagittal and transverse growth of the maxilla up to age 8–9 years, dissuaded them from undertaking such early-secondary procedures. This rationale was also expressed by Jackson et al (1981).

Enemark et al (1985) also cautioned against bone grafting “too early” in the developmental stage of the cleft-side canine. In their study of 56 cases, 5 out of the 23 patients operated on prior to eruption of the canine showed adverse change in the path of eruption and subsequent retention of these teeth. They pointed out that the average in these 5 patients was 9.6 years in contrast to 10.7 years for the total pre-eruption group. However, in view of the poor correlation between dental and chronological age, it would have been desirable if a comparison had been made of the actual tooth development stages.

Johanson et al (1974) in a follow-up period ranging from 3–14 years, also found alveolar form to be much better when the graft was placed prior to eruption of the canine tooth. In contrast, Lilja et al (1987) reported a lack of correlation between eruption state of the permanent tooth distal to the cleft and the form of the grafted alveolar ridge. However, closer evaluation of the latter’s sample shows that, at worst, the canines would have been only partially erupted at time of grafting.
Hence their conclusion is not relevant to a comparison of results between cases grafted prior to eruption and those where the canine is already fully established in the arch.

Sindet-Pedersen and Enemark (1985) reported on another large sample which was very well distributed in terms of age of patient at operation and cleft type (Tables 4. and 5.). The secondary grafting procedure was performed according to the technique of Simonsen (1986). The follow-up period varied from 5.5–8.5 months. The success of the grafting procedure was assessed in terms of the alveolar bone level at the cleft site, recurrence of oronasal fistula, gingival height and periodontal pocketing of the teeth adjacent to the cleft site (Table 6.). Tables 7. and 8. show the pattern of the post-operative results. The post-operative results were significantly better for each of the criteria tested, in the group grafted prior to the eruption of the canine tooth. The authors attributed this partly to "reduced healing potential" in the older age groups. The notable exception is the cleft which is limited to the lip and alveolus (Table 7., 8.), where optimal results were obtained for all criteria tested, regardless of age group, for virtually all cases. Another interesting finding of this study was the similar distribution of alveolar sulcus shortening for all age groups, again with the exception of the isolated cleft lip and alveolus cases. Table 9. shows the authors' conclusions on the overall success of the procedure for the various cleft types and age groups.

In a retrospective study of 64 canine teeth in 46 patients subjected to secondary alveolar bone grafting, El Deeb et al. (1982) concluded that the prognosis for the canine eruption through the graft appeared to be best in those cases where 1/4–1/2 root formation of this tooth had occurred at the time of graft placement\(^1\). These findings however are not conclusive, in that the radiographs were taken at one-year intervals and were not standardised. This introduces the possibility of significant measurement error.

\(^1\) The patients were in the age range 9–12 years.
Table 4. Age-distribution of patients. (from Sindet-Pedersen and Enemark 1985).

<table>
<thead>
<tr>
<th>Group</th>
<th>Dentition</th>
<th>Age</th>
<th>Mean age (range)</th>
<th>Sex</th>
<th>M</th>
<th>F</th>
<th>n</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>Before eruption of canine</td>
<td>&lt;14 years</td>
<td>&lt;14 years (5–14)</td>
<td>70</td>
<td>26</td>
<td>96</td>
<td></td>
</tr>
<tr>
<td>B</td>
<td>After eruption of canine</td>
<td>&lt;16 years</td>
<td>&lt;16 years (11–15)</td>
<td>75</td>
<td>30</td>
<td>105</td>
<td></td>
</tr>
<tr>
<td>C</td>
<td>After eruption of canine</td>
<td>≥16 years</td>
<td>≥16 years (16–38)</td>
<td>63</td>
<td>29</td>
<td>92</td>
<td></td>
</tr>
</tbody>
</table>

For bilateral clefts, the longest erupted canine caused classification of the patient.

Table 5. Distribution of cleft-types. (from Sindet-Pedersen and Enemark 1985).

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Diagnosis</th>
<th>Group A</th>
<th>Group B</th>
<th>Group C</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip and alveolar process only</td>
<td>Cleft lip and alveolar process only</td>
<td>19</td>
<td>11</td>
<td>2</td>
<td>32</td>
</tr>
<tr>
<td>Unilateral cleft-lip and palate</td>
<td>Unilateral cleft-lip and palate</td>
<td>61</td>
<td>70</td>
<td>67</td>
<td>198</td>
</tr>
<tr>
<td>Bilateral cleft</td>
<td>Bilateral cleft</td>
<td>16</td>
<td>24</td>
<td>23</td>
<td>63</td>
</tr>
</tbody>
</table>

Total* 96 105 92 293
Percentage of total number of patients 32.8% 35.8% 31.4% 100%
* The total number of clefts was 356.


<table>
<thead>
<tr>
<th>Findings</th>
<th>Findings</th>
<th>Marginal bone-level</th>
<th>Recurrence of oro-nasal fistula</th>
<th>Periodontal pathology or gingival retraction</th>
<th>Possibility of total dental rehabilitation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total score</td>
<td>Total score</td>
<td>≥75%</td>
<td>0</td>
<td>≥4 mm</td>
<td>Optimal</td>
</tr>
<tr>
<td>1 Optimal</td>
<td>2 Acceptable</td>
<td>50%</td>
<td>0</td>
<td>+</td>
<td>Acceptable</td>
</tr>
<tr>
<td>3 Unacceptable</td>
<td>4 Failure</td>
<td>&lt;25%</td>
<td>+</td>
<td>+</td>
<td>Impossible</td>
</tr>
</tbody>
</table>

Table 7. Marginal bone-level on teeth adjacent to the cleft. (from Sindet-Pedersen and Enemark 1985).

<table>
<thead>
<tr>
<th>No. of clefts</th>
<th>Cleft lip alveolar process (19)</th>
<th>Cleft lip alveolar process (61)</th>
<th>Total (112)</th>
<th>Cleft lip alveolar process (70)</th>
<th>Total (129)</th>
<th>Cleft lip alveolar process (67)</th>
<th>Total (115)</th>
<th>Cleft lip alveolar process (126)</th>
<th>Total (356)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>18</td>
<td>56</td>
<td>101</td>
<td>11</td>
<td>45</td>
<td>28</td>
<td>84</td>
<td>2</td>
<td>38</td>
</tr>
<tr>
<td>2</td>
<td>1</td>
<td>4</td>
<td>10</td>
<td>1</td>
<td>22</td>
<td>12</td>
<td>34</td>
<td>2</td>
<td>23</td>
</tr>
<tr>
<td>3</td>
<td>-</td>
<td>1</td>
<td>-</td>
<td>2</td>
<td>8</td>
<td>10</td>
<td>-</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>4</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>1</td>
<td>-</td>
<td>1</td>
<td>-</td>
<td>4</td>
<td>8</td>
</tr>
</tbody>
</table>

Mean score: 1.05 1.10 1.16 1.11 1.00 1.41 1.58 1.44 1.00 1.58 1.96 1.72 1.03 1.37 1.61 1.43
Table 8. Other post-operative results. (from Sindet-Pedersen and Enemark 1985).

<table>
<thead>
<tr>
<th>Finding</th>
<th>No. of clefts</th>
<th>Group A (Unil. cleft)</th>
<th>Group B (Unil. cleft)</th>
<th>Group C (Unil. cleft)</th>
<th>Groups A – B + C (Unil. cleft)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Cleft lip alveol. palat. process (112)</td>
<td>Bilat. (24)</td>
<td>Total (136)</td>
<td>Bilat. (40)</td>
</tr>
<tr>
<td>Recurrence of fistula</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Minor gingival retrac.</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Major gingival retrac.</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Periodontal pocket</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Insufficient alveol. sul.</td>
<td>1</td>
<td>9</td>
<td>5</td>
<td>15</td>
<td>1</td>
</tr>
</tbody>
</table>

Minor gingival retraction is < 4 mm. Major gingival retraction is ≥ 4 mm. Periodontal pocket is ≥ 4 mm.


<table>
<thead>
<tr>
<th>Score</th>
<th>No.</th>
<th>Cleft lip alveol. proc. patients</th>
<th>U/ml. cleft lip palate patients</th>
<th>Bilateral</th>
<th>Clefts</th>
<th>Patients</th>
<th>Clefts</th>
<th>Patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group A</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 &amp; 2 Success</td>
<td>19</td>
<td>61</td>
<td>69 (98.4%)</td>
<td>32 (100%)</td>
<td>16</td>
<td>112</td>
<td>96</td>
<td></td>
</tr>
<tr>
<td>3 &amp; 4 Failure</td>
<td>–</td>
<td>–</td>
<td>8 (11.6%)</td>
<td>–</td>
<td>–</td>
<td>1 (0.9%)</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Group B</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 &amp; 2 Success</td>
<td>11</td>
<td>70</td>
<td>62 (88.6%)</td>
<td>36 (75%)</td>
<td>15</td>
<td>109</td>
<td>84.5%</td>
<td>88</td>
</tr>
<tr>
<td>3 &amp; 4 Failure</td>
<td>–</td>
<td>–</td>
<td>8 (11.4%)</td>
<td>–</td>
<td>–</td>
<td>29</td>
<td>15.5%</td>
<td>17</td>
</tr>
<tr>
<td>Group C</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1 &amp; 2 Success</td>
<td>2</td>
<td>67</td>
<td>52 (77.6%)</td>
<td>38 (60.9%)</td>
<td>9</td>
<td>82</td>
<td>71.3%</td>
<td>63</td>
</tr>
<tr>
<td>3 &amp; 4 Failure</td>
<td>–</td>
<td>–</td>
<td>15 (22.4%)</td>
<td>18 (39.1%)</td>
<td>14</td>
<td>33</td>
<td>28.7%</td>
<td>29</td>
</tr>
</tbody>
</table>
4.2.4 Sources of bone for secondary grafting procedures.

Matthews and Grossman (1964) and Matthews et al (1970) reported harvesting cortical plate from the inner table of the ilium, which was contoured and wedged into place in the alveolar defect. A contoured extension was also included for alar-base support. If an oronasal fistula had been closed, the periosteal surface of the graft (which was considered more resistant to infection) was placed against the nasal layer of closure. If no previous fistula existed, then the periosteal surface was placed against the labial layer. Although the graft was successfully incorporated in 89 percent of cases and post-graft stability of the dental arches was also very good, the authors did not mention factors such as tooth eruption into or orthodontic movement of teeth through the graft.

Epstein et al (1970) used split rib onlay grafts, with or without inlay of chips of rib within the defect and reported an 80 percent success in functional and 60 percent success in aesthetic improvement (correction of depressed alar base). However the only criterion for functional success was the unification of the cleft maxilla—factors such as dental arch form and stability, eruption and movement of teeth through the graft were not considered. In fact, these authors noted, in agreement with Pickrell et al (1968), that eruption or orthodontic movement of teeth through the rib graft did not occur. Furthermore early grafting led to notching of the alveolus due to lack of growth and remodelling at the graft site. They consequently suggested that secondary grafting be performed after the full establishment of the secondary dentition. Hogeman et al (1972), also noted the progressive notching of the grafted cleft in cases of primary and early secondary repair with rib grafts. A study of 145 cases followed up to 12 months after operation showed relative success rates of 34 percent for rib and 58 percent for iliac crest cancellous bone. The success rate rose to 98 percent when a buccal onlay cancellous “bar” was added to the cancellous inlay graft.

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1 The grafting procedure was considered a success if there was complete bony union radiographically, there was no oronasal fistula and no change had occurred in the occlusion post-operatively.
Ames et al (1981) found, in a sample of 15 patients with previous autogenous rib grafts, the most common appearance was that of a small strut bridging the cleft. There was no evidence of return of form or function to the cleft. Åbyholm et al (1981) reported initially using either rib or iliac-crest cancellous bone for grafting. However, the rib grafts were abandoned after 3 patients, when the 3 month radiographic and clinical results were found to be less favourable than for the cancellous grafts.

Boyne and Sands (1972, 1976) and Boyne (1974) stated that, in contrast to particulate marrow and cancellous bone grafts, "nonviable" rib or solid one-piece grafts from the ilium could be used only after the major growth and development of the premaxillary region had occurred. This was due to the inability of these "nonviable" grafts to "keep pace" with the growth of adjacent bony segments. Such grafts would therefore be best used in late-secondary procedures. The strong osteogenic properties of the particulate marrow and cancellous bone led to rapid and complete incorporation of the graft. The bone grafts were remodelled to form an apparently normal trabecular pattern and alveolar cortical surfaces of lamellar bone (Boyne 1974). Hence, in cases of less severe maxillary arch distortion, alignment of the segments could be carried out just as easily after as before placement of the bone graft.

Witsenberg (1985), in his review of the literature, noted that autogenous bone from the anterior iliac crest is the most frequently used and advocated source of bone in secondary grafting of residual clefts.

Early in their series, Johanson et al (1974) harvested the graft bone from the iliac crest, but later changed the donor site to the tibia. No comparison was made between the grafts from the different sites and the change was probably due to factors related to the harvesting of the graft bone and the donor site, rather than any differences in the quality of the graft bone itself.
Wolfe and Berkowitz (1983) suggested the use of diploic cancellous bone from the calvarium in preference to the iliac crest cancellous bone for the following reasons:

i. Rapid time of harvesting cancellous bone with an assurance of as much bone as needed.

ii. The donor area is in the same operative field.

iii. Virtual absence of post-operative pain in the donor area and an "invisible" scar.

iv. Shorter length of hospitalisation (1–2 days) as a result of less pain in the donor area.

Turvey (1987) also supported the efficacy of cranial cancellous bone in cleft grafting. He argued that although it may be more brittle than iliac bone, it is more densely cellular and can also be more densely packed. This allowed for a greater density of viable cells in the graft.¹

4.3 Orthodontic treatment associated with secondary bone grafting procedures.

Matthews and Grossman (1964) and Matthews et al (1970) reported the use of fixed acrylic appliances for rapid maxillary expansion. This procedure takes about 3 weeks. The appliance was also modified in bilateral cases, to allow forward movement of the premaxilla. The expansion appliance was left in situ at the time of operation and 7 weeks post-operatively for retention. A removable appliance was worn at night for a further 3 months, followed by either a denture or (in an adult) a fixed bridge. The timing for commencement of pre-surgical orthodontics was not specified. However, the authors did recommend that the secondary grafting be performed after establishment of the full permanent dentition.

¹ This appears to be a contradictory statement, since the research would tend to indicate it is the proportion of vital marrow which determines the viability of the graft itself.
Kling (1961) started the pre-surgical orthodontic treatment "as early as possible" with the aim of achieving good alignment of the different maxillary segments in the dental arch and a good intermaxillary relationship. The first step was to establish a normal inter-incisal relationship, followed by adjustment of the displaced maxillary segments. An "operation plate" was used in the first weeks post-surgery, which was then replaced with a removable retainer. Space closure and/or root- paralleling was carried out after the bone graft had "taken".

The most common pre-surgical orthodontic procedure undertaken by Johanson et al (1974) was the expansion of the maxilla to correct crossbite or regain space for erupting teeth. Inter- and intra-arch traction was used for correction of open-bite. Fixed as well as removable appliances were used. A combined retention/"operation" appliance was used to prevent relapse and protect the palate in the immediate post-operative period. After a few weeks, a conventional removable retainer was worn for at least a year. Permanent prostheses were constructed after the year's retention, if residual gaps existed in the dental arch. In cases with a complete maxillary arch, the removable retainer was gradually ceased over a further twelve month period.

Boyne and Sands (1976) claimed that the use of fresh autogenous particulate marrow and cancellous bone grafts from the iliac crest allowed considerable flexibility in the timing of orthodontic treatment. The maxillary arch expansion could be performed before or after the grafting procedure with equally successful results. Boyne (1985) stated that in patients bone grafted at 5 or 6 years of age, rapid posterior palatal expansion could be accomplished within two months of graft placement. In contrast, where "non viable" grafts such as rib and solid one-piece grafts from the ilium were used, pre-surgical expansion was considered essential since extensive arch alignment was not usually possible after the graft was in place. In the older patient, the optimal time for orthodontic movement of the cleft-side canine tooth was considered to be between eight and twelve weeks after placement of the autogenous marrow graft.
Åbyholm et al (1981), who also used iliac crest cancellous bone grafts, felt that the position of the maxillary segments could not be significantly altered by orthodontic methods once bony union had been established across the cleft. Thus, they advocated that repositioning of the maxillary segments should be done prior to surgery, whereas the alignment of the dental arch could be finished after graft placement.

Enemark et al (1985) commenced pre-surgical orthodontic treatment once the maxillary permanent incisors had erupted. The aim of initial treatment was simply to correct inversions, forced bites and lateral crossbites. Uprighting of tipped incisors and correction of midline deviations were delayed until the alveolar defect had been grafted. Turvey et al (1984) also preferred to start orthodontic alignment about 2–3 weeks after bone grafting. Pre-surgical orthodontics was limited to maxillary arch expansion.

Broude and Waite (1974) advocated repositioning the more "extensively" collapsed maxillary arch segments by means of osteotomy, with lateral rotation and advancement. The cleft grafting procedure would be carried out at a second stage.

4.3.1 Oriented stress and retention of bone graft

The effect of oriented stress on retention of bone volume after transplantation into the cleft was investigated by Chierici (1977), who postulated that it was necessary for bone regeneration. Chierici conducted a controlled experiment in adolescent rhesus monkeys with surgically created unilateral complete cleft palates. He compared the quality of bone regeneration between maxillary segments which were, unsupported, stabilised by a passive appliance or expanded with an active appliance i.e subject to oriented stress. He found that, when split-rib grafts were placed, the clefts subjected to oriented stress had the best bone regeneration. Those with a passive appliance showed

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1 They felt that, ideally, the orthodontic appliances should be in place at the time of surgery. This was carried out, where possible, at 1/2-2/3 root formation of the permanent canine.
inconsistent bone formation. The unsupported segments, where no modification of functional stress occurred, had total failure.

Vagervik (1978), in an uncontrolled clinical study, showed development of new bone across the alveolar cleft in all of 15 maxillary clefts\(^1\) which had been expanded with a palatal wire appliance. The expansion was commenced two weeks after placement of autogenous rib grafts.

Vagervik emphasised the need to subject the cleft area to "stretch" and to stabilise the maxillary segments after secondary alveolar graft placement. She maintained that this created the optimal environment for bone regeneration.

A note-worthy aspect of both Chierici and Vagervik's studies, is the use of rib-grafts. Boyne and Sands (1976) pointed out that in situations where "non viable" grafts such as rib were used, later tooth alignment was difficult. Hence they suggested that maxillary expansion should be performed pre-surgically in these cases. However, neither Chierici nor Vagervik mentioned any problems with tooth movement in their respective studies.

\(^1\)The ages of the patients ranged from 12-20 years.
CHAPTER FIVE

DISCUSSION

5.1 Evaluation of the secondary bone grafting procedure.

As Witsenberg (1985) points out, results of the numerous studies are not easily comparable. With very few exceptions, the studies were uncontrolled, almost always retrospective and rarely uniform with regard to surgeon, treatment protocol and objectives. There was no uniformity in the criteria for success and many of the earlier reports could even be described as anecdotal. Hence, strict analytical principles cannot be applied in the evaluation and comparison of these results.

5.1.1 Dental arch correction

5.1.1.1 Reconstruction of the cleft alveolar ridge.

Johanson et al (1974) reported successful healing of the bone graft in 96 percent of cases. The bone density in the graft corresponded well with the surrounding alveolar bone in 77 percent; bone density was slightly decreased in 13 percent and considerably decreased in 6 percent of the 108 cases grafted. Anterior crossbite prevalence was reduced from 72.2 percent to 3.4 percent; prevalence of buccal cross bite was reduced from 80.4 percent to 26.3 percent.

Jackson et al (1982) reported an 80 percent bone graft survival at an average follow-up of 7.5 years, in a sample of 112 cases. The criteria for “graft survival” were not defined.

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1 As observed on anterior occlusal and panorex radiographic examination.
In a study of 40 patients aged 6-20 years, with a mean age of 12.7 years, Ames et al. (1981) reported an overall success rate of 85.4 percent. Unilateral and bilateral cleft repairs had similar rates of success. Criteria for success in this study were: radiographic evidence of bone formation bridging and obliterating the alveolar cleft, clinical evidence of complete oronasal fistula closure as tested by periodontal probing and orthodontic movement or eruption of teeth into the previously grafted site at long term follow-up.

Åbyholm et al. (1981) reported complete orthodontic closure of the cleft defect in 13 out of 14 cases where the grafting procedure was performed prior to eruption of the cleft-side canine tooth. Orthodontic closure was also achieved in 32 of the 43 clefts grafted after the eruption of the canine. Even in the 5 cases where the gap could not be fully closed, the authors reported better bone support of the teeth adjacent to the cleft. Furthermore, fissural teeth which would previously have been lost due to lack of bony support were successfully incorporated in the dental arch, resulting in better incisor symmetry. In the continuation of this study, Bergland et al. (1986) reported on 292 cases where final canine position had been achieved. Total space closure was obtained in 90 percent of cases grafted prior to eruption of the canine, compared with 72 percent of cases grafted after the canine had fully erupted.

In 21 patients subject to secondary bone grafting prior to eruption of the canine tooth, Bertz (1981) reported "variable migration" of the canine into the graft in 75 percent of cases. However, post-operative orthodontic treatment was required in all cases to achieve final tooth alignment. Bertz concluded that the graft provided alveolar bone support for the teeth in the cleft region, but did not attempt a quantitative assessment.

Enemark et al. (1985) achieved full orthodontic space closure in 40 percent of 56 patients, two-thirds of whom had the grafting performed prior to eruption of the cleft-side canine. 72 percent of the patients having surgery after eruption of the canine needed later prosthetic treatment. The

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1 When the graft failures were disregarded, orthodontic closure was obtained in 90 percent of the clefts, with prosthetic closure needed in only 10 per cent.
authors stated that although, from a technical point of view, space closure could have been achieved in a higher proportion of patients, it was not performed in a number of cases for aesthetic reasons. The alveolar ridge repair was also noted to allow unrestricted tooth positioning in the vicinity of the cleft.

Sindet-Pederson and Enemark (1985) (see 4.2.3), found the success rate much higher when the bone grafting was performed before eruption of the canine than after (Table 9.). They suggested that bony healing is impaired with increasing age. Repair of clefts restricted to the lip and alveolus, however, were all successful, regardless of the timing of graft. The frequency of complications was much higher in the late-grafting groups (Table 8.). In fact, with the exception of vestibular shortening, no post-operative complications were noted in the secondary-grafting group.

Hillerup et al (1987) described four cases of successful auto-transplantation of teeth into the previously grafted alveolar cleft region. All four patients had repaired complete cleft lip and palate defects and had previously undergone late-secondary repair of the alveolar cleft with incorporation of autogenous iliac crest cancellous bone grafts. The teeth were transplanted 4, 7, 15 and 20 months after bone grafting. Clinically and radiographically, the periodontal attachment appeared normal. Follow-up of 18 months to 4 years showed no morbidity in 3 cases. The fourth case showed external root resorption at 18 months, which was related to periodontal ligament damage during the transplant procedure. The authors claimed that a further 5 teeth were successfully transplanted, including one case where the tooth was removed at the time of the secondary bone-graft procedure and cryopreserved in a special storage medium in liquid nitrogen for 7 months prior to successful transplant into the graft site.

5.1.1.1 Periodontal status following alveolar ridge reconstruction.

Johanson et al (1974) reported that the tooth immediately distal to the cleft had better bony support than the tooth immediately mesial to it. In 90 percent of cases the former tooth showed bony support for at least 80 percent of root length. The remaining 10 percent of cases showed between 80
percent and 66 percent bony attachment. In 85 percent of cases the mesial tooth had bony support less than 66 percent of its root-length, while the remaining 15 percent had only between 33–66 percent bony support.

Åbyholm et al (1981) reported a good alveolar bone height (defined as greater than 3/4 of the "normal" height) in 81 percent of 69 completely treated clefts. Complete lack of bony union was seen in only 8 out of the total of 89 clefts that were grafted. The bone formed in the grafted area was described as a fine trabecular network "hardly distinguishable from normal alveolar bone". In all 13 clefts where the bone grafts were inserted before the eruption of the canine, the tooth erupted into the graft and achieved an acceptable septum. Septal height in 10 of the 13 cases was classified as "normal" (see 4.2.3).

Troxell et al (1982) found that their four-cornered flap technique gave an average width of attached gingiva of 4.3mm. (range 0–10mm.) in the graft region. No periodontal pocket depths more than 4mm. were noted.

Sindet-Pedersen and Enemark (1985) found marginal bone levels to be significantly better when the bone grafting was performed prior to eruption of the canine (Table 7.). The frequency of gingival retraction also increased with increased delay of grafting (Table 8.).

Bergland et al (1986) reported that, in the 292 clefts where final canine tooth or pre-canine fissural tooth position in the cleft had been achieved, 90 percent showed an acceptable alveolar bone height. The alveolar bone height was significantly better and orthodontic space closure was also significantly enhanced, when the graft was placed prior to eruption of the canine tooth (see 4.2.3).

Enemark et al (1985) pointed out that improvement of the gingival topography in the cleft alveolus, as well as removing the gingival irritation caused by a patent oronasal fistula, would improve the periodontal prognosis of the teeth adjacent to the cleft.

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¹ Minimal observation time was 12 months post-operatively.
El Deeb et al (1986)\(^1\) compared the periodontal status of 34 canines that had erupted through the grafted alveolus in 26 cleft lip and palate patients with that of 58 canines that had erupted through a normal alveolus in 29 non-cleft control patients. The graft material was autogenous iliac crest bone. The labial closure was achieved with either a mucogingival or mucobuccal flap. The mean age at time of grafting was 10.5 years; the mean age at eruption of the canine tooth was 11.9 years for the control patients and the contralateral control canines in the unilateral clefts; the mean age at eruption of the canines through the graft was 12.8 years in the unilateral and 13.1 years in the bilateral cases.\(^2\) The periodontal evaluation was carried out at a mean age of 16.2 years for the unilateral clefts and 16.8 years for the bilateral clefts. The control group had a mean age of 16.9 years at evaluation. The probing depth of the gingival sulcus, clinical periodontal attachment level and width of labial attached gingiva were compared:

There was no significant difference between the graft canines and canines of the control patients with respect to probing depth and attachment loss. Although the contralateral control canines in the unilateral cleft cases did have significantly smaller values than the graft canines, for these two parameters, the authors felt it was too small to be of direct clinical significance.

The control canines had almost twice the width of labial attached gingiva as did the graft canines. However, the authors pointed out that the significance of width of attached gingiva to periodontal breakdown has been questioned (Wennstrom et al 1982, Wennstrom and Lindhe 1983).

Although the mucogingival flaps generated more attached gingiva than the mucobuccal flaps, they showed no differences in attachment loss or probing depth.

Finally, the canines erupted through the bone-grafted alveolus were shown to have more than 90 percent of their possible clinical periodontal attachment intact.

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\(^1\) Hinrichs et al (1984) had previously reported very similar findings in the first 18 cases of this study.

\(^2\) El Deeb et al (1982) also reported a mean eruption time of the canine, through the grafted alveolus, of 3.4 years, though range varied from 1.8–5.0 years.
5.1.1.2 Oral hygiene status

Johanson et al (1974) reported an improvement in oral hygiene following secondary repair, as demonstrated by a low Green-Vermillion plaque index (0.53) and a low Löe-Silness gingival index (.62). They attributed this improvement to normalisation of the anterior palatal contour with the closure of the vestibulo-palatal fistula.

Ramstad (1989) reported a study of 50 subjects who had finished treatment for complete unilateral cleft lip and palate at a mean age of 19.5 years, when a fixed partial denture was inserted into the cleft region. He found that the oral hygiene and periodontal health status was worse in relation to the abutment teeth for the prosthesis than for non-abutment control teeth. He concluded from a comparison of abutment and non-abutment canines, that the evidence indicated this situation to be due to the presence of the prosthesis rather than anatomic deviations created by the repaired cleft.

One could argue from these findings that improving gingival contour and eliminating the need for prosthetic intervention, by bone grafting prior to eruption of the cleft-side canine, would improve the long term oral hygiene and periodontal status of the teeth in the cleft region.

5.1.1.2 Stability of dental arch correction.

The dental objectives of Stenström and Thilander (1963) were to obtain an ideal shape of the maxillary dental arch and to ensure the best possible occlusion. Their sample of 19 patients underwent late-secondary bone grafting, after arch alignment and establishment of final occlusion. A space was created for the missing lateral incisor on the cleft side, which would require later bridging. These authors advocated prolonged post-operative retention of the maxillary arch expansion to allow proper healing and reorganisation of the transplanted bone. The two cases considered failures had almost total
relapse of the maxillary expansion. This was considered to be the reason for the severe resorption of the graft in these two patients. Stability of the graft was tested in terms of resistance to flexion in the region of the graft. Although "excellent stability" was achieved at the graft site, when the stability of the maxilla as a whole was tested, the resistance was not much greater than for the unoperated controls. This led Stenström and Thilander to conclude that, in spite of good stability in the region of the cleft, prolonged retention would be essential to prevent relapse of the transverse buccal expansion.

Matthews et al (1970) found that 74 percent of 55 cases subjected to maxillary expansion and mainly late—secondary grafting with autogenous rib, maintained a stable occlusion. The follow-up period varied between 8 months and 9 years, but this was not correlated with the results.

Johanson et al (1974) felt that the bone graft stabilised the premaxilla in bilateral cleft lip and palate cases and eruption of teeth through the graft normalised the alveolar development. The maxillary arch was retained with a removable appliance post—treatment, for one year in cases where prosthetic closure was required and up to two years in cases where full space closure was orthodontically achieved. Over the mean follow-up period of 7.5 years (range 3—14 years), mean relapse of cross bite was insignificant (1.16mm. for molar and 0.06mm. for canine region). No relapse of established overjet and overbite relationships had occurred.

Bertz (1981) claimed that stability of the maxillary segments was improved in a sample of 21 cases, but gave no quantitative assessment nor the period of follow—up.

Åbyholm et al (1981) emphasised the distinction between segment stability and dentoalveolar stability. They found segment relapse was not a significant problem over the follow—up period. Segments repositioned just prior to surgery remained stable without any retention device, following

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1 Stenström and Thilander associated the severe maxillary collapse with poor wear of the removable retainer.
2 The displacement produced by expansion and contraction forces applied in the transverse plane between the second premolars was used to ascertain whether the graft provided greater stability in the maxilla as a whole.
3 Age range 8—18 years, mean age 14.5 years.
successful bone grafting. The dentoalveolar complex, in contrast, had shown a greater tendency toward relapse and the authors attributed this mainly to palatal scar contracture. This was counteracted by the placement of a bonded wire retainer on the palatal surfaces of the teeth, extending two teeth either side of the cleft.

Jackson et al (1982) concluded that bone grafting does not completely stabilise the expanded maxillary arch, although it did appear to reduce the tendency for relapse. They found that in a group of patients who received bone grafts, any removable or fixed prosthetic appliance design could be less extensive without compromising its retentive function.

Enemark and Pederson (1983) and Enemark (1984) reported a delayed surgical/orthodontic procedure, which was claimed to greatly improve the stability of late-secondary (14-17 years) correction of cleft-related severe crossbite and lateral open bite. The surgery was performed in two stages, separated by a period of rapid maxillary expansion. The initial surgery consisted of mobilising the lesser maxillary segment. A rapid maxillary expansion appliance was cemented at the end of this operation. The rapid maxillary expansion was completed in 2-3 weeks, after which conventional fixed appliances and vertical elastics were used to close the lateral open bite. The fistula closure and bone grafting of the alveolar process was carried out as a later procedure. Average movements of the maxillary segments of 8.4mm. transversely and 4.9mm. vertically, were achieved. Follow-up ranging from 1 year 10 months to 3 years 8 months had not shown any significant relapse of the maxillary segments.

The above authors also reported on another group of ten patients treated for the same condition by a one-step surgical procedure, followed by orthodontic tooth alignment. The lesser maxillary segment was repositioned, alveolar defect bone grafted and fistula closed at operation. The initial result had been good, but five year follow-up had shown gingival retraction in six patients and vertical segmental relapse in two others.
Rune et al (1980) conducted an implant study of maxillary stability in 5 patients who had undergone either rapid or slow maxillary expansion followed by secondary bone grafting by the method of Hogeman et al (1972). In follow-up ranging from 1.8-4.6 years, they found that firstly, the expansion brought about complicated three-dimensional movement of the maxillary segments and secondly, a stable relationship between the segments was not achieved by the secondary grafting procedure. The authors speculated however, that the grafts may well have stabilised the adjoining parts of the alveolar segments, whose movements could otherwise have been even greater.

Helms et al (1987) reported a comparative study of three groups of patients with unilateral and bilateral cleft lip and palate, who had undergone primary, secondary and late-secondary bone grafting procedures. The primary grafting had been performed before one year of age, using rib bone. The secondary and late-secondary grafts consisted of autogenous iliac crest cancellous bone. The patients were a minimum of 15 years of age and at least 5 years post-surgery, at the time of comparison. These authors concluded that primary grafting gave a superior end-result to both secondary and late-secondary grafting protocols, based on the following observations:

1. Both primary and secondary graft groups had bony bridging of the alveolar cleft in 85 percent of cases, whereas the late-secondary group showed only 56 percent success.

2. There were no significant differences between the three groups with regard to frequency of anterior crossbite at any time prior to the secondary treatment. At final evaluation, the primary group showed no cases of anterior crossbite, in contrast to 29 percent for the secondary and 10 percent for the late-secondary groups.

3. The primary group had shown a posterior crossbite history in 6 percent compared to 74 percent for the secondary and 56 percent for the late-secondary groups. At final evaluation, the primary group showed no cases of posterior crossbite, in contrast to 53 percent for the secondary

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1 Four patients had bilateral cleft lip and palate and one had a unilateral cleft lip and palate.
and 63 percent for the late-secondary groups.

iv. There were no teeth lost in the primary group due to lack of bone support, whereas 22 percent of the secondary and 59 percent of the late-secondary had lost one or more teeth for this reason.

v. There were no instances of graft failure in the primary group, in contrast to 26 percent for the secondary and 11 percent for the late-secondary group.

vi. When level of bone attachment on the central incisor surface adjacent to the cleft was considered, the primary group showed a mean attachment of 84 percent of total root length, compared with 60 percent for the secondary group and 44 percent for the late-secondary group.

vii. 55 percent of the primary group, 42 percent of the secondary group and 6 percent of the late-secondary group had a canine or lateral incisor present in the grafted area.

The validity of Helms et al’s conclusions however, cannot be accepted at face value for several reasons:

The 'baseline' relative frequencies of anterior and posterior crossbite between the three groups (ii. and iii.) differ substantially from those of larger and in some cases, better standardised studies (Koberg 1973; Robertson and Jolley 1968, 1983; Rehrman et al 1970; Friede and Johanson 1974). A much more detailed analysis of the secondary-grafting groups, with regard to their early surgery as well as a comparison of skeletal and dental relationships with other published samples would be required before they could be accepted as a representative sample for comparison of these different treatment protocols.

The values for the secondary and late-secondary groups, of the various parameters of graft success (i., v., vi., vii.) were again, much lower than for several larger, well structured studies

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1 Helms et al did, in fact, present their conclusions “with recognition of the limitations of the study”.
(Johanson et al. 1974; Åbyholm et al. 1981; Bergland et al. 1986; El Deeb et al. 1982, 1986;).

The orthodontic records of the secondary grafting groups appear not to have been fully documented. In view of the importance of adequate pre- and post-surgical orthodontic treatment and retention for the success of secondary grafting procedures, valid comparisons would be difficult to make in the absence of such records.

Finally, the nature of case selection and sample size in the three groups should also be questioned, especially since not only are the inter-arch relationships achieved in the primary group at odds with other maxillary growth studies, but maxillary dentofacial corrections achieved with rib grafts in these cases are also so much better than reported by other studies (Pickrell et al. 1968, Epstein et al. 1970, Boyne and Sands 1972, Ames et al. 1981, Åbyholm et al. 1981). Ross (1987a) noted in his comparative study of unilateral cleft lip and palate cases from several different centres, that the Chicago group did have better overall facial proportions than the other primary-graft groups. It also compared well with secondary-graft groups in the long term. However, this comparison is tenuous, since the proportion of growth retardation in the secondary-graft group, attributable to primary repair, cannot be quantified.

5.1.2 Closure of the oronasal fistula

Johanson et al. (1974) reported successful closure of fistula in 70 out of 82 cases. The 12 recurrent fistulae were mainly pin-hole size and asymptomatic. Lehman et al. (1978) reported total successful closure in a sample of 19 cases.

Åbyholm et al. (1981) reported successful closure in 24 of the 25 patients (96 percent) with a fistula. This compared favourably with the success rates achieved by Åbyholm in 1979, with a soft

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1 Åbyholm et al. reported a success rate for the fistula closure varying from 85 percent for the two-layer approximation and 74 per cent for the two-layer closure combined with lip-nose repair, to 60 per cent for the one-layer closure.
tissue-only closure. Bergland et al (1986) also reported permanent closure in all but one of the 30 percent of their sample who had an oronasal fistula.

Jackson et al (1982) reported 84 percent success in fistula closure at the first operation. A further 11 percent needed re-operation. 5 percent had a persistent, though asymptomatic, fistula.


Fig.46. Improvement of nasal tip and alar symmetry associated with bone grafting. A. Prior to grafting; B. 1 year post-operatively (from Bergland et al 1986).

Table 10. Pre- and post-operative speech assessment after fistula closure. (from Jackson 1976).

<table>
<thead>
<tr>
<th>Category</th>
<th>Preoperative Assessment</th>
<th>Postoperative Assessment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>1 (1)</td>
<td>1 (1)</td>
</tr>
<tr>
<td>2</td>
<td>6 (6)</td>
<td>12 (10)</td>
</tr>
<tr>
<td>3</td>
<td>9 (10)</td>
<td>15 (16)</td>
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<tr>
<td>4</td>
<td>23 (25)</td>
<td>13 (16)</td>
</tr>
<tr>
<td>5</td>
<td>5 (4)</td>
<td>—</td>
</tr>
</tbody>
</table>

The figures in brackets are those assessed by an independent speech therapist.

| 2 category change | 4 (6) |
| 1 category change | 24 (26) |
| No category change | 16 (12) |

Total: 44 44

The figures in brackets are those assessed by an independent speech therapist.
5.1.3 Facial aesthetics

Åbyholm et al (1981) reported achieving good skeletal support of the alar base following the grafting procedure. They made the purely subjective assessment that it was easier to achieve a satisfactory result in those patients subsequently undergoing final correction of the nasal deformity. Bergland et al (1986) added that, in some cases, a marked spontaneous improvement was found (Fig.46). Nearly all the follow-up studies tended to make a similar subjective assessment.

5.1.4 Speech

Jackson et al (1976) reported on the pre- and post-operative speech assessment of 44 patients aged 8–43 years who had undergone fistula closure with secondary bone grafting as described by Jackson (1972). Many of the patients were noted to have velo-pharyngeal incompetence, but this was not corrected and was assumed to be a constant for the study. Speech was assessed by tone, degree of nasal emission of air and articulation in both single utterances and continuous speech. All patients were noted to achieve immediate speech improvement, with tone change being the most apparent. The majority of patients had improved by one category on the assessment scale (Table 10.). Of the 16 patients who did not change categories, 7 were judged to have a decrease in nasal emission, but not enough to warrant a category change. The remaining 9 patients showed no change in nasal air emission. The authors felt that their results showed the closure of oronasal fistula to be a significant factor in improvement of speech in cleft lip and palate patients.

Jørgensen and Enemark (1977) (cited in Enemark et al 1985) had reported on a sample of 21 patients who exhibited nasal distortion on (s) sound in a standardised speech test which included samples of conversational speech. When re-evaluated 6 months after the secondary bone grafting procedure, only one patient continued to exhibit the sound distortion.
5.1.5 Effect on maxillary growth

Bergland et al (1986) postulated that the development of lateral discrepancy in the maxilla was unlikely, since transverse maxillary growth has virtually ceased by age 8-9 years. Although vertical growth continues until adolescence, no disturbance was noted in the clinical sample.

Ross (1987a), found that the cases bone grafted between 9-11 years did not show any consequent unfavourable maxillary growth anteroposteriorly\(^1\). However, both the 9-11 year group as well as an older, 11-13 year group tended to show a retardation in anterior maxillary vertical growth following bone grafting. Ross suggested that vertical growth effects would be avoided only if the procedure was delayed until age 15 years or later. In view of the fact that the soft tissue repair-only group also showed comparable retardation of maxillary vertical growth, Ross speculated that the vomerine flap repair, rather than the bone graft itself, was the major inhibitory factor. He concluded that assessment for secondary bone grafting should be delayed until late mixed dentition and that in cases of poor vertical maxillary growth tendency, grafting should be delayed even further to minimise the growth retardation.

Semb (1988) also studied the maxillary growth effects of secondary bone grafting in a sample of unilateral cleft lip and palate cases from the same centre (Oslo) as Ross (1987a). Cephalograms taken at 9 years and 16 years were compared: firstly, 28 cases grafted between 8-12 years age relative to a group of 30 non-grafted controls and secondly, a group of 70 cases grafted between 8-15 years, in which a linear regression analysis of maxillary growth versus age at grafting was performed. Semb found no adverse effect on sagittal or vertical maxillary growth due to secondary bone grafting, even when performed as young as 8 or 9 years of age. He refuted the findings of Ross (1987a) and queried their statistical significance.

\(^1\)Radiographic follow-up occurred for a minimum of 3 years post-operatively.
5.1.6 Complications related to surgery

5.1.6.1 Shortening of vestibular sulcus

Boyne and Sands (1972) found shortening of the vestibular sulcus and consequent tension on the philtrum an occasional complication of their labial flap design. This could be corrected by a later vestibuloplasty. Several other authors have also mentioned this complication (Lehman et al 1978, Bertz 1981, Sindet-Pedersen and Enemark 1985).

5.1.6.2 Wound dehiscence and wound infection

Dehiscence of the palatal incision1 with loss of the more superficial fragments of the bone graft was also an occasional problem (Boyne and Sands 1972; Johanson et al 1974; Bertz 1981; Troxell et al 1982; Enemark et al 1985; Lilja et al 1987). The bulk of the graft however still remained intact in these cases.

Broude and Waite (1974) attributed wound dehiscence to either tension in the flap or more likely, to venous congestion and subsequent oedema within the flap. They suggested that daily "milking" of the flap towards its base may help.

Enemark et al (1985) reported a "secondary resorption" of bone from the grafted region 1-2 years after surgery in two cases where a cheek flap had been used for the labial closure, even though 6 months post-operative radiography had shown a well healed, reorganised bone graft. They felt this may have been associated with the lack of attached gingiva due to flap design.

1 The thick, scarred, palatal flaps were found to be difficult to mobilise and appose.
Åbyholm et al (1981) found the only significant complication in their study to be infection of the grafted area, leading to loss of the graft. It was thought to be the direct cause of failure in two cases and may have been a contributory factor in the other six. The authors advocated three measures to reduce the risk of infection. Firstly, bone grafting should be undertaken only in the presence of healthy gingiva and dentition. Secondly, the bone grafts must be completely covered by mucoperiosteal flaps sutured without tension. Lastly, all patients should be given pre- and post-operative penicillin antibiotic cover—although the authors acknowledged that there was no evidence to show that this measure influenced the results. However, the majority of authors tend to advocate antibiotic prophylaxis.

Haag (1977) (cited in Witsenberg 1985) had found a positive relation between a soft, swollen mucosa over the graft post-operatively and the extent of resorption of the grafted bone.

Braunn and Sotereanos (1981) also emphasised the importance of “impeccable oral hygiene” to reduce the likelihood of wound breakdown.

Thompson (1989) feels that although good gingival health prior to surgery is a factor in the success of the grafting procedure, it is not as important a factor as the water tight closure of nasal and oral flaps around the bone graft.

Water tight closure of the nasal layer during surgery has been emphasised by many surgeons, as a critical measure for reducing the risk of post-operative infection of the graft (Epstein et al 1970, Bertz 1981).

5.1.6.3  Donor-site morbidity

A number of complications at the donor site, following the harvesting of bone grafts, have been reported in the literature. The more common intra-, and post-operative problems are haemorrhage, infection, pain and scar formation. Other, less frequent complications have included
disturbance of sensation in the lateral femoral cutaneous nerve and alteration in gait following detachment of gluteal muscle attachments when harvesting bone from the ilium, as well as pleural laceration\(^{1}\) when harvesting bone from the chest (Crockford and Converse 1972; Mrazik, Amato, Leban, Mashberg 1980; Laurie, Kaban, Mulliken and Murray 1984, Keller and Triplett 1987). Herniation of abdominal contents has also been reported after removal of massive full-thickness iliac grafts (Reid 1968). Weikel and Mutoz (1977) reported another rare complication of iliac graft harvesting: Meralgia paresthetica, a syndrome characterised by severe paraesthesia in the branches of the lateral femoral cutaneous nerve due to its entrapment in the scar tissue or sutures.

There do not appear to be any reports in the literature specifically investigating donor-site morbidity related to secondary grafting of alveolar cleft defects. Breine and Johanson (1966) investigated donor-site morbidity related to tibial grafts used in primary cleft repair. They reported that, although no significant radiographic changes were noted in the first two years, the donor tibia was 1-4 mm longer than its unoperated counterpart at three to four years post-operation. Wolfe and Kawamoto (1978) reported that there was no evidence of growth disturbance in cases where the graft was taken before the final fusion of the iliac epiphyses (10-16 years of age), using their method of splitting the iliac crest.

Laurie et al (1984) assessed the post-operative morbidity at the 104 donor sites\(^{2}\) in 72 patients who had undergone craniofacial surgery. 60 patients were studied retrospectively and 12 prospectively by a combination of examination of medical record, patient interview and physical examination:

i. Early morbidity

These investigators found much less early morbidity at the chest donor sites than at the ilium. 9 percent (4 patients) who had more than one rib harvested suffered pleural laceration requiring

\(^{1}\)This may lead to haemothorax and pneumonia.
\(^{2}\)60 iliac crest and 44 rib donor sites.
chest drainage. Otherwise, all the chest wounds had healed primarily and no patients had chest infection or excessive haemorrhage. All patients however, had post-operative pain of a "moderate degree" lasting from 1-8 weeks.

The iliac graft harvesting procedure had resulted in significant intra- and post-operative blood loss\(^1\). 21 percent (3 out of 14) of patients with incisions over the crest showed delayed healing. However, all the patients with medial or lateral incisions had exhibited primary healing. All these patients too, had reported moderate post-operative pain lasting from 2-8 weeks.

Interestingly, those patients who had both rib and iliac crest grafts harvested, remembered the pain from the latter as being the more severe and prolonged.

\[\text{ii. Late morbidity}\]

The average length of the mature chest scar was about 7.5mm. 26 of the 44 chest scars had shown suture track marks and the majority of patients were noted to have a mild chest-wall contour defect on examination. 6.8 percent (3 patients) had persistent chest pain after 2 years.

The average length of the mature hip scar was about 7.7cm. All the secondarily healed incisions over the crest of the ridge resulted in depressed and adherent scars that were painful under belts or clothing with waistbands. 32 percent (13 out of 41) of sagittally split iliac crests showed bony contour abnormality. 10 percent (6 patients) had persistent pain after 2 years, but this was usually brought on by heavy exercise and did not have the same unpleasant quality as did the persistent rib pain. 8.3 percent (5 patients) had hyperaesthesia or anaesthesia over the distribution of the lateral cutaneous nerve. None of the iliac graft patients in this study had gait disturbance after 1 year.

Laurie et al suggested that crestal incisions should be avoided when harvesting bone from the ilium, in order to reduce likelihood of morbidity. However, the medial and lateral incisions have to be

\(^1\) Intra-operative blood loss ranged from 500-800 mls. and post-operative blood loss ranged from 42-493 mls.
placed carefully to avoid interference with the function of underlying muscle. Keller and Triplett (1987) recommended the medial-superior cortex, since it provided ample cortical and cancellous bone and reduced contour abnormalities by avoiding the iliac crest, as well as sparing the attachment of the gluteal muscles with consequent reduction in gait disturbance. Laurie et al found that the significant long-term problem with rib grafts, of an unsightly scar and persistent pain, appeared to be more difficult to relate to purely technical error. They concluded that although early morbidity at iliac donor sites was far greater than for chest, the long-term morbidity for the former was minimal while the latter led to persistent, unpleasant pain in a significant number of cases. In considering the above findings, it must be remembered that only 6 of the 72 subjects were operated on for alveolar cleft. The vast majority of cases had craniofacial deformities which would have required much larger bone grafts and consequently, more invasive procedures at the donor site. Mrazik et al (1980) for example, pointed out that the relatively small amounts of cancellous bone required for bony periodontal defects could be obtained via a 3cm incision, stripping of a small section of the iliac crest and the excision of a piece of cortex sufficient to admit a medium-sized curette.

Thompson (1989) feels that small areas of paraesthesia or anaesthesia due to sectioning of branches of the lateral femoral cutaneous nerve are the only significant long-term complication in his cases of iliac grafts for secondary cleft repair. However this complication does not appear to have caused undue distress in these patients, certainly not enough to warrant discontinuation of the grafting procedure.

Braun and Sotereanos (1981) also stated that lateral femoral cutaneous anaesthesia was not uncommon, but when it did occur, it was usually not uncomfortable for the patient. These authors also suggested that packing the iliac donor site with microfibrillar collagen obviated the need for drains or suction post-operatively.
5.1.6.4 Root resorption

Bergland et al (1986) found in 15 cases, where the bone graft had been placed after eruption of the canine tooth, external root resorption occurred at the cemento–enamel junction, which had been exposed during surgery. Mechanical trauma to root cementum was considered a significant factor. They suggested that this was a further reason for grafting prior to eruption of the canine. Enemark et al (1985) also reported virtually identical resorption in two cases, which was discovered 3–4 years after the surgery. Both teeth were lost. They suggested that this particular complication could be avoided by covering the canine at risk with lyophilised bone at the time of surgery.

Boyne (1985) warned that if fresh autogenous marrow–cancellous grafts are placed against any part of a fully erupted tooth in the adult or the cervical margin of a fully erupted tooth in the child, root resorption may ensue. His ten-year study of 45 cases revealed a single instance of cervical root resorption. This occurred in a central incisor which had been fully erupted at the time of graft placement and was successfully treated with calcium hydroxide.

A degree of apical root resorption was also seen “quite frequently” (Bergland et al 1986), mainly in the central incisor on the cleft side. The patients most severely affected were those with bilateral clefts who received late grafts. The authors attributed this to the relatively little cancellous bone around the roots of the teeth in the pre-maxilla.

5.1.6.5 Eruption of canine tooth through the bone graft.

Bergland et al (1986) found that, in 15 percent of the patients, surgical exposure of the canine tooth was necessary.

Enemark et al (1985) warned that grafting the cleft too early in the development of the canine tooth could lead to unfavourable path of eruption with subsequent retention of the tooth (see 4.2.3).
El Deeb et al (1982) found that, of 64 canines erupting through an alveolar ridge reconstructed with autogenous iliac crest bone, 17 percent required surgical uncovering alone, while 56 percent required orthodontic traction as well.

El Deeb et al (1986), in a second study, reported that of 34 canine teeth erupting through an alveolar ridge reconstructed with autogenous iliac crest bone, 9 percent required surgical exposure only. A further 50 percent required orthodontic traction, as well as surgical exposure, to achieve their final position. In contrast, 10 percent of the control canines needed surgical exposure. Several factors were thought to be related to delayed eruption and impaction of canines in the graft, including, the presence of excessive cortical bone in some grafts; scar tissue in the anterior palatal region and the absence of lateral incisors. The last factor was thought to result in a loss of eruptive guidance for the canine, with an increased risk of palatal impaction.

In contrast, Troxell et al (1982) [95 percent, or 18 out of 19 cases] and Turvey et al (1984) [94 percent, or 16 out of 17 cases], reported much higher rates of unassisted eruption of the adjacent canine tooth into the graft site. Turvey et al also noted that the time interval from graft placement to eruption of canine was much shorter in their sample than generally reported. They speculated that delaying the grafting procedure until just before the normal canine eruption time and removal of any potentially obstructive supernumerary teeth, might explain the improvement in canine eruption.

5.2 Risk/benefit considerations of secondary alveolar bone grafts.

The assessment of the risk/benefit relationship in secondary grafting procedures involves the consideration of the following factors:
Firstly, 'negative' factors such as donor-site morbidity related to harvesting of the bone graft; the disruption to the lives of the patients and their families of yet another operation.\footnote{The financial cost to the community, as well as the allocation of health-care resources is also a question that needs to be addressed. However, I feel that this is more an administrative matter and is thus not relevant to a treatise on the actual individual health consideration of a clinical procedure.}

Secondly, the potential benefit to the patient in terms of overall facial appearance, as well as the restoration of appearance, structure and function of the maxilla and its dental arch.

Discussion is hampered somewhat by the lack of studies dealing specifically with this issue. Paradoxically this may well be an indication that there are no complications of consequence and the benefits of the secondary grafting outweigh any drawbacks. Donor-site morbidity related to the harvesting of bone grafts has been well documented (see 5.1.6.3). The significant long term morbidity reported tended to be associated with much larger bone grafts and hence more invasive harvesting procedures, than would be required for alveolar cleft repair. According to Thompson (1989), damage to branches of the lateral femoral cutaneous nerve was a significant complication of the alveolar grafting procedure. However, he did not feel this complication was serious enough to warrant abandonment of the procedure.

The physical benefits of the secondary correction have been discussed at length and have been clearly demonstrated\footnote{With the exception of Ross (1987), who stated that in some cases, vertical alveolar development may be retarded due to the placement of a secondary bone graft.} however, are a more complex issue:

Richardson (1963) investigated the perception of a handicap in a group of handicapped, as well as a group of "normal" children aged 10 and 11 years. These two groups showed remarkable uniformity in the preference order for different disabilities: The non-handicapped child was ranked the highest in order of preference; the child with a leg-brace and crutches was ranked second; the child in a wheel-chair was third; the child with a missing left hand was fourth; the child with a slight

\footnote{This refers to the improvement in both dental and overall facial appearance, as well as any improvement in speech and their combined effect on the self-perception of the patient and the perception of the patient by others.}
facial disfigurement was ranked fifth, preferred only to the obese child. A significant functional disability such as confinement to a wheelchair appeared to be preferred to a slight facial disfigurement, even by children thus handicapped.¹

As mentioned previously, Clifford et al (1972), conducted a psychological survey of 98 ‘completed’ cleft lip and palate patients. They found that although overall body-image scores were high, when various items of the body-satisfaction scale were ranked according to the mean satisfaction levels, those associated with clefts rated low. The item with the lowest satisfaction level was teeth, closely followed by speech. Talking, nose and lips had followed in ascending order of satisfaction. Although 65 percent of the sample had received no orthodontic treatment and 76 percent had reported no previous speech therapy, these findings still point out the significance of these factors in the context of total repair of cleft-related deformity.

Shaw et al (1980) described two interrelated ways in which unaesthetic occlusal traits may operate against the psychological well-being of the individual. These are ‘social response’ and ‘poor self-esteem’.

1. Social response

The body of research appears to indicate that physical appearance is important in biasing judgements of social acceptability, ability and personality in both children and adults, as well as the more obvious form of teasing and ridicule in childhood.

Miller (1970), studied the way a group of male and female subjects² correlated a series of seventeen personality traits with photographs of persons of varying physical attractiveness. A consistent pattern was obtained, where low attractiveness was perceived in terms of negative and undesirable traits, while very attractive persons were judged significantly more positively. He

¹ This early study was rather crude in that sample sizes, selection methods and statistical data were not provided.
² 360 male and 360 female undergraduate university students.
concluded that, in a first-impression situation, a person's level of attractiveness may evoke in a perceiver, a consistent set of expectations by a process of trait inference. Dion (1973) found a similar tendency in a group of 3–6 year old children, who were shown a series of photographs of children who had previously been rated attractive or unattractive by a panel of adults. The subjects rated the attractiveness of the children in the photographs in the same way as the adult panel. When asked about having the photographed children as friends, the subjects most often chose the attractive children and indicated a dislike for the unattractive children. Cavior and Dokecki (1973) reported a similar finding in a group of adolescents, in whom physical attractiveness tended to determine popularity for the most and least attractive groups, regardless of whether the faces viewed were known to the judges or not. In contrast, rating of the average-attractiveness group was influenced to a small but significant degree by the judges' prior acquaintance with the respective subjects. Several studies had shown that teachers tended to rate general behaviour and personality less favourably and have lower academic expectations in the case of unattractive children. Richman (1976) studied the relationship between facial appearance in cleft children and the accuracy of teachers' ratings of intellectual ability. A group of 87 children aged 8–14 years, with either cleft lip and palate or cleft palate only, were divided into two groups on the basis of independent ratings of facial appearance. The two groups did not differ significantly with respect to speech, hearing, intellectual behaviour and achievement factors. However, the results of the classroom teachers' estimates of intellectual functioning showed that, while they were accurate in assessing the children with relatively normal appearance, they were significantly less accurate with respect to the children with more severe facial disfigurement. In the latter group of children, the teachers tended to underestimate the ability of the brighter children, relative to their intelligence-test results and interestingly, they overestimated the ability of children who had scored below average.

Goldman and Lewis (1977) reported that in a sample of college students, the physically unattractive were not only judged as less socially attractive, but were in fact less socially skilled.
Shaw et al (1980) stated that the face and oral region in particular, appear to be of primary importance in determining attractiveness. Shaw (1981) found that the biasing effect of the different types of dentofacial anomaly varied with the personality variable. The dentofacial anomaly was seen to detract more from the girls' attractiveness than from the boys. Furthermore, background attractiveness of the face also emerged as possibly a more important modifying factor than the dental anomaly. Attractive children with a dentofacial anomaly, still scored better in many instances than unattractive children with a normal dental appearance.

Tobiasen (1987) reported an elegant study of age- and gender-related personality and ability judgements associated with facial deformity of cleft lip and palate. Background facial configuration was held and the facial deformity varied by showing pictures of the same individuals with and without cleft deformities. The study is especially relevant to the present discussion since the cleft children (six boys and five girls) were in the age groups 9-12 years and 13-16 years, both of which are in the critical age range for secondary and late-secondary bone grafting and related secondary correction of lip and nose. There were 317 subjects in four age groups ranging from 8-16 years age - a much larger sample than most previous studies. The deformed faces rated as significantly less friendly, less popular, less likely choices as friends, less smart and less good-looking than the non-deformed faces, regardless of the subject's age or gender. The cleft deformity tended to detract to a significantly greater degree from the girls' faces than the boys. The forced-choice nature of the questions was a significant limitation of this study.

Shaw et al (1980) pointed out that although the bulk of the studies showing the salience of appearance cues were non-interactive (i.e. the subjects rated photographs and diagrams rather than interacting with people), even where interaction had taken place, differences in physical attractiveness continued to be systematically related to social acceptance.
ii. Self-esteem

The literature pertaining to a subject's self-perception in relation to dental anomaly and body-image appear to be more contradictory than for 'social response'.

McGregor (1951) (cited in Klima et al 1979) had suggested that grossly disfigured people are better adjusted than those with a mild anomaly, e.g. 'buck teeth'. He felt the latter group were more anxiety-prone since unfavourable social responses occurred irregularly.

Watson (1964), found no difference for five measures of personality-disturbance, between a group of 34 boys with cleft lip and palate, aged 8–14 years and peer groups of either normal or chronically handicapped boys. However Watson conceded that this situation may well change with onset of adolescence.

Shaw et al (1980) concluded that the relationship between actual dental arrangement and self esteem would depend on a critical balance between the individual's perception of his own dental appearance, social feedback and his constitutional strength and environmental support. These factors would appear to apply equally well to the cleft lip and palate deformity and its attendant handicaps.

Peter, Chinsky and Fisher (1975) found that a group of cleft adults functioned within normal population range with regard to employment and compared favourably with their non-cleft siblings and random controls, in occupational mobility over the levels attained by their fathers. However, levels of income for the cleft subjects were substantially lower than for control groups. The authors concluded that cleft subjects appear to experience some limitation in their ability to secure vocational and economic rewards from society.

The psychosocial studies therefore tend to indicate that individuals with the cleft condition, though in themselves well adjusted, are significantly handicapped in the context of social
response, because of their dentofacial deformity. This finding reinforces the validity of the traditional aims of cleft treatment as listed by Dey (1974):

The production of normal speech.

The provision of normal appearance.

The procuring of normal dentition.

McLachlan (1985), while acknowledging the need for continued efforts to improve treatment, also pointed out that, if normality is the aim, then we are forced to accept that the ultimate result will be a compromise solution. He speculated that although some cases exhibit cosmetic and speech results which are less than satisfactory (and presumably justifying secondary correction), others may well be due to "unreasonable expectations" on the part of the patient. McLachlan also questioned whether the absence of a mechanism to relate the outcome of treatment to the severity of the original defect is a factor which results in "excessive therapeutic effort", with a diminishing return for each attempt. He posed the question asked at the Jubilee Cleft Palate Clinic of the Royal Alexandra Hospital for Children in 1981:

"When is enough, enough?"

The answer to this question is not to be found in the biological studies, which are quite dispassionate. They tell us whether or not a procedure will achieve its clinical objectives and the nature of any attendant morbidity. The answer lies in the psychosocial studies, which can tell us whether the various therapeutic efforts make a significant contribution to improving the recipients' quality of life. This literature appears to show that the cleft sufferers, when taken as a group, are well-adjusted members of the community. However, any significant deviation from 'normality' appears to act as a social handicap.
Therefore, the consensus of opinion in the psychosocial literature relevant to people with cleft lip and palate, tends to indicate that only 'normality' would be 'enough'.

Three patients who underwent secondary alveolar bone grafting as part of their maxillary arch reconstruction, are presented in Appendix-3. These cases illustrate the benefits of alveolar bone grafting, not only in the "ideal" situation, i.e. mild/moderate defect with cleft-side canine half to two-thirds root-formed at time of grafting, but also in "mutilated" cases where the defect is more severe and the situation complicated by multiple tooth loss and malformation.
CHAPTER SIX

SUMMARY AND CONCLUSION

An important observation in the preceding review of the literature is the vast array of uncontrolled and sometimes uncontrollable variables which could influence the outcome of any programme of cleft treatment. These variables relate to nature of growth in the individual patient; the nature and severity of the defect; the nature, timing and complications of the primary repair and any related orthodontic/orthopaedic treatment; the nature, timing and sequencing of secondary procedures and perhaps most importantly, the skill of the individual surgeon and orthodontist, which may vary from case to case and certainly from one type of procedure to another.

There is, for example, a profound influence of the severity of the initial cleft defect and nature of primary repair on the subsequent development of the facial processes and consequently on the nature, difficulty and outcome of any secondary procedures. Furthermore, the same surgical procedure, when performed by two different surgeons and even by the same surgeon on different occasions, may give quite different results. This can be attributed to variations in factors such as extent of flaps raised, damage to adjacent tissue, extent of haematoma and scarring.

The literature on bone grafting emphasised the importance of correct handling of the graft intra-operatively in maintaining optimum osteogenic potential; but with the exception of Boyne and Sands (1972) and Wolfe and Berkowitz (1983), none of the reports either explained their choice of graft bone or elaborated on their methods of graft handling.

There were other, more subtle problems related to comparison of results between the different techniques and their modifications. These would have been difficult to resolve in clinical studies from both an ethical and logistical point of view. For example, when considering the success
rate of fistula-closure, the relevant studies were not specific enough to definitely state whether the increased success noted with the secondary grafting procedures was in fact due to the bone graft, as suggested by Jackson (1982) and not just a result of the change to a consistent two-layer closure (see 4.2.1.1).

The conclusions reached with regard to the original hypothesis on the efficacy of secondary alveolar bone grafting in the management of the cleft lip and palate condition, have therefore been based, in some aspects, on the broad consensus of opinion in the literature as much as the critical analysis of results:

i. Secondary bone grafting, when performed in conjunction with the appropriate orthodontic/orthopaedic alignment of the maxillary segments, was generally successful in restoring the continuity of the maxillary alveolus. Although most studies had short periods of follow-up, two studies showed high rates of graft stability at average follow-up periods of 7-8 years and another at 10 years.

ii. Reconstruction of the alveolus at the cleft site resulted in good periodontal support for the adjacent teeth and allowed complete orthodontic space closure in a majority of cases. Both the level of bone support and the frequency of full space closure were best when the graft was placed prior to the eruption of the canine tooth on the cleft side. However, even in those cases where full space closure could not be achieved, the improvement in tooth positioning and periodontal support of the adjacent teeth and the consequent simplification of the prosthodontic needs was thought to justify the procedure.

iii. There appears to be unanimous agreement in the literature that secondary grafting performed after 8-9 years of age does not lead to any adverse maxillary growth effects in either the sagittal or coronal planes. With the exception of one study (Ross 1987), the consensus of opinion in the literature appears to be that vertical maxillary growth is not significantly affected either.
iv. There was general agreement that bone grafting stabilised the anterior relation of the maxillary segments. However, there was controversy regarding the stability of the transverse orthodontic/orthopaedic expansion of the buccal segments. The general consensus was that although bone grafting improved the transverse stability of the maxillary segments, long term retention would still be required to avoid relapse of the buccal occlusion. However, the various investigators differed in their expectations of degree of relapse and its clinical significance.

v. The stability of closure of oronasal fistulae in the anterior palate was significantly improved by the placement of the bone graft.

vi. No quantitative analysis of the aesthetic improvement was attempted. However, the universal opinion was that, in addition to the improvement in dental aesthetics, bone grafting to augment bony support of the alar base not only led to a spontaneous improvement in the alar contour, but also greatly enhanced the success of secondary correction of the nasal defect.

vii. The few studies investigating speech benefits of the secondary grafting procedure tended to indicate that an improvement, especially in tonal quality, can be expected.

viii. Successful bony union of the maxillary segments was achieved with all the autogenous graft types reported, i.e. cancellous, cortical and corticocancellous, used in solid-block, particulate or a combination of these two forms. However, adaptation of graft-bone to growth changes in the maxillary segments, alveolar bone support for the teeth in the cleft, tooth eruption and orthodontic tooth movement through the graft were most favourable when a particulate marrow-cancellous graft was placed in the cleft defect. The autogenous marrow-cancellous graft was also the type most commonly reported in the recent literature.

ix. Wound dehiscence and infection of graft site occurred as complications in a small percentage of cases in most of the studies, but rarely led to substantial loss of the graft. Apical root resorption (attributed to orthodontic tooth movement) and inflammatory root resorption at the
cemento-enamel junction of teeth adjacent to the graft site (attributed mainly to surgical trauma), were rare complications. Failure of the canine tooth to erupt through the graft was a more common complication, but in all these cases the tooth was successfully exposed and orthodontically aligned. Vestibular shortening was also a complication of some techniques of soft-tissue closure, but again these appear to have been rectified by a simple vestibuloplasty where needed.

x. The physical and psychosocial benefit to the patient from the successful secondary correction with bone grafting of the alveolar cleft, in general outweigh the negative aspects of undertaking the procedure.

Therefore, it is possible to conclude that secondary alveolar bone grafting is able to achieve and maintain all its stated objectives.
BIBLIOGRAPHY


* Speech pathologist to the cleft palate clinic, Westmead Hospital.


** Plastic surgeon to the cleft palate clinic, Westmead Hospital.


APPENDIX-1

The Kernahan and Stark classification of cleft lip and palate

(from Kernahan and Stark 1958)

1. UNILATERAL SUBTOTAL CLEFT OF PRIMARY PALATE
   Fig. 1

2. UNILATERAL TOTAL CLEFT OF PRIMARY PALATE
   Fig. 2

3. BILATERAL TOTAL CLEFT OF PRIMARY PALATE
   Fig. 3

4. SUBTOTAL CLEFT OF SECONDARY PALATE
   Fig. 4
5. TOTAL CLEFT OF SECONDARY PALATE
   Fig. 5

6. UNILATERAL SUBTOTAL CLEFTS OF PRIMARY AND SECONDARY PALATES
   Fig. 6

7. UNILATERAL TOTAL CLEFT OF PRIMARY AND SECONDARY PALATE
   Fig. 7

8. BILATERAL TOTAL CLEFT OF PRIMARY AND SECONDARY PALATE
   Fig. 8
APPENDIX-2

The American Cleft Palate Association classification of cleft lip and cleft palate

(from Harkins et al 1962)

A CLASSIFICATION OF CLEFT LIP AND CLEFT PALATE

Clefts of prepalate (synonym: cleft of lip and alveolar process; cleft of embryologic primary palate)

Cleft lip (synonym: cleft cheiloschisis)

Unilateral: right, left; extent - 2 3, 3 3 (fig. 1)
Bilateral: right, left; extent - 1 3, 2 3, 3 3 (figs. 2 and 3)
Median: extent - 2 3, 3 3 (fig. 4)
Prolabium: small, medium, large (fig. 3)
Congenital scar (synonym: congenital or prenatal cicatrix): right, left, median; extent - 1 3, 2 3, 3 3 (figs. 5 and 6)

Cleft of alveolar process (synonym: cleft glossochisis)

Unilateral: right, left; extent - 3 3 (fig. 7)
Bilateral: right, left; extent - 1 3, 2 3, 3 3 (fig. 8)

* Cleft extends into nostril; synonym: complete; i.e., involves conformation with widespread clinical practice; however, a cleft lip alveolus extends into the nostril without some indication—between the lobe and midline—of involvement of the alveolar process (refers to figures 1 and 2).

# Width of cleft may be expressed in millimeters.
" Middle (central) part of upper lip (refers to figure 3).
+ Hard palate terminates at incisive foramen (refers to figures 7 to 12).
% Width may be expressed in millimeters.
& Cleft extends to incisive foramen; i.e., into nasal cavity; (synonym: complete) (refers to figures 7 to 11).

Fig. 1. Unilateral cleft lip: left 2 (or left complete)
Fig. 2. Bilateral cleft lip: right 2 (or right complete), left 2 (or left incomplete)
Fig. 3. Bilateral cleft lip: right 2, left 2 (or complete bilateral)

Fig. 4. Median cleft lip 2 (incomplete)
Fig. 5. Congenital lip scar: left 2
Fig. 6. Cleft lip: right 2 (or right complete), left 2; congenital scar: left 2
Fig. 7. Unilateral cleft alveolar process: left, $\frac{3}{4}$ (or left complete).
Fig. 8. Bilateral cleft alveolar process: right $\frac{2}{3}$; left $\frac{2}{3}$ (or complete).
Fig. 9. Unilateral cleft lip and alveolar process, left (or unilateral cleft palate); left, lip $\frac{3}{4}$, alveolar process $\frac{3}{4}$ (or complete).

Fig. 10. Bilateral cleft premaxilla, right $\frac{3}{4}$, left $\frac{3}{4}$ (or complete). Premaxilla protrusion—moderate.
Fig. 11. Bilateral cleft premaxilla. Premaxilla rotation, right—moderate; protrusion—moderate.
Fig. 12. Premaxilla arrest: (median cleft) total. Premaxilla (premaxilla and overlying lip) absent.

Fig. 13. Cleft soft palate: $\frac{1}{4}$ (or bifid uvula, or staphylochiasis)
Fig. 14. Cleft soft palate: $\frac{1}{3}$ (or bifid uvula); with palatal shortness

Median: extent—$\frac{1}{4}$, $\frac{2}{3}$, $\frac{3}{4}$
Submucous: right, left, median

Cleft of premaxilla (synonym: cleft lip and alveolar process)*,

Any combination of foregoing types of cleft:
- Unilateral: right, left; extent—$\frac{3}{4}$, $\frac{2}{3}$, $\frac{3}{4}$ (fig. 9)
- Bilateral: right, left; extent—$\frac{2}{3}$, $\frac{3}{4}$, $\frac{3}{4}$ (figs. 10 and 11)
- Median: extent—$\frac{3}{4}$, $\frac{2}{3}$, $\frac{3}{4}$
Clefts of palate (syonym: cleft of soft and hard palate, i.e., embryologic secondary palate)\(^a\)

**Cleft soft palate (syonym: cleft velum palatini, staphyloschisis veloschisis)**

**Extent:**
- Postero-anterior: \(1/3, \ 2/3, \ 3/3\) (fig. 13)
- Width: maximum (in millimeters)
- Palatal shortness: none, slight, moderate, marked (fig. 14)
- Submucous (occult) cleft: extent --\(1/3, \ 2/3, \ 3/3\) (fig. 15)

**Cleft hard palate (syonym: neuruschisis)\(^b\)**

**Extent:**
- Postero-anterior: \(1/3, \ 2/3, \ 3/3\)
- Width: maximum (in millimeters)

\(^{a}\) Hard palate terminates at incisive foramen.
\(^{b}\) Cleft of soft palate \(1/3\); i.e., equivalent to bilh uvula (refers to figure 13).
\(^{c}\) Synonym: complete; ends at posterior nasal spine (refers to figure 13).
\(^{d}\) Synonym: complete; terminates at incisive foramen (refers to figs. 16 and 17).
Vomer attachment: right, left absent.
Submucous (occult) cleft; extent—$1\frac{1}{3}$, $2\frac{2}{3}$, $3\frac{1}{3}$ (fig. 10)

Cleft soft and hard palate (synonyms: isolated cleft palate, u ranostaphy loschisis, u ranovelaschisis, embryologic secondary palate)

Any combination of clefts of the soft and clefts of hard palate described above (figs. 16 and 17). Such combination should be specified in detail.

Clefts of prepalate and palate (synonyms: embryologic primary and secondary palate; partial syn., gnathop aloschisis)

Any combination of clefts described under "clefts of prepalate" or "clefts of palate" (figs. 18, 19, and 20). Such combination should be specified in detail.

Facial Clefts Other than Prepalatal and Palatal $^1$ (Rare)

Mandibular process clefts

Lip: extent—$1\frac{1}{3}$, $2\frac{2}{3}$, $3\frac{1}{3}$ (figs. 21f and 22)
Mandible: extent—$1\frac{1}{3}$, $2\frac{2}{3}$, $3\frac{1}{3}$ (fig. 21f)
Lip pits: (synonym: congenital lip sinuses) (fig. 23)

Naso-ocular: extending from narial region toward medial angle of palpebral fissure$^m$ (fig. 21a)

Oro-ocular: extending from mouth slit (rima oris) toward palpebral fissure (rima palpebrarum)

Oro-medial canthus—from mouth slit toward medial angle of palpebral fissure (fig. 21b)

Oro-lateral canthus—from mouth slit toward lateral angle of palpebral fissure.$^a$

(fig. 21c). May extend beyond lateral angle toward temporal region (fig. 21d).

Oro-aural: extending from mouth slit toward ear$^p$ (fig. 21c).

$^1$ Less typical, and more severe clefts representing developmental stages of approxi mately 5 1/2 to 6 weeks (or possibly 7 weeks) of the embryonic period.

$^m$ Represents open nasolacrimal groove; usually associated with cleft lip (refers to figure 21a).

$^a$ May involve alveolar process and hard and soft palate (refers to figure 21c).

$^p$ If cleft is sufficiently oblique, alveolar process, and hard and soft palate may be involved (refers to figure 21c).

APPENDIX-3

Three cases of secondary alveolar bone grafting performed at Westmead Hospital are presented below. Secondary alveolar bone grafting has been an integral part of the cleft lip and palate management protocol at Westmead Hospital since 1986.

CASE 1

**Patient:**
HW

**Date of birth:**
19. 11. 1978

**Type of cleft:**
Left unilateral complete cleft lip and palate.

**Primary lip repair:**
1979

**Primary palate repair:**
1980

**Revision of lip and nose:**
1982

**First seen at Westmead Hospital:**
April 1987; aged 8 years and 5 months.

**Pre-graft orthodontic treatment:**
A Quadhelix appliance was placed in June 1987 (aged 8 years and 7 months).

The erupted supernumerary tooth in the cleft was removed in September 1987. An unerupted mesiodens was left in situ at this time, due to difficulty of removal and risk of damage to the adjacent structures. An unerupted supernumerary lateral incisor in the lesser segment was also left in situ. The treatment plan called
for incorporation of this tooth in the dental arch, if possible.

Upper fixed appliance treatment, for the alignment of the maxillary teeth prior to alveolar bone grafting, was commenced in December 1987.

**Bone grafting procedure:**
Maxillary alveolar graft with iliac crest cancellous bone was performed in September 1988 (aged 9 years and 10 months). The cleft-side permanent canine tooth was unerupted and had undergone approximately half root formation at the time of surgery. The maxillary mesiodens was again electively left in situ.

**Post-graft orthodontic treatment:**
A passive quadhelix appliance is in place to maintain arch expansion until the cleft-side canine tooth and the adjacent supernumerary lateral incisor erupt into the arch. Once these teeth erupt, upper and lower fixed appliances will be placed for final alignment and arch coordination. The use of a long term bonded maxillary retainer is envisaged.
PATIENT HW:
MIRROR VIEW OF MAXILLARY ARCH

AT PRESENTATION

PRE-GRAFT

3 MONTHS
POST-GRAFT
PATIENT HW:

AT PRESENTATION

PRE-GRAFT
PATIENT HW:

1 MONTH POST-GRAFT

7 MONTHS POST-GRAFT
PATIENT HW

PERI-APICAL RADIOGRAPHS OF CLEFT:

A. 4 MONTHS PRIOR TO GRAFT

B. 1 MONTH POST-GRAFT
PATIENT HW: LEFT PROFILE

POST-GRAFT
CASE 2

Patient: SF

Date of birth: 20.9.1976

Type of cleft: Left unilateral complete cleft lip and palate.

Primary lip repair: January 1977; aged 4 months.

Primary palate repair: July 1977; aged 10 months.

Revision of lip and nose: -

First seen at Westmead Hospital: November 1984; aged 8 years.

Pre-graft orthodontic treatment: Upper and lower fixed appliance treatment, for initial tooth alignment, commenced May 1985 (aged 8 years and 7 months). Debanded January 1986.

Quadhelix placed for buccal expansion prior to alveolar bone grafting, in February 1986.

Bone grafting procedure: Maxillary alveolar bone graft with iliac crest cancellous bone was performed in July 1986 (aged 9 years and 9 months). The cleft-side canine tooth was almost fully formed and nearly erupted, at the time of surgery.

Post-graft orthodontic treatment: Tooth 23 had erupted palatally. Hence the upper fixed appliance was replaced to align 23.
Post-graft orthodontic treatment:

Tooth 62 showed a pathological degree of mobility just prior to deband, in January 1988 (18 months post-grafting). Radiography showed extensive root resorption of this tooth. Tooth 62 was subsequently extracted on debanding, with immediate insertion of a maxillary retainer.

The patient will now be kept under regular long term review until growth is completed.

There was a positive incisor overjet/overbite relation immediately after the initial repositioning of the maxillary segments. However, this relation had deteriorated to an edge-to-edge situation by 32 months post-graft. This manifestation of a Skeletal III tendency prior to the pubertal growth phase would indicate, at best, an uncertain prognosis for the maintenance of favourable jaw relation. On completion of growth, the orthodontic needs will be reassessed.

Ultimately, complete space closure in the maxillary arch and consequent avoidance of prosthetic tooth replacement would be desirable. However, a discrepancy in future growth between maxilla and mandible would preclude such a result. Finally, a bonded maxillary retainer will be placed.
PATIENT SF:
FRONTAL VIEW

AT PRESENTATION

PRE-GRAFT
(mirror-view; left and right transposed)

20 MONTHS
POST-GRAFT
PATIENT SF:
FRONTAL VIEW IN RETENTION
(32 months post-graft)

Without prosthesis

With prosthesis
PATIENT SF:
MIRROR VIEW OF LEFT SIDE

AT PRESENTATION

PRE-GRRAFT

20 MONTHS
POST-GRRAFT
PATIENT SF:
MIRROR VIEW OF
LEFT SIDE
IN RETENTION

(32 months
post-graft)

With prosthesis
PATIENT SF:
MIRROR VIEW OF MAXILLARY ARCH

AT PRESENTATION

PRE-GRAFT

20 MONTHS POST-GRAFT
PATIENT SF:

MIRROR VIEW OF
MAXILLARY ARCH
IN RETENTION

(32 months
post-graft)

Without
prosthesis

With
prosthesis
PATIENT SF:

AT PRESENTATION

PRE-GRAFT
PATIENT SF:

5 MONTHS POST-GRAFT

32 MONTHS POST-GRAFT (14 months post extraction of 62)
PATIENT SE:

PERI-APICAL RADIOGRAPHS OF CLEFT:

A. Pre-graft.

B. 1 month post-graft.

C. Root resorption of 62 and adjacent bone. (18 months post-graft)

D. Residual bony defect following extraction of 62 (32 months post-graft). Note retention of bony support on cleft-side of 21 and 23.
PATIENT SF

OPPOSITE: AT PRESENTATION

BELOW: IN RETENTION
PATIENT SF: LEFT PROFILE

AT PRESENTATION

POST-GRAFT
PATIENT SF: RIGHT PROFILE

AT PRESENTATION

POST-GRAFT
CASE 3

Patient: SS

Date of birth: 2.11.1976

Type of cleft: Left unilateral complete cleft lip and palate.

Primary lip repair: February 1977; aged 3 months.

Primary palate repair: November 1977; aged 12 months.

Revision of lip and nose: —

First seen at Westmead Hospital: November 1986; aged 10 years.

Pre-graft orthodontic treatment: Quadhelix placed for arch expansion in August 1987 (aged 10 years and 9 months).

Upper fixed appliance treatment, for the alignment of maxillary teeth prior to alveolar bone graft, was commenced in February 1988.

Bone grafting procedure: Maxillary alveolar graft with iliac crest cancellous bone was performed in September 1988 (patient aged 11 years and 10 months). The cleft-side canine tooth had just erupted at the time of surgery.

The original treatment plan had called for bonding a gold chain to unerupted tooth 21 at time of grafting, with subsequent orthodontic alignment. However, at the time of operation, tooth 21 was found to be within the cleft,
significantly malformed and completely surrounded by scar tissue. The prognosis for repositioning tooth 21, either orthodontically or surgically, was considered, by the surgeon, to be poor. This tooth was consequently extracted prior to graft placement. Unfortunately, extraction of the erupted, albeit malformed, supernumerary tooth had been necessary to gain access to tooth 21.

**Post-graft orthodontic treatment:** Post grafting, a bilateral buccal open bite situation existed, with incisor contact only.

Upper and lower fixed appliances with Class III elastics are at present employed to achieve buccal occlusion. A removable maxillary retainer will then be constructed, which will incorporate prosthetic replacement for teeth 21 and 22.

In the long term, a cast, bonded retainer extending to the premolars and incorporating pontics for 21 and 22, is envisaged.
PATIENT SS:
MIRROR VIEW OF MAXILLARY ARCH

AT PRESENTATION

PRE-GRAFT

4 MONTHS POST-GRAFT
PATIENT SS:

AT PRESENTATION

PRE-GRAFT
PATIENT SS:

4 MONTHS POST-GRAFT

8 MONTHS POST-GRAFT
PATIENT SS:

PERI-APICAL RADIOGRAPHS OF CLEFT.

A. Pre-graft

B. 1 month post-graft

C. 8 months post-graft.
PATIENT SS

AT PRESENTATION

POST-GRAFT
PATIENT SS: LEFT PROFILE

AT PRESENTATION

POST-GRAFT
PATIENT SS: RIGHT PROFILE

AT PRESENTATION

POST-GRAFT