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DENTAL ANOMALIES ASSOCIATED WITH

CLEFT LIP AND/OR PALATE

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A thesis submitted for part fulfillment of the requirements for the degree of Master of Dental Science.

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INTRODUCTION

"The human organism is an organism characterized beyond anything else by the infinite number of variations which may affect its particular structures."

Ashley-Montagu (1935).

Boucher (1953:p98) defines a dental anomaly as:--
"an abnormality in which a tooth or teeth have deviated from the normal in form, function or position."

The existence of dental anomalies associated with cleft lip and palate has been known for a long time. The abnormalities include not only the number, shape and structure of the teeth but also their position and nerve and blood supply. There is considerable variation in the occurrence of these abnormalities between one patient and another and this makes the problem a difficult one to evaluate. These dental anomalies are not exclusive to cleft lips and palate but occur in the non-cleft population as well. The difference is that they occur with much greater frequency in the cleft population. (Jordan et al, 1966;p.51).

This study is the introductory phase of a more detailed overall (surgical, dental, orthodontic, speech and related fields) survey now commencing of cleft lip and palate patients treated at Royal Alexandra Hospital for Children, Camperdown over the years 1951 to 1960 inclusive.
The study will be confined to anomalies in form, number and structure occurring in the permanent dentition. Third molars have not been included in the survey.

Malpositions of teeth and anomalies of tooth formation are practically universal in cleft individuals and with their relatively high birth frequency present a substantial dental and orthodontic problem; so much so that in various places, for example, New York State, Denmark, Liverpool (England), there are special services developed to answer the many treatment needs of these afflicted children.

The thesis will take the following form:-

1. Normal development of the face.
2. Mechanism of cleft formation
3. Aetiology of clefts.
4. The development of the teeth.
5. Aberrations in the development of the teeth.
6. Dental anomalies in cleft lip and cleft palate.
7. Aetiology of anomalies in general.
8. Results of pilot survey into anomalies.
9. Discussion.
10. Summary.
11. References.
NORMAL DEVELOPMENT OF THE FACE

INTRODUCTION.

The development of the face and oral cavity involves a dynamic series of events that begin during the second month of intrauterine life. The complex origin of this region from different growth centres makes the relatively infrequent occurrence of malformations remarkable. Critical changes lead to the formation of the embryonic face and to the separation of the oral and nasal cavities by the formation of the palate. This period can be divided into two phases.

In the first phase, which occurs during the fifth and sixth weeks of embryonic life, the building blocks of the face are prepared.

In the second phase, during the seventh and eighth weeks of embryonic existence, the development of the palate takes place, leading to the separation of the oral and nasal cavities.

The most common malformations of the face, cleft lip and cleft palate, originate during the first and second phases respectively.

Three main theories describe the formation of the face. The "classical" theory described by two German anatomists, Dursy and His, has persisted for nearly one hundred years.
The mesodermal penetration theory, advocated by Streeter, may be considered a more modern concept of the embryology of the face. The last theory is a combination of both these theories.


The classical theory states that there are five major processes or masses of ectoderm and mesoderm which develop about the rim of the invaginated oral cavity. The external depression which breaks through to establish the oral opening into the foregut is known as the stomodaeum. It acts as the topographical centre of the developing facial structures. As the stomodael depression is deepened, its ectodermal floor comes to lie against the blind endodermal end of the foregut. This two-layered membrane is known as the bucco-pharyngeal membrane. At the end of the fourth week, this membrane breaks down and thereafter there is no sharp dividing line between the ectoderm and endoderm of the mouth. This leads to some uncertainty as to the layer from which structures in the mouth are derived.

Towards the end of the fourth week, the first external features of the face are becoming apparent. These are the invaginating olfactory placodes, the protruding eye cups, and the first branchial arch or mandibular arch. It is interesting to note that these
features are associated with early development of neural tissues, the neural ectoderm of the olfactory placode, the neural plate of the lens vesicle, and the early occupation of the appropriate branchial arch by its branchial nerve. This indicates the importance of the neural tissues and derivatives in regulating many of the developmental processes of the embryo.

During the fifth week all the major primordia which are involved in the formation of the face and jaws become clearly distinguishable. The frontal process is superior to the oral cavity. On either side of the frontal process, the olfactory placodes have become bordered by rapidly growing horse-shoe shaped elevations so that they appear to have sunk below the general surface and to lie at the bottom of depressions called the nasal pits. The medial limbs of these elevations around the nasal pits are known as the median nasal processes, and the lateral limbs are called the lateral nasal processes.

Growing towards the midline from the cephalo-lateral angles of the oral cavity are the maxillary processes, which are destined to form the lateral parts of the upper jaw.
Caudal to the stomadaeum, on the lateral walls of the pharynx, is a series of elevations with deep grooves between them. These are the rudiments of the branchial arches. The tissue contained in the branchial arches is usually referred to as mesoderm but it is formed by migration downwards of cells from the neural crest. The paired elevations grow and merge with each other. The arch that lies just below the primitive mouth is called the mandibular arch. The two parts of the mandibular arch called mandibular processes grow towards the centre line and fuse together forming the lower lip and jaw.

Other than a short maxillary ridge and the beginning of the primary palate on each side, there is no upper jaw. The lower jaw, as stated above, is definitely laid down and exhibits multiple centres of condensation.

During the sixth week marked progress is made in the development of the upper jaw. The maxillary processes become more prominent and grow toward the midline, crowding the nasal processes closer to each other. The nasal processes have grown to such an extent that they overshadow the frontal process. Now the upper lip and premaxilla are ready to be formed. The two medial nasal processes fuse with each other in the midline and with the maxillary processes.
laterally. The median nasal process gives rise to the premaxilla and the prolabium or median portion of the upper lip. The maxillary processes give rise to the rest of the upper lip and cheeks. The nasolateral process forms the sides and alae of the nose.

The transformation of the right and left olfactory pits leads to the establishment of bilateral nasal passages which by-pass the mouth. A flat plate of epithelium extends backwards from each nasal sac and ultimately provides, by its vertical splitting at the bucco-nasal membrane, continuity with the oral cavity at the posterior nares. The nasal sac becomes more definitely separated from the roof of the mouth by the active proliferation of the mesenchyme of the premaxilla and maxilla which blend across with the corresponding centres of the other side establishing the primordium of the palate.

By the end of the seventh week then, the primitive face is apparent and with it the various structures such as lip, alveolar and palatine processes and dental lamina can be noted.

SIX STAGES IN THE DEVELOPMENT OF THE PRIMARY PALATE.

A and A' Face of a human embryo. Broken line marks plane of Section A'. Medial Nasal Process to fuse with maxillary and lateral nasal process.

B and B' Medial Nasal Process has fused with maxillary process on inferior border of nasal pit. By this fusion an epithelial wall has been formed.

C and C' Medial Nasal Process has fused to maxillary and lateral nasal process. Epithelial wall lengthened. Arrow in C' points to area where epithelial wall separates oral and nasal cavities.

D Mesoderm has strengthened fusion of processes. Epithelial wall thinning at arrow.

E Mesoderm has proliferated (crosses). Epithelial wall thins to form naso-buccal membrane (arrow).

F Nasal Cavity communicates with oral cavity through primary choana (arrow). Superior part of epithelial wall with mesoderm forms primary palate.
FORMATION OF SECONDARY PALATE.

The formation of the palate begins during the sixth week of embryonic life, when the upper jaws have been established. Since the nasal pits break through above the level of the palate, its formation in effect elongates the nasal chambers backwards so that they open eventually into the region where the oral cavity becomes continuous with the pharynx. The palate is contributed to by the median nasal process which forms the premaxilla. The main part of the palate is derived from that portion of the upper jaw which arises from the maxillary processes. The tissue that separates the two primitive nostrils grows backwards and downwards to form the future nasal septum.

The oral cavity now has an incomplete horse-shoe shaped roof formed anteriorly by the primary palate and laterally by the oral surface of the maxillary processes. On either side of the nasal septum, the oral cavity communicates with the nasal cavities.

Shelf-like outgrowths appear at the edges of the maxillary processes and grow downwards on either side of the tongue. At this stage the tongue is narrow and high and reaches to the nasal septum. In time however, the tongue drops lower due to mandibular growth in width and length. Between the eighth and ninth weeks,
the palatal processes begin to arch up over the tongue and lie in a horizontal plane. The shelves do not show appreciable growth during this period, changing merely from the vertical to the horizontal orientation in a wave-like fashion that progresses from a posterior to an anterior direction (Stark 1958). The means by which the palatal shelves proceed from a vertical to a horizontal position has been studied with interest. As early as 1913, it was proposed that the tongue actively elevated the shelves. Recent animal experimentation by Walker and Fraser (1955-1956) made these authors conclude that a "shelf force" seemed to make these processes slide over the tongue from a posterior to an anterior direction. They also noted that the movement of the shelves was too fast to be accomplished by growth alone.

After this positional change, the palatal shelves meet and fuse with each other and with the nasal septum. Contrary to the shelf movement, fusion occurs from an anterior to a posterior direction. Palatal closure occurs at about the 8½ week stage of embryonic development. Anteriorly, the fusion of the premaxilla with the palatal shelves is incomplete, and the incisive foramen remains. This represents the position of the primitive nostrils.
The soft palate is derived from a fold, which arises from the palatal shelves and develops posteriorly towards the pharynx. By the twelfth week, the soft palate and uvula are completed and fusion with the hard palate occurs.

It must be noted that palatal fusion occurs from the incisive foramen posteriorly. A palate that is cleft as far anteriorly as the incisive foramen represents a complete cleft of the palate. Involvement of the alveolus occurs only with a coexisting cleft of the lip. An alveolar cleft is a part of the lip anomaly and is not a part of the cleft of the palate.

**MESODERMAL PENETRATION THEORY.**

Streeter, as quoted by Burston (1959) and Harrison (1957) considered the description of the development of the face by the fusion of a number of processes an oversimplification of the process. He considered it more precise to speak of these processes as swellings or ridges which correspond to the centres of growth in the underlying mesoderm. These mesodermal masses migrate between the ectodermal covering of the face. In this way no ectodermal fusion is called for but rather a smoothing out of ectodermal grooves by the expanding mesoderm beneath. Ectoderm requires the support and nutrition of underlying mesoderm if it is to survive.
Streeter has emphasized that the growth centres are bilateral. They blend with each other and in some cases meet across the midline. This occurs, for example, in the region around the olfactory pit and leads to the formation of the upper lip. The upper lip is thus not differentiated as a separate structure but is formed jointly by coalescence of the centres of two sides.

Streeter agrees that the palate is formed by a process of fusion, as previously described.

Therefore, this theory applies only to the development of the lip, premaxilla and primary septum (The "Primitive Palate"). This tissue is directly under the nasal pits. The ectoderm in this region thickens to form an "epithelial wall". This wall exists initially as the anlage of the upper lip, premaxilla, and upper incisor teeth (primary palate). Three mesodermal volumes are located in it, one in the midline and two laterally. These grow and fuse, forming the normal upper lip, premaxilla and four upper incisor teeth. If one mesodermal volume is absent or deficient the epithelial wall will rupture and a cleft will occur in that area. The mesodermal infiltration is usually complete by the seventh week of foetal life.
The premaxilla develops from the primary palate and is thus a lip structure. The fate of the premaxilla has long been a contentious issue. One school of thought (Callender 1869) holds the view that the maxilla overgrows the premaxilla, leaving the anterior nasal spine as the only vestige of its presence. But Chase (1942) states that in Man these two bones fuse early in development. All skulls at birth and some adult ones show an incisive suture. This suture runs from the septum between the lateral incisor and canine teeth backwards, then mesially to the incisive foramen.

COMBINATION THEORY.

Maurer and Hoepke (1938) have inferred that the facial processes do occur during normal development of the face and their fusion is the mechanism of normal development of the middle face. However, they feel that if a pathologic cleft should occur because of interrupted fusion, the body makes an attempt to heal the cleft and it does so by means of mesodermal penetration. They based their belief on two factors which they found in their cleft embryo: (i) the plug of epithelial cells in the floor of the nares, (ii) "stream of mesoderm" directed towards the nasal plug.
They found mitotic activity in the cells of the nasal plug and felt that the plug was the scaffolding along which the mesoderm penetrates to heal the cleft. Their final proof was the orientation of the stream of mesoderm directed towards the nasal plug. If incomplete penetration of mesoderm occurs, then a web-like band (Simonart's Band) persists.

Stark (1954) quotes both Veau and Ströer as objectors to this theory. Veau, whose interpretation of the mesodermal penetration theory varied, in that he envisaged the epithelial wall as existing from the inception of development, felt that Simonart's band was the result of attenuation and degeneration of this wall after incomplete mesodermal penetration. Ströer doubted the existence of a typical "cell-stream" arrangement of the mesoderm and felt that once a cleft was present, recovery was not possible.

**DISCUSSION OF EMBRYOLOGY.**

It must be remembered that the description of the classical theory of the embryology of the face was based on observations made on non-human material nearly one hundred years ago. Plastic surgeons, like Veau, made several clinical observations, which did not agree with classical concepts.
1. The occasional presence of a Simonart's band across the cleft at the floor of the nares, suggested the resultant of a traction force rather than one of fusion of these regions.

2. Patients who show anatomically intact palates but who possess typical cleft palate speech. This suggests an intrinsic defect of the musculature, which would be due to a mesodermal deficiency.

3. The patient with an incomplete cleft of the lip without a demonstrable defect of the alveolus, may possess no lateral incisor tooth upon the affected side. This reviewer did see such a case himself in the course of his clinical study. But Stark's statement that such an anomaly represented "an abortive form of harelip" must be strongly questioned. It will be shown later, mainly from the work of Woolf et al (1965), that the above statement is questionable. Stark further states that the absence of the lateral incisor indicates a defect of mesoderm as well as ectoderm for the pulp of the tooth arises from mesoderm.

4. The rare occurrence of a median cleft of the upper lip cannot be satisfactorily explained by the classical theory.
The theory of mesodermal penetration is certainly an attractive one and must at the moment be accepted as fundamentally true. But even Stark (1954) admits that a "refinement of details is needed."

From the preceding description of the mesodermal penetration theory, it is now clear that cleft lip and cleft palate are separate and distinct anomalies occurring at different times. Clefts of the lip occur during the seventh week of intrauterine development, while clefts of the palate develop during the ninth or tenth weeks of formation.
MECHANISM OF CLEFT FORMATION

Töndury (1961) states that the mechanism of cleft formation is still under discussion. He feels that none of the theories is sufficient to explain all cases of cleft lip. There are four theories, according to Töndury.

1. Retarded growth of the facial swellings or primary non-fusion hypothesis (Reed).

2. Failure of the epithelial wall to develop (Töndury).

3. Incomplete substitution of the epithelial wall by mesoderm with the result that a secondary pulling apart of the swellings occurs leaving cleft lips with or without bridges (Veau).

4. Ruptures of previously formed cysts resulting in the formation of harelips with soft-tissue bridges (Steniger).

Therefore the harelip can no longer be regarded as a simple arrest-deformity, as the classical theory considers it to be.

Töndury says that the formation of a cleft lip is connected with a defective formation of the nasal cavities. He mentions the existence of an epithelial wall, which extends from the floor of the nasal cavity to the roof of the nasal cavity. This wall has been
formed by the lying together and fusing of the epithelial coverings of the nasal swellings and remains for only a few days and is then lost. Its dissolution and its substitution by mesenchyme forms the primitive palate. Disturbances of these processes account for the formation of a cleft lip.

The theory proposed by Veau varies slightly from the theory of mesodermal penetration. Veau states that the epithelial wall exists from the inception of development. In the mesodermal penetration theory, this epithelial wall is said to form by a thickening of the ectoderm which lies above the oral cavity to form the anlage of the primary palate. If mesoderm fails to penetrate and support this delicate epithelial wall, the wall thins, then pulls apart either completely or incompletely.

According to the mesodermal penetration theory, as reported by Stark (1954), three mesodermal volumes are located in the epithelial wall mentioned above. One of these is situated in the midline and two are lateral. 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 cleft lip. Absence of the medial volume will result in a median cleft of the lip, a rare
anomaly, and absence of the two lateral volumes will cause a bilateral cleft lip to occur.

After studying sections of six cleft human embryos, Stark (1961) states that a lack or even a diminution in mesoderm will lead to the formation of a cleft in the area that is mesoderm-poor, but the exact mechanism is not clear. A relative deficiency of mesoderm on the cleft side was found whenever a cleft of the lip and premaxilla occurred. Since the palatal clefts are midline (Stark 1961) the paucity of mesoderm exists bilaterally and equally. The fact that a relative lack of mesoderm may exist as well as a total lack in a given area, must account for the wide variation in severity of these deformities seen. Should no cleft occur the defective mesoderm may cause defective function.

Tondury mentions the investigations of Steiniger on a family of mice with cleft lip. The latter found 2 week old embryos with cysts in the region of the epithelial wall. The cysts were not present in the newborn cleft animals. Steiniger concluded that such cysts are embryological formations, which up to the time of birth either split open to become clefts or if they are small enough, are filled out in later development.
Burton (1958, 1959) states that cleft formation in the palate is due to:

1. A lack of growth of the palatal processes.
2. Failure of the epithelium of the palatal processes to degenerate preventing the mesoderm to fuse.
3. Failure in co-ordination between the increase of the width of the head as a whole, and the development of the palatal processes.
4. Cystic formation in the degenerating epithelium between the palatal processes. These cysts may rupture producing a cleft.
5. Failure of the mandible and the tongue to descend could prevent the normal development of the palatal processes in the horizontal plane.

Stark (1958) quotes Fraser as postulating that the following factors are important in the pathogenesis of cleft palate:

1. Palatal shelves that are too narrow to meet and fuse.
2. Excessive resistance to fusion of the palatal shelves by the tongue.
3. Interference with a postulated intrinsic force which moves the palatal shelves from a vertically downward position beside the tongue, to a horizontal upward position above the tongue.
4. Excessive width of the head at the time when the palatal processes normally fuse. The "mechanism" behind these various failures in growth processes are still not explained.

DISCUSSION.

The embryology of cleft lip shows an early effect (5 to 6 weeks) of non-penetration of mesoderm. From one to five weeks later (7 to 12 weeks) there may be a similar but not necessarily related happening in the secondary palate. In either instance there is the rare occurrence of incomplete breakdown:

1. Simonart's band of ectoderm at the base of the nostril, which joins two segments of the lip and prevents a complete cleft.

2. "Notching" of the lip.

3. Cleft of the uvula which does not extend into the soft or hard palate and may have little effect on palate function.

4. "Submucous cleft" which is a special case of deficiency of mesoderm in the soft palate without a cleft but characterized in the affected person by cleft-palate speech.

Additional support comes from a consideration of genesis of mid-line clefts.

The reviewer feels that the mesodermal penetration theory, seems at the moment, to best explain the clinical findings by surgeons in cleft patients.
CLASSIFICATION OF CLEFTS

Classification is necessary for uniformity of description, comparison of surgical results and promotion of understanding of these greatly variable defects. The English Classification, suggested by Ritchie and Davis, is an anatomical one, using the alveolar ridge as the line of division between lip and palate anomalies. Thus they had prealveolar clefts as Group I; Group II were postalveolar clefts and those in whom the alveolus was cleft were Group III. Unusual cases were hard to classify.

Stark (1961) proposed a new classification based on newer concepts of the embryology of the centre of the face. He makes the incisive foramen and not the alveolus as the dividing point between the different deformities of cleft lip and cleft palate. There are three distinct groups:

1. Clefts lying anterior to the incisive foramen. These occur in the primary palate and would include minor clefts of the lips or clefts which encircle the premaxilla.

2. Clefts lying posterior to the incisive foramen. These are due to failure of fusion of the palatal shelves and occur in the secondary palate.
3. Clefts which combine both of these. They are due to failure of normal development of both the primary and secondary palates. The words unilateral, bilateral, median and total or subtotal or incomplete are added as necessary to the clefts classified as those of the primary or secondary palates.
AETIOLOGY OF CLEFTS

Aetiology is the study of causes. The aetiology of cleft lip and cleft palate is not clear but two factors are of proven significance.
1. Heredity.
2. Unfavourable maternal environment during the critical period of the first twelve weeks of gestation until the palate of the embryo is formed.

HEREDITY

The primary lesion which leads to a congenital malformation must always be the result of some abnormal functioning of a gene-controlled process. A few years ago all congenital defects were simply dismissed as hereditary accidents.

There is no general agreement on the mechanisms involved in hereditary predisposition to cleft lip and or cleft palate.

Recently Carter (1964) has produced evidence to show that polygenic systems (involving a number of genes) are involved in the occurrence of cleft lip with or without cleft palate. He has suggested that the most plausible explanation of the mode of action of the hereditary predisposing factors is that they make the embryo more susceptible to minor, perhaps haphazard changes, of the intrauterine environment.
Such minor disturbances would be very difficult to detect. Carter considered the best place to search for such environmental factors would be in the families "at-risk" and hoped that once such factors were found it might be possible to see that the children "at-risk" were not exposed to them.

Other authors have produced evidence to suggest that impaired reproductive capacity as manifested by failure to conceive, foetal abnormalities and foetal wastage may show a familial pattern. (Drillen et al 1966).

Since his classic publication in 1942, Fogh-Andersen, the Danish plastic surgeon, has been a great advocate of the importance of heredity in the aetiology of clefts. He reiterates this stand (Fogh-Andersen 1961) in spite of publications supporting the role of exogenous factors. He further shows that there are two different malformations with no genetic connections:

1. Cleft lip with or without cleft palate, most often in males.

2. Cleft palate alone, most often in females.

In 37% (thirty-seven percent) of cases of cleft lip with or without cleft palate and in 19 percent of cleft palate alone it was possible to demonstrate cleft formations in the families.
Other evidence supporting heredity is in the results of twin studies, showing a greater frequency of concordance among monozygotic twins than among dizygotic twins. The most likely manner of inheritance with cleft lip and palate is by "conditioned dominance" with sex limitation to males and the gene occurs generally as a recessive gene but under favourable conditions the heterozygotes are also manifested (Fogh-Andersen 1961).

In localized clefts of the palate alone, the influence of heredity is rather small (Fogh-Andersen 1961). The manner of inheritance is likely to be simple dominance with sex limitation to females and failing manifestation.

Patients with other malformations associated with clefts makes up about 10 percent of all cases (Fogh-Andersen 1961). The cases of cleft lip or cleft palate alone seem to be more frequently associated with other malformations. This indicates that the complicated, multiple malformations may be due to other environmental causes or to genetic mutations or to operation of gene complexes.

Information given to patients and their relations who are concerned by the cleft lip and palate deformity in their family should be based on a careful study of each particular family history,
in an attempt to distinguish between those cases which may be primarily of genetic origin and those of an environmental nature. Advice must be based on risk tables calculated from a population sample (Rank and Thomson 1960:p.688).

ENVIRONMENT.

A vast array of mechanical, chemical, nutritional, infectious and metabolic factors can affect a change in an organism of normal genetic make-up. Yet, there is probably a continual interplay with heredity as it exerts a modifying influence on host susceptibility. It should further be recognized that not only may a single type of injurious agent cause different types of developmental arrest but that a specific type of malformation may be caused by a variety of environmental factors. (Kreshover 1960).

In general, the incidence and type of defect, both oral and extra-oral will vary with the agent used, its dosage, the stage of gestation when it is applied and the genetic constitution of the host animal. (Kreshover 1960).

Kreshover lists the following environmental factors:-

1. Emotional disturbances

2. Physical interference with palatal closure caused by a foetal finger being in the mouth.
3. An inadequate vascular supply to the palatal region.

4. Amniotic bands caught in the lines of embryonic closure.


6. Infectious disease.

7. Radiation.

Stark et al (1962) states that emotional disturbances must be largely discounted since they cannot be evaluated properly. Greene (1963) came to the same conclusion. Stress is known to induce increased function of the adrenal cortex and secretion of hydrocortisone. The production of cleft palate in laboratory animals by injection of cortisone led to the study of mothers subjected to stress during the first trimester of pregnancy. (Strean and Peer 1956).

Harrison (1957) states that it is possible, but not proved that some defect in the embryonic circulation may be responsible for cleft palate, since during the second month the cardiovascular system develops to replace the simple fluid permeation as a means of circulation. Defective development at the periphery of adjoining processes at this time would interfere with fusion, and once the appointed time had passed, growth would increase the gap.
Sanvenero-Rosselli (1953) also suggests that clefts could be due to aberrations in vascularity during the change in blood supply from the stapedial branch of the internal carotid artery to the anastomosis from the external carotid artery, which usually occurs at the time when facial processes are fusing.

In considering the possible effect of malnutrition or disease, it must be remembered that the embryonic face and palate are formed by the tenth week of intrauterine life and that these factors would have to act before that time. Laboratory studies on maternal nutrition showed that vitamin deficiencies can cause clefts. Deficiencies in folio acid, riboflavin, pantothenic acid, vitamin E, nicotinamide and hypervitaminosis A gave rise to abnormal offspring. It should be emphasized that dietary deficiency need only be slight. The extent to which these laboratory findings apply to human beings is difficult to assess.

Abnormalities of the foetus can also be produced as a result of maternal infection during early pregnancy with Rubella. Twelve to twenty five percent of mothers who contract rubella in the first trimester will give birth to a congenitally deformed child (Greeno 1963). Cleft palate does not often occur with this condition, perhaps because the period during which palatal closure occurs extends longer than the infection. There seems to be an apparent
association between toxoplasmosis and cleft palate but no definite conclusions can be drawn at this stage. (Greene 1963).

Irradiation by x-rays has also been proved effective in causing clefts in laboratory animals. It is very interesting to note that no clefts were reported among children exposed in utero during the atomic bombing of Hiroshima (Plummer 1952). The relationship between diagnostic x-rays and congenital clefts has not been reported in the literature.

OTHER CONGENITAL ABNORMALITIES ASSOCIATED WITH CLEFTS.

There is a significantly frequent but unpredictable association of both external and visceral malformations in cleft palate individuals. Any organ or structure or combination of several could be affected and to varying and unpredictable degrees.

McKenzie (1958) considers the above association as a "first arch syndrome". He lists eight features of abnormal development of the first visceral arch in the embryo.

1. The Treacher Collins syndrome or mandibulofacial dysostosis. This condition shows a marked hereditary trait and affects the facial bones.


Affected infants show:

(i) hypoplasia of the mandible.
(iii) glossoptosis - a tendency for the tongue to block the pharynx.

(iii) frequent complication of cleft of the secondary palate.

3. Mandibular dysostosis: micrognathia in varying degrees.

4. Deformities of external and middle ear.

5. Congenital deaf-mutism - caused by an abnormality of the middle ear.

6. Cleft lip and cleft palate.

7. Hypertelorism: a developmental fault thought to lie in the cartilaginous base of the skull.


Seven specific malformations have been found to coexist with facial clefts, according to Steigler and Berry (1958).

1. Syndactyly: webbing or fusion of fingers or toes.

2. Polydactyly: extra fingers or toes.

3. Clubfoot.

4. Malformed ears.


7. Micrognathia.

There is an additional syndrome, which exhibits malformations of the face, oral cavity and digits, and is now generally known as the oro-facial-digital
syndrome. Gorlin and Psaume (1962) found a number of features which occur constantly in this condition.

1. Occurrence in females.
2. Numerous frena in the upper and lower vestibular sulci that cleft the mandibular and maxillary alveolar processes.
3. Clefts of the tongue.
5. Pseudo-cleft of the upper lip.
6. Dental anomalies including malposition, supernumerary teeth, missing teeth especially lower lateral incisors.
7. Digital anomalies including osteoporosis, polydactyly, syndactyly and others.
8. Hypertelorism and epicanthic folds. The cheeks are flattened due to a hypoplasia of the maxillae.

Less commonly found were benign tumours of the tongue, alopecia, dryskin, mucous pits of the lower lip, mental retardation and trembling.

The authors estimate that one case of oro-facial-digital syndrome occurs in every one hundred cases of cleft palate.
Kitamura and Kraus (1964) found that visceral abnormalities were more frequent and more severe in cleft palate foetuses. There seemed to be no apparent correlation between type and severity of cleft with type or severity of visceral malformation. This suggests that cleft lip and cleft palate are not just isolated malformations but are part of one or more syndromes affecting visceral and external features alike.
THE DEVELOPMENT OF THE TEETH

In the introduction to their book, Kraus and Jordan (1965) state:

"The embryonic period of man covers the first two months of prenatal life, but the embryology of the primary dentition begins at the end of this period and extends to about six months after birth. There is thus, no other organ in the human body which takes so long to attain its ultimate morphology."

Schour and Massler (1958) state:

"Instead of the usual one or two developmental processes in the formation of other organs, the tooth undergoes four or five such processes. The tooth not only must grow and calcify, it must also erupt before it can carry out its masticatory function."

The development of the tooth has been arbitrarily divided into five stages:

1. Initiation (bud stage).
2. Proliferation (cap stage).
3. Histodifferentiation and morphodifferentiation (bell stage).
4. Apposition and Calcification.
5. Eruption (prefunctional and functional).

Each stage merges imperceptibly into the subsequent stage.
It must be remembered that while the tooth is developing, concomitant maturative changes are going on in the face and jaws and in the supporting structures of the tooth. All the teeth along the dental arch are not developed at the same time. Hence, each tooth not only develops independently but is in a different stage of development. The stage of development of a given tooth will vary with its location. The steps involved in the development of all teeth are identical but the duration of each stage varies.

In previous discussion it has been mentioned how clefts of the primary palate develop in the fifth to sixth weeks of embryonic life and clefts of the secondary palate begin in the seventh to twelfth weeks of intrauterine existence. The first signs of activity leading to the development of teeth occurs when the embryo is six weeks old. It has further been shown how a cleft defect is not to be considered as a merely local abnormality but is part of a "first arch syndrome". The aetiological agents have been listed as (i) heredity and (ii) environment. There is probably a fine interplay between the two factors. It is quite feasible to assume that these very same factors acting on the developing teeth will produce the anomalies to be described later. The eventual results are probably aggravated by surgical
procedures in and around the developing tooth
germ.

Hereditary-environment interplay could affect
development of the primary and secondary dentitions
in utero, while neonatal nutritional problems and
later post-natal surgery could exert additional
influences perhaps more on the development of the
permanent dentition.

DEVELOPMENTAL STAGES. (Provenza 1964, p.159; p.111-139).

The earliest known feature which presages the
development of the teeth is the appearance of the
dental lamina. This may be observed in the human
embryo at the age of about six weeks. Subsequently,
groups of cells bud off from this lamina and become
specialized in various ways to form tooth germs. The
dental lamina and its lateral bud continue to grow
in size disproportionately so that the tooth
primordium outgrows its parent tissue.

The tooth anlage passes through various
developmental stages. These stages follow one another
but may overlap.

Localized increased mitotic activity of the
dental lamina results in the formation of bud-shaped
structures in which the cells are all morphologically
similar. This is the bud or initiation stage.
An increase in the size of this structure, together with disproportionate growth, produces a cap-shaped structure. The new form simulates the shape of the future crown. Meanwhile, cells, which were initially morphologically and cytologically similar, have changed their form and internal structure. By the end of this **cap or proliferative stage**, differentiation of the tooth germ has progressed to the point where the location of peripheral outer enamel epithelium; the basal invaginated epithelial layer (inner enamel epithelium) and the intervening layers, stellate reticulum and stratum intermedium can be recognized.

The stage of **histodifferentiation and morphodifferentiation** (bell stage) is characterized by the fact that complete cell differentiation is effected. Four definite layers are now to be seen. Total differentiation throughout the various layers does not occur simultaneously. The first cells to differentiate are those which are destined to form the incisal or cuspal areas. The formative cells arrange themselves along the future dentino-enamel junction so as to outline the size and shape of the future crown. The dentino-enamel junction acts as a blueprint for the shape of the tooth.
In the stage of apposition and calcification, the matrix for dentine and enamel is deposited in incremental fashion. This is the most sensitive phase of the development of the tooth (Schour and Massler 1940). The initial apposition of dentine and then enamel establishes the dentino-enamel junction. Dentinogenesis occurs in two stages (Takuma 1967 p.368). First an uncalcified collagenous matrix is formed, which in the second stage becomes calcified.

Provenza (p.123; 1964) states that differentiation of the cells of the inner enamel epithelium into ameloblasts does not begin until odontoblasts have matured and deposited the initial quantity of dentine. The first stage in amelogenesis is the deposition of a partially calcified matrix at the dentino-enamel junction. Crabb and Darling (p.93; 1962) summarize recent research findings about the subsequent stages of amelogenesis. The partly calcified enamel at the dentino-enamel junction becomes hypercalcified, as a foundation for the enamel rods. This zone of higher calcification spreads out towards the enamel surface, beginning just over the cusps of the teeth and spreading cervically. The process of calcification is not incremental in pattern. The pace of prenatal calcification is slow. Kraus (p.1128; 1959) has reported initial calcification in the deciduous dentition.
as early as twelve weeks in utero. He also found the sequence of initial calcification to be rigid and did not run in regular sequence from the central incisor to the second molar. The usual condition was for upper teeth to calcify before lowers except with lower cuspids. Garn et al (1958) state that calcification occurs earlier in girls than in boys. Garn, Lewis et al (1965) report the rate at which teeth calcify to be strongly affected by genetic and endocrine factors.

Both dentinogenesis and amelogenesis occur simultaneously, proceeding from the crest of the developing tooth to the root. (Provenza p.160;1964).

The teeth do not erupt until the crown is fully matured and root formation has begun. The time of emergence of all teeth varies widely. In the final stage of attrition, tooth surfaces are worn away by function and together with eruption, it is a continuous process throughout the life of the tooth. (Fanning,1961).

**PERMANENT TEETH.** (Provenza p.159; 1964).

The permanent tooth develops from the lateral band which attaches the developing deciduous tooth germ to the dental lamina. It therefore lies lingual to the developing deciduous tooth and on a level with the crest of the deciduous crown.
By the end of the seventh month of foetal life, the permanent tooth germ detaches itself from its predecessor. From the position described above, the permanent tooth germ is shifted to a location inferior to the apex of the primary tooth root. As the alveolar arch increases in size, the permanent germ becomes deeply embedded. Later, as the arch is widened, the permanent tooth germ undergoes translocation to a site lingual, mesial or labial to the apex of its successor. The incisors, cuspids and bicuspids originate in the manner described above but the permanent molars arise directly from the parent dental lamina.

RELATIONSHIP OF BONE AND TOOTH DEVELOPMENT (Provenza p.139; 1964).

In the earlier stages of development, the tooth germ appears to be independent of the adjacent tissue (bone and primitive periodontal tissue). But in later stages, a marked dependence is observed. Extensive growth takes place in that area of the jaws where the alveolar crest ultimately develops. The tooth germs maintain their relationship to the growing alveolar margin by moving occlusally and buccally.
ABERRATIONS IN THE DEVELOPMENT OF THE TEETH
(Schour and Massler, 1940)

A given disturbance, such as trauma to a developing tooth, will have entirely different effects, depending on the time of its occurrence and the stage of development which it affects. Aberrations in tooth development can therefore be classified according to the stage of development at which they occur.

If aberrations occur during the stage of initiation, single or multiple supernumerary teeth may result. A lack of initiation is evidenced as an absence of teeth. This may occur in isolated areas or there may be a complete lack of teeth.

Disturbances during the stage of proliferation may cause new parts to be differentiated, for example, supernumerary teeth, extra cusps or roots. Gemination or a complete suppression of parts may also occur. The result seems to depend on the severity of the disturbance rather than upon its specific character.

Disturbances to the stage of histodifferentiation and morphodifferentiation may disturb the form and size of the organ as well as the function of the formative cells. In the case of the peg lateral incisor the form and size of the tooth are affected without impairment to the function of the formative cells.
The form and size of the tooth can be affected only if a disturbance occurs at the bell stage of the enamel organ, when the morphogenetic pattern of the tooth is outlined.
### CLASSIFICATION OF ABNORMALITIES IN TOOTH DEVELOPMENT

*(Modified from Massler and Schour 1954)*

<table>
<thead>
<tr>
<th>Character of Disturbance</th>
<th>Initiation Proliferation</th>
<th>Histodifferentiation</th>
<th>Morphodifferentiation</th>
<th>Apposition</th>
<th>Calcification</th>
<th>Eruption</th>
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<tbody>
<tr>
<td><strong>Deficient Development</strong></td>
<td>Abnormal Lumen</td>
<td>Atypical Structure</td>
<td>Atypical Form and Size</td>
<td>Abnormal</td>
<td>Abnormal</td>
<td>Abnormal Eruption</td>
</tr>
<tr>
<td>Axodentin</td>
<td>Axogenisis</td>
<td>Pyg Teeth</td>
<td>Hypoplasia</td>
<td>Hypo-</td>
<td></td>
<td>Delayed Eruption</td>
</tr>
<tr>
<td>Partial or Complete</td>
<td>Imperfecta</td>
<td>Microdentia</td>
<td>Systemic -</td>
<td>Calkification</td>
<td></td>
<td>Impacted Teeth</td>
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<tr>
<td>Dentinogenesis</td>
<td>Imperfecta</td>
<td></td>
<td></td>
<td>or Local</td>
<td></td>
<td>Erupted Teeth</td>
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<td></td>
<td></td>
<td></td>
<td>Submerged Teeth</td>
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<tr>
<td><strong>Excessive Development</strong></td>
<td>Supernumerary Tooth</td>
<td>Extra cusps and Roots,</td>
<td></td>
<td>Odontoma</td>
<td></td>
<td>Haloconclusions</td>
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<td>Microdentia</td>
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<td>Dens in Dente</td>
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DENTAL ANOMALIES IN CLEFT AND CLEFT PALATE

INTRODUCTION.

The occurrence of dental anomalies associated with cleft conditions has been known for a long time. For good orthodontic prognosis, it is not sufficient to deal with just problems of dental caries and special types of malocclusion in cleft children. It is also necessary to know that there are problems of congenitally missing teeth, not just located adjacent to the cleft. In addition, there are abnormal forms of tooth crowns and roots, the results of abnormal development and perhaps surgical interferences at critical stages of development. Such conditions greatly increase the difficulties of orthodontic treatment in cleft children, let alone conservation of the teeth themselves.

This review will take the following form:-

2. Relationship of an alveolar cleft to the incisor teeth.
3. Abnormalities of tooth number.
4. Abnormalities of tooth form.
5. Abnormalities of tooth structure.
6. Cleft microforms.
GENERAL OBSERVATIONS.

NATAL AND NEONATAL TEETH.

The dental problems of cleft children may begin at birth. Dixon (p.178; 1966) states that it is not uncommon to observe exfoliation of a deciduous lateral incisor from the alveolar margin of a cleft. These teeth show normal structure on histological observation. They are said to be more common at the mesial margin of an alveolar cleft but may also occur at the distal margin. The tooth germ is thought to penetrate the surface epithelium, due to a local deficiency of tissue. The tooth germ is soon extruded and shed but if troublesome may be extracted. The deciduous incisor crypts are also very superficially placed and therefore subject to local and surgical trauma. This appearance tends to disappear slowly as a compensatory layer of bone covers the teeth but Graber (1964) warns that eighteen percent of all cleft cases show an absence of the anterior nasal spine.

Massler and Savara (1950) divide these into natal and neonatal teeth. The former are present in the oral cavity at birth, while the latter erupt during the neonatal period (birth to thirty days). The authors report the incidence of natal teeth as very low and neonatal teeth as of much lower frequency.
They are characterized by their looseness and lack of root formation. The teeth most affected are the deciduous lower central incisors. The aetiology of the condition is given as being due to:

1. the superficial position of the tooth germ,
2. heredity.

Massler and Savara issue a warning about the management of natal teeth. They should be left alone, if they are not causing any difficulty to the infant or to the mother. The authors state that indiscriminate extraction can lead to complications of trauma to the gingival tissues, which are strongly adherent to the cementum, and to excessive bleeding due to the physiologic hypoprothrombinemia, occurring at this transitional period.

Thoma (p. 37; 1960) considers them as being predeciduous and "white, rudimentary epithelial structures sitting on the gingiva." He states that their removal does not affect the formation of the deciduous teeth.

Godfrey (1967) has only seen the occurrence of neo-natal teeth in cleft infants in the lower anterior region. Their presence there affected the function of the pre-surgical orthopaedic appliance being used.
The position and number of teeth adjacent to a cleft has been a matter of dispute for a long time.

Goethe (1831), who discovered the premaxilla in man, suggested that a cleft would follow the premaxillary suture, thus separating the lateral incisor from the canine. He described this suture as running from the incisive foramen to the region between lateral incisor and cuspid, as shown in the figure below.

Diagrammatic illustration of theories available of the effect of an alveolar cleft on incisor development.
Albrecht (1879), after his studies of the comparative anatomy of the premaxilla, realized that the above was not an invariable finding in every case. He reported that the premaxilla comprised of two bones, the endognathion, supporting the central incisor and the mesognathion, supporting the lateral incisor, as seen in the previous figure. He suggested that a cleft could be situated on either side of the mesognathion, thus explaining the variable position of the lateral incisor. Difficulty arose in explaining an occasional tooth on the maxilla in the area between the cuspid and the alveolar cleft. Albrecht called this a "precanine".

Turner (1885) attempted to test Albrecht's theories by examining cleft palate casts. He said that these did not ",..., enable one to state with absolute precision the particular part of the jaw in which the alveolar fissure is situated, and still less do they permit one to determine if a maxillo-intermaxillary suture coexists with the alveolar cleft." The reviewer agrees with the above statement as he faced the same problem when examining dental casts of cleft children. In twelve of the fifteen casts examined, Turner saw Albrecht's precanine tooth. If the premaxilla (intermaxillary bone) supports the incisor teeth and if Goethe's theory of failure of intermaxillary-maxillary union is valid, then the lateral incisor
should always be found mesial to the cleft. (See previous figure).

In 1885, Coles followed the example of Albrecht and Turner, and examined thirty-one models of cleft patients. He reported that there was not a single instance of a cleft occurring between a true lateral incisor and a canine on either side of the mouth. He found insufficient evidence in any case of an increase in the number of teeth in the precanine region; on the other hand, there was a distinct evidence of the reverse condition and also of "imperfect development." In unilateral clefts the central incisor was missing in thirteen out of twenty-seven cases and in eleven out of thirty-one cases, a precanine was observed.

Keith (1909) also examined forty-one specimens to test the relationship of the cleft to the teeth. He found the cleft to be situated between the central and lateral incisor in twenty-one instances and between the lateral and canine in nine instances. In nine further cases there was absence of the lateral incisor and in two cases a supernumerary lateral incisor was present.

Inouye (1912) suggested that the cleft initiated supernumerary tooth formation by splitting the developing lateral incisor tooth germ. In direct contrast, Warnekros observed supernumerary teeth in
cleft palate patients and believed that this dental anomaly affected embryonic development in such a way that clefts of the lip and palate occurred.

Federspiel (1923) emphasized the frequency with which lateral incisors were absent in cleft patients. He presented evidence to strengthen Albrecht's theory that the premaxilla develops from four centres which fuse and later unite with the maxilla proper. Federspiel's interpretation of Albrecht's theory of development as it related to the lateral incisor is pertinent. "At each point where the tooth is going to be formed, a bell-shaped thickness appears at about the forty-eighth day or about the seventh week. It is interesting to know that about that time the union of the endo-, meso- and exognathion should occur. If the mesognathion is delayed in its formation, the exognathion and the endognathion may grow towards each other and prevent the full development of the mesognathion. The result is that the epithelial thickness to form a lateral tooth will be retarded, or arrested in its growth, or it may be obliterated."

Kirkham, writing in 1931, states. "... as the defect in the alveolus occurs at or near the lateral incisor, this tooth shows more embryologic irregularity than all the others."
He further says, "... there should be no special reason why dentition should be disturbed except in the lateral incisor area, unless we include .... supernumerary teeth, which ... more common in cleft cases ...." In unoperated cleft cases, Kirkham reports that, "there is invariably no more disturbance in dentition than in the normal month of similar age, except ... in the region of the lateral incisor." In operated cases however, he considered that the percentage of missing teeth, misplaced teeth and caries was greater than in unoperated cases.

Veau (1934) offers an opinion, based on observation of several hundred children on whom he operated and subsequently followed up. He found a variable situation for the lateral incisor in all his cases. In the primary dentition, a supernumerary tooth either mesial or distal to the fissure, was his most frequent finding. In the permanent dentition, a single fissural tooth distal to the cleft was more common.

The various hypotheses which are used to explain the variation in incisor positions relative to a cleft are shown in the previous figure. Supporters of each theory are still to be found. The concept of the dental lamina differentiating into tooth germs after the cleft is established is based
on the observations of Töndury (1961). However, Robinow (1958) suggested, after the examination of an embryo, that if two lateral incisors were present, one medial and one distal to the cleft, they were at one time one tooth, while another author thinks that supernumerary teeth may be caused by the extension of the dental lamina along the side of an alveolar cleft.

Early studies did not distinguish between the deciduous and permanent tooth adequately, but Milhon and Stafne (1941) radiographed eighty-one patients and suggested there was a tendency for deciduous supernumerary lateral incisors to be followed by permanent supernumerary lateral incisors. Böhn (1950, p.57) described the teeth of sixty-three children with clefts of the lip and palate. He found frequent absence of the permanent lateral incisors in patients with an alveolar cleft while patients with cleft lip showed supernumerary deciduous lateral incisors in three-quarters of the cases. In both dentitions, most of the laterals were found distal to the cleft. These were more frequently hypoplastic than laterals medial to the cleft.
Dixon (p. 84, 1966) presents the findings of a radiographic study of one hundred children with alveolar clefts. The photograph below shows his findings in graph form.

Findings on position of teeth in relationship to an alveolar cleft.

The most frequent position of the cleft, according to Dixon, was to separate the central and lateral incisors in both dentitions. The least common position was for the alveolar cleft to separate the lateral incisor from the canine. Complete absence of the lateral incisor was more common in the permanent (thirty-nine percent) than in the deciduous dentition.
The presence of supernumerary lateral incisors was more frequent in the deciduous dentition.

Holdsworth (p. 28, 1957) states that the germ of the lateral incisor may be damaged in the process of cleavage, so that this tooth is suppressed or poorly developed. It may erupt in the wrong place and direction and is sometimes double, "as if the tooth germ had been split."

Harvold (1954) mentions the fact that the position and direction of eruption of lateral and central incisors varies near the cleft. The development of the alveolar process is connected with the number, size and direction of eruption of teeth. Usually, all the teeth are present in the deciduous dentition. Occasionally the lateral incisor in the region of the cleft has an irregular form or perhaps a supernumerary tooth may be present. Often the crown of the lateral incisor erupts in the centre of the cleft. In the permanent dentition, absence of teeth is found in many cases, especially when the anterior part of the cleft palate is surgically closed within the first few months after birth. The eruption of the teeth is often normal but may be retarded especially in the margin of a cleft. In the deciduous dentition the incisors adjacent to a cleft may be slow in appearing and may erupt palatally
(Dixon p.179, 1966). In the permanent dentition the canine may be deeply placed and supernumerary incisors slow to erupt.
AETIOLOGY IN GENERAL

"Nature is nowhere accustomed more deeply to display her secret mysteries than in cases where she shows traces of her workings apart from the beaten path."

William Harvey, 1657.

Dental anomalies may be due to genetic, environmental or phylogenetic influences. Generally speaking, the deciduous teeth are much more constant and less frequently involved by gross abnormalities and alterations in number and size. The tendency for the permanent teeth to be more readily involved may be partially explained by their succedaneous position on the dental lamina of the primary teeth. (Tiecke et al, p. 443, 1959).

GENETIC INFLUENCES. (Sperber, 1967)

As with all biological structures, the final tooth morphology (the phenotype) is the product of its genetic directive (the genotype) modified by the environment in which it develops. Therefore, malformations may stem from either inherited defective genes or from mutation in a "normal" set of genes prior to them becoming operative.
When these defective genes operate in an optimum environment for tooth development, they produce abnormal forms of teeth. If they act in an adverse environment, the resultant tooth morphology can be severely aberrant. At the moment, the relative importance of defective genes and adverse environment in producing defective morphology is speculative. It is known that defective teeth can result from normal genes in an adverse environment. (Sperber, 1967, p.436).

The, as yet, undefined genetic complement concerned with dental development ultimately determines the presence or absence and morphology of teeth.

Sperber (1967, p.437) quotes Jacob and Monod (1961) as postulating that two gene types exist. "Structural" genes are responsible through ribonucleic acid (RNA) for the amino acid composition of proteins. "Non-structural" genes or. operator and regular genes control structural gene action. Chromosomal aberrations that produce malformations may result from atypical structural genes that initiate abnormal development or from failure or mis-timing of non-structural genes.

The different shapes of the teeth are due to the sites at which they develop. This forms the basis of Butler's "field theory". This theory will be dealt with in a later section. Butler (p.9, 1963)
suggests that "field" genes exist which influence the shape and size of the field in relation to individual tooth germs. He also postulated the existence of "pattern" genes influencing the way in which tooth germs react to any given level of the field. The pattern of the crown thus established reflects the dental evolutionary inheritance that is transmitted through the genes (Sperber p.440, 1967).

The total number of tooth germs is probably determined by genes causing the dental lamina to bud at specific sites (Sperber 1967, p.440). The complexity of cusp patterns is probably also genetically determined.

Gabriel (p.9, 1965) after examination of teeth from identical twins, suggests that the anatomical features of the various surfaces of the teeth are to a large extent genetically determined. He states that though two things are never exactly the same, their points of difference may be difficult to detect.

Genetic variation may be spread throughout the dentition (as in hereditary amelogenesis imperfecta) or may give rise to conditions affecting individual teeth.

A complex involving multiple genes, acting directly and indirectly, is responsible for the presence of supernumerary teeth (Sperber 1967, p.440). On the
evidence of supernumerary teeth being inheritable, their initiation is probably genetically determined.

Tooth size is an inherited characteristic.

(Garn et al 1965, p.228).

Enamel hypoplasia represents adverse environmental conditions affecting a presumably normal genetic make-up- (Sperber 1967, p.441).

This anomaly will also be discussed in a later section.

ENVIRONMENTAL INFLUENCES.

The environment influencing tooth development can be intra-uterine or post-natal.

The frequent occurrence of abnormal tooth development, not only in the offspring of mothers with abnormal pregnancies, but also in the children of mothers with seemingly normal pregnancies, as well as the reverse situation of normal tooth formation in complicated pregnancies; attests to the non-specific nature of congenital dental defects. This makes it difficult to establish a cause-effect relationship.

INTRA-UTERINE INFLUENCES.

Kreshover, writing in 1960, states that the majority of studies, linking a wide variety of metabolic and other disturbances to tooth abnormalities, are concerned with the postnatal period of dental development.
established whether such disturbances to the system during pregnancy can have similar effects on the developing dental tissues of the foetus in utero. "From studies it seems quite reasonable to expect that any disturbance in the highly specialized function of amelogenesis and dentinogenesis occurring during the prenatal period might be indicative of a more widespread foetal response to the particular injurious agent occurring at the time." (Kreshover, p.574, 1960). The teeth might also contribute much needed information on the subject of the placenta as a barrier to or a pathway for transfer of various injurious agents from mother to foetus.

With the thalidomide disasters still fairly fresh in the mind, the study of Axrup and his co-workers (1966) into the dental conditions of these children is interesting. They found the following disturbances in tooth development:

1. partial anodontia: in every case the missing teeth were primary lateral incisors or the germ of the permanent lateral incisors.

2. anomalies in the shape of the crowns.

3. disturbances in mineralization: the enamel hypoplasia showed a wide range of variation in both appearance and localization.
Maternal infection with micro-organisms and viruses, rhesus factor incompatibility (Wienor 1946), dietary deficiencies of vitamins (Giroud 1954) and mineral salts, irradiation of the maternal pelvis during pregnancy and maternal administration of cortisone (Fraser et al 1953) and of toxic substances such as trypan blue, have all been shown to produce more or less specific disturbances of development in the offspring (Warkany, 1947; Kalter and Warkany, 1959). Much of the evidence, but not all, relates to laboratory animals and is only indirectly applicable to the problem of malformations in Man and even more indirectly still to the problem of the causation of dental malformations. It is interesting to note that not all strains of laboratory animals are equally susceptible to environmentally induced developmental abnormalities (Fraser et al 1953) so that it is possible that at least some of the examples of aberrant development result from a combination of both genetic and non-genetic factors.

It is well known that infection of the foetus of a syphilitic mother produces characteristic malformations of the teeth. The foetus is not infected until a relatively late stage of pregnancy, as is exemplified by the fact that the deciduous teeth are never affected, because in earlier stages
the placental barrier is effective against the passage of the Treponema pallidum (Sarnat and Shaw 1943, p.279). The same authors found that thirty percent of the seventy-three patients they studied, showed dental changes characteristic of the disease. These were hypoplastic changes in the incisors and molars to produce the commonly called Hutchinson's incisors and mulberry molars (Brauer et al 1959, p.52).

Nagai et al (1963) observed the following changes in transplanted tooth germs influenced by polyoma virus:

1. loss of tooth morphology
2. degeneration of ameloblasts with loss of enamel formation.
3. modified dentine formation.

In laboratory animals, maternal vitamin deficiencies too slight to produce signs of avitaminosis in the parent animals, have been shown to cause many types of malformation in the offspring (Giroud 1954). Warkany (1954) noted the absence of mandibular incisors in some instances. Knudsen (1966) has shown that hypervitaminosis A (lathyrisn) also produces oral malformations. These include fusion of upper incisors and agenesis or abnormal molar germs.
It is now well established that attacks of rubella (German measles) during the first trimester of pregnancy can cause congenital defects in the infant. (Evans 1944, p.225). Cataract, deaf-mutism and cardiac anomalies are the most common abnormalities. Interest in maternal rubella from the dental point of view arises from the fact that there is now an extensive literature on the subject with ill-defined references to delayed eruption, malformed teeth and absence of teeth (Miles 1954, p.825). Gregg and his colleagues have also noted retarded eruption in these cases. Evans, an Adelaide dentist, carried out the only clinical studies that throw any light on the exact nature of the dental anomalies (Miles 1954). Evans (1944, pp.225 and 227) carried out his studies in two stages. He first examined thirty-four affected children for:

1. variation in the number of teeth from normal.
2. hypoplasia
3. abnormal tooth form.

Twenty-three of these children showed evidence of one or more dental defects. Sixteen children exhibited retarded eruption of teeth. Hypoplasia was found in eight cases, mainly of a minor degree of severity.
The surprising fact is the incidence of hypoplasia in the deciduous dentition. Normally the incidence is approximately 0.5 percent but here it was 23.5 percent (Evans 1944, p.227). Abnormal tooth form was present in seven cases with the incisal third affected in every case. Evans (1947, pp.780-784) made a second study of sixty-seven children, including the thirty-four from the first study. Radiographs were not used routinely except to confirm a finding. Three children had congenital absence of incisors, comprising of two lower lateral incisors and two upper lateral incisors. Thirteen children showed hypoplasia which was localized to two to four teeth. Eight cases exhibited abnormality of tooth form but this was not very pronounced. Premature eruption of permanent teeth was present in four cases.

Miles (1954, p.825) quotes Stocker who examined serial sections of the jaws of a one month premature infant, whose mother had rubella at the end of the sixth week of her pregnancy. There was complete absence of all the permanent tooth germs and marked degenerative changes in the enamel organs of the deciduous teeth, of a kind which would no doubt have resulted in hypoplastic defects of the teeth later.
Dental anomalies are common in mental deficiency and in mongolism. Spitzer (1963) describes the teeth in mental defectives as being stunted with their crowns cone-shaped in the anterior and occlusally tapering in the posterior regions. Generalized microdontia and often partial anodontia occur bilaterally and may involve all four quadrants. The eruption times of the permanent teeth may be retarded.

McMillan and Kashgarian (1961) list the dental anomalies found in mongolism as:

1. hypoplasia
2. congenitally absent teeth
3. supernumerary teeth
4. abnormal morphology

The most frequent defects in a total of seven hundred and sixty-seven subjects were:

1. congenital absence of teeth (thirty-five percent)
2. teeth with abnormal morphology (thirty-four percent)

These defects are representative of faulty development in early embryonic life (McMillan and Kashgarian 1961). They were confined to the maxillary and mandibular deciduous and permanent incisors. The authors suggest two aetiological possibilities to explain the above observations. First, "the insult or deficiency may be operative over a sufficiently long period to
include the period of initiation of the deciduous
tooth bud and its permanent successor." Second,
"the anlage of the deciduous tooth bud may contain
the potential for growth for both the deciduous
tooth and its permanent successor."

A further study of one hundred and twenty
mongoloid patients by Otero and Sznajder (1967)
reports that thirty-three percent of these patients
had anodontia of one or more permanent teeth. The
teeth most frequently missing were the lateral
incisors. Among the forty patients with anodontia,
the majority exhibited it bilaterally. The prevalence
was slightly greater in the maxilla.

Brown and Cunningham (1961) made a survey of
eighty mongoloid patients. Their main finding was
the congenital absence of teeth. Thirty-three
percent had both permanent upper lateral incisors
missing. Others had the same tooth missing on one
side only.

Via and Churchill (1957) recorded an incidence
of enamel hypoplasia among fifty children with
cerebral disorders, whose births had been marked
by poor respiration or prematurity. Hypoplasia was
also seen in seven of the sixteen children born to
mothers with antepartum haemorrhage. Stein (1947)
describes the hypoplasia seen in the teeth of
prematurely born children as "grooves and pits with depressions of the enamel surface."

Stack (1963, p.445) says that pathological events affecting the foetus may produce an apparently non-specific growth response from the dentition. Via and Churchill (1959) defined a definite correlation between the timing of abnormal events of gestation and birth to enamel hypoplasia.

Miles (1954, p.825) cites Spitzer and Mann as attributing a common cause for the association between mental deficiency, lens defects and dental anomalies: All the affected tissues are of ectodermal origin and the syndrome could be explained on the basis of a defect of that layer. Since the maternal and family histories of the cases studied were unknown in most cases, the above explanation must remain a subject for speculation. It is interesting to note that Spitzer and Mann postulate the presence of enamel hypoplasia in the permanent dentitions of these children as being probably due to post-natal nutritional deficiencies associated with difficulties of feeding of mentally deficient debilitated infants.
POST NATAL INFLUENCES.

These can be divided into systemic and local influences.

SYSTEMIC INFLUENCES.

Sarnat and Schour (1941, p.1989) introduce the concept of the growing tooth being a biologic recorder of both health and disease. They state that the developing enamel and dentine "yield accurate prompt and permanent records of both normal fluctuations and pathologic accentuations of mineral and general metabolism." On the basis of the severity and duration of the disturbances, the records may be microscopic or macroscopic. In enamel, the history of systemic disturbances is indelibly recorded by a cessation of ameloblastic activity. A lack of enamel formation is noted in the particular portion of the tooth that is developing during the disease period.

Stein (1947) says that metabolic disturbances even of relatively short duration exert a greater influence on the developing tissues in infancy than they do in later life.

Sicher (1967, p.88) points out that systemic influences are in the majority of cases active during
the first year of life. Therefore, the teeth most frequently affected are the permanent incisors, canines and first molars. The upper lateral incisor is sometimes found unaffected because its development starts later than that of the other teeth mentioned.

Some of the systemic influences which are involved are the exanthematosus diseases of infancy, the change from the relatively stable intrauterine existence to the rigors of extrauterine life, including endocrine disorders and vitamin deficiency states. Downs (1928, p.372) writing in the early part of this century, states that "while the endocrines do play an important part in calcium metabolism, they do not act in such a way as to cause regular types of dental anomalies." Schour and Massler (1943, p.948) state that there is no evidence that alterations in endocrine function can give rise to oral or dental symptoms without producing other systemic symptoms. Sicher (1967,p.89) states that recent investigations have demonstrated that exanthematosus diseases are not so frequently a cause of enamel hypoplasia as was previously believed.

**LOCALIZED INFLUENCES.**

These may affect single teeth in most instances. The cause may be infection of the deciduous pulp with subsequent infection of periapical tissues and
irritation of enamel formation of its permanent successor (Sicher 1967, p.39).

The localized influence of most concern to us is trauma to the developing tooth. This can originate from the surgical procedures employed in the repair of cleft lip and palate. It is suggested that with modern surgical techniques, trauma may not involve the tooth germs in their crypts directly but the blood circulation to the developing tooth germs may be altered in such a way as to cause some of the anomalies we see later (Godfrey, 1967).

In order to clarify later descriptions, a short description of the circulation to developing tooth germs is presented. Two vascular plexuses lie within the dental papilla and about the dental sac of the developing tooth (Echeverria, 1963; Saunders and Rockert, 1967, p.225). One plexus surrounds the dental sac or capsule-like condensation of embryonic tissue which invests the developing tooth. This was called the peridental plexus by Saunders. It is closely related to the outermost layer of the enamel organ. The second plexus lies within the dental papilla below the developing crown and is known as the intradental plexus (Saunders and Rockert 1967, p.225).
Both plexuses are derived from branches of the alveolar arteries, which are the main arterial supply trunks of the maxilla and mandible. Saunders and Rockert (1967, p.227) report the interesting observation that the intradental plexus precedes the appearance of enamel and mineralization of the crown. The authors regard this plexus as a nutritional source from which the odontoblasts draw their raw materials while secreting their final product (Saunders and Rockert 1967, p.230). The peridental plexus, surrounding the dental sac, has been shown to be continuous with the intradental plexus (Saunders and Rockert, p.231). Prenatal infections (for example, syphilis) must gain entry to the tooth germ via these plexuses (Saunders and Rockert 1967, p.241). Histological sections have shown syphilitic exudate in the region of the peridental plexus and spirochaetes within the tooth buds. The staining of children's teeth by some of the antibiotics (for example, tetracycline) and induction of dental anomalies thereby must be dependent on vascular transport via the intradental plexus during pregnancy. (Saunders and Rockert 1967, p.241).
Before considering trauma to the developing teeth in children with cleft lip and palate, the following discussion will deal with the effects of trauma on tooth germs.

Incompletely formed teeth, lying in their crypts, may be injured by trauma.

Rushton and Cooke (1963, p. 69) see the most common effect as, "the part of the tooth already formed at the time of injury is dislocated without serious damage to its vascular supply. The root then continues to form in the original direction so that on completion the tooth presents an abruptly angulated form. These "dilacerated" teeth show no histological abnormalities except alteration in alignment of tissues." Colyer and Sprawson (1942, p. 290) state that the above abnormality occurs more commonly in the permanent dentition. Thoma and Goldman (1960, p. 97) comment on the possibility of the deformity of the root occurring "at any place from the neck to the apex." When the causative trauma is of a "gentler" nature, the deformity produced may be a bend in the direction of the root. (Colyer and Sprawson, 1942, p. 292). The injury may occur either to the crown or to the root according to the development of the tooth at the time of injury.
If the injury is serious enough to damage the vascular supply to the developing tooth, then this temporary interruption in circulation is reflected in "qualitative changes in the dental hard tissues." (Rushton and Cooke 1963, p.69). The circulation is later restored by the formation of new vessels and the apposition of dentine is usually resumed. In the case of premolars, the calcified portion occasionally appears to be forced into the developing part causing a kind of "impacted dislocation" (Colyer and Sprawson 1942, p.291). The authors state that this can sometimes occur from careless removal of a deciduous tooth.

The shape of the crown and the arrangement of the tissues may be altered by very early injuries to the tooth germ (Rushton and Cooke, 1963, p.71). These authors describe two other effects of this early injury: (i) hypoplastic defects of the enamel, (ii) formation of additional cusp or denticle joined to the same root.

Williamson (1966, p.284) gives case histories to show that trauma during exodontia can be an aetiologic factor in producing hypoplastic premolars.

Glasstone (1952, p.12) has shown by experiments on rabbits that the very early tooth germ has remarkable powers of recovering from mechanical injury.
Tooth germ were halved before the appearance of cusps and the halves were cultivated separately. The halved tooth germs formed complete tooth rudiments. This did not happen once morphodifferentiation had proceeded further. Rushton and Cooke (1963, p.71) feel that in man "most injuries appear to occur at a stage after this 'regulative power' is lost."

Butcher and Taylor (1951, p.274) report on experiments on the effects of denervation and ischaemia on the teeth of monkeys. They state that development of teeth and maintenance of tooth structure do not depend on innervation. Vascular disturbances at the tooth apex could permanently injure a tooth, though the authors state that a collateral circulation is quickly established.

Little correlation can be made between the type of surgery and dental abnormalities in cleft lip and palate because of the broad range of insults and the diversity of modern surgical techniques. At one extreme are the techniques used by Brophy in the early part of this century. Jolleyes (1954, p.229) cites Brophy as believing that the maxillae tended to separate as growth continued. He, therefore, used wires to pull the two halves of the upper jaw together. As early as 1933 (p.413) we see Logan and Kronfield warning about the dangers to developing tooth germs from such procedures.
They state that there is not enough space present between the deciduous tooth germs to pass a wire from the outside of the maxilla between the germs nor would it be possible to avoid injury to the permanent germs lying on the lingual side of the deciduous germs. The authors think that a common injury from this procedure is "major displacement or destruction of the germ of the permanent lateral incisor." Thoma and Goldman (1960, p.96) also warn about the Brophy method in causing injury to tooth germs, the canine and lateral incisor, in particular. Jolley's (1954, p.240) confirms the findings of Graber and Slaughter and Brodie that there is a reduction in maxillary development due to surgical operations. He felt that this was due to fibrosis rather than ischaemia, as suggested by Graber.

DISCUSSION.

The reviewer feels that even though children with cleft lip and palate have dental problems before any surgical intervention whatever, these could only be accentuated by the lip repair and perpetuated by the palate repair. Operative injury today is more common in the region of the alveolar cleft and could be associated with anomalous development of the central and lateral incisors adjacent to the cleft.
Figures I to V are examples of clinical conditions seen at the cleft palate clinic at Royal Alexandra Hospital for Children. Surgery can often result in shifting in position of a tooth or teeth buds. Figure V is the plaster model of a patient with a cleft of the secondary palate. The gross shifting of teeth evident is most probably due to the result of surgery. Fibrosis, mentioned above, can cause this effect. Disturbances to the circulation of developing teeth by fibrosis and actual surgical trauma is also suggested as an important cause of anomalous development. The periosteum is one of the vascular sources of the developing tooth (Saunders 1967, p.206). Any injury to this layer can affect a source of blood supply to the tooth. A corresponding situation exists in a tooth socket where the capillary plexus of the periodontal ligament remaining on the pocket wall after extraction of the tooth is important in facilitating vascularization in the socket by giving out new blood vessels from it. (Noma 1967, p.40).

As Harvold stated in 1954 (p.506), the major part of the deformities seen in cleft lip and palate cases cannot be due to reduced growth potential. Alteration in environment will modify growth potential.
Further improvement in surgical and orthodontic treatment will come with extension of our knowledge as to the extent of the different anomalies in the facial skeleton (Harvold 1954, p.495). Perhaps this lies in the direction in which Silverman (1965, p.213) has suggested. This author describes a family of craniofacial malformations with certain common features among them. He mentions cleft lip and palate (p.215) as one of the less severe forms of this group.
Unilateral cleft of primary and secondary palates showing rotated and anomalous upper central, absence of lateral incisor, canine and both bicuspids on cleft side.

Bilateral cleft of primary palate showing absence of one central incisor and both lateral incisors. The central incisor present is anomalous.
Figure III

Unilateral cleft showing grossly hypoplastic central incisors. A "dens in dente" is also seen in the canine region.

Figure IV

Unilateral cleft showing grossly rotated central incisor with congenital absence of lateral incisor, canine and first and second bicuspid.
Figure V

Cleft of secondary palate, showing gross displacement and shifting of teeth. This is suggested as being most probably due to the effects of surgery on the palate.
PHYLOGENETIC INFLUENCES. (BUTLER'S FIELD CONCEPT)

(Gaunt and Miles, 1967; Boyle 1955, p.13)

The dentition of man is changing in form, size and number. There is a trend towards simplification of the patterns and a reduction in measurements of the teeth (Dahlberg 1945, p.676).

Butler in 1939 adapted the concept that a field of influence governing size and form existed in each quadrant or tooth group. Each unit of the dentition develops in this "morphogenetic field" (Gaunt and Miles 1967, p.153) according to its position in the field. The morphogenetic capacity is not the same at both ends of the field. The field is differentiated into regions corresponding to the incisor, canine and molar regions and these regions have some degree of independence. The field manifested itself in decreasing strength as the distance was increased from a hypothetical pole located within the field. Butler observed that morphologic variability became greater in the more distal tooth of each group.

Dahlberg in 1945 amended this concept by separating the premolars from the molar group in the human dentition. There are thus four "types" of fields to consider. In each group one particular tooth is remarkably constant in form.
This tooth is practically never absent except when other teeth in the group are also absent. Since it is almost invariably present and normal in pattern, it has been designated the stable tooth in contrast to the other more variable teeth in the group. The upper central incisor, the lower lateral incisor, the cuspids, the first bicuspids and the first molars are the stable teeth. The others are more subject to variation in size and form and are more likely to be congenitally absent. In other words, the relative variability in size and form was shown to be smallest for the more anterior or mesial tooth in each group with the exception of the mandibular central incisor, which showed a larger variation.

The maxillary lateral incisor is the most variable tooth and the maxillary first molar the least.
CONGENITAL ABSENCE OF TEETH (ANODONTIA) (MISSING TEETH)

Boucher (1963) defines anodontia as the congenital absence of teeth: it may be partial or total and may affect the permanent dentition or both the deciduous and permanent dentitions.

Thoma and Goldman (1960, p.36) warn that true anodontia may be difficult to recognize sometimes as tooth may have been lost years ago by extraction, by accidental removal of a tooth germ with a deciduous tooth or by exfoliation. Groups of teeth or individual teeth may be affected.

DIAGNOSIS.

Radiographs are suggested as the most satisfactory method by Dolder (1937, p.142).

Werther and Rothenberg (1939, p.61) also suggest a complete radiographic examination to determine true anodontia. They also suggest a medical and family history because "they very often throw light on the aetiology of the deficiency." The condition of the alveolar ridge is usually smaller where true anodontia is present. (Werther and Rothenberg 1939, p.61).

A true anodontia can only exist in cases where neither retained tooth germs nor their developed forms can be identified by radiographs. It also exists where
the age of the child accounts for the presence of the necessary number of teeth and where an early extraction of the missing teeth does not agree with the age of the child.

AETIOLOGY.

The cause of congenitally missing teeth has been attributed to several factors:

1. heredity
2. local influences in the direct region of the jaw
3. systemic diseases
4. endocrine disorders
5. phylogenesis.

All these influences have already been considered earlier. Whatever the cause, the absence of a tooth is due to failure of the dental lamina to develop or the dental lamina may become exhausted after the deciduous tooth is formed and not be able to produce a permanent one. "There is a strong influence to pass such acquired characters on to future generations, the defect then becoming a simple Mendelian dominant". (Thoma and Goldman 1960, p.34).

In the past, missing lateral incisors were held to be the result of hereditary syphilis and the original defect was thought to be handed down to numerous generations as a dominant character. (Thoma and Goldman 1960, p.31).
But Stern (1960, p.483) states that any changes made by individual adjustment will not affect the future generations.

**CLINICAL FINDINGS.**

Congenital absence of teeth is unusual in the deciduous dentition but quite common in the permanent dentition (Grahmén 1962, p.198). Several investigations of the frequency have been published but with different results. Grahmén (1962) states that it varies between three and six per cent. The variations have been greatest in regard to the third molars. No statistically significant differences have been found between the sexes by Grahmén (1962) and Bolder (1937, p.143) though Lind (1959, p.187) and Kline (1967) have found a higher frequency in girls. Meskin and Gorlin (1963, p.1479) demonstrated a higher frequency for agenesis of the permanent lateral incisors in girls. (It is interesting to note that supernumerary teeth seem to be more common in men (Grahmén 1962, p.198)).

According to a study by the same author, the mandibular second premolars showed the highest frequency of agenesis (about 2.3 percent), followed by the maxillary lateral incisors with 1.6 percent and the maxillary second premolars with 1.4 percent. The frequency of congenital absence of one or more third molars was
25 percent. Garn et al (1963, p.1358) found that agenesis of the third molar was associated with a marked increase in the number of missing teeth of other morphologic classes and delayed calcification of the posterior teeth. Lind (1959, p.188) makes the observation that numerical variations of teeth have a strong tendency to appear symmetrically. When asymmetrical hypodontia is seen in the anterior region, it appears to be more common on the right hand side. Dolder (1937, p.143) states that he found no difference between right and left sides in his study of deficient dentition. He makes the further comment that absence of teeth was observed more frequently in the lower than in the upper jaw whereas supernumerary teeth occurred more often in the upper jaw than in the lower jaw. He also noted that girls tended to have agenesis of the upper lateral incisor more commonly and boys to have missing upper second premolars. Meskin and Gorlin (1963, p.1479) in their study of absence of upper lateral incisors found that if agenesis of this tooth was unilateral, it was twice as common on the left hand side as on the right hand side. Clayton (1956, p.206) found that in cases where more than one tooth was absent, there was usually a bilateral occurrence.
Oliver et al (1945, p. 220) after a study of congenitally missing second premolars state that "lack of space in the arches is not a factor in the congenital absence of the second premolars". In a study of the genetic pattern of congenitally missing teeth by Grahnén (1962, p. 199) he came to the conclusion that "congenitally missing teeth are primarily genetically determined". He thought that congenitally missing teeth may represent one group of anomalies in a genetic syndrome but if so, is manifested as the only alteration in most individuals. He cites Grüneberg whose animal studies indicate that congenital absence of a tooth occurred when the tooth size reached a certain lower limit. The same may also be the case in humans. Therefore, in cases of congenitally missing teeth, one should find intermediate forms of smaller teeth and variations in tooth form. Peg-shaped teeth in the upper lateral incisor region often occurred in association with congenitally missing teeth (Grahnén 1962, p. 201).

Grahnén (1962, p. 202) states that his studies indicate a correlation between the deciduous and permanent dentitions. Individuals with hypodontia in the deciduous dentition also have hypodontia in the permanent dentition in 75 percent of cases.
Böhn has noted that missing teeth in the area of the cleft occur in both deciduous and permanent dentitions, but it is much more common in the latter. (Böhn, 1963, p.100). His findings can be tabulated as follows:—

1. There was no distinct difference between right and left sides in the occurrence of missing teeth.

2. Boys with missing teeth lacked significantly more teeth than girls with similar condition.

3. The number of missing teeth in the posterior region was higher in bilateral than in unilateral cases.

4. More teeth were missing in the upper than in the lower jaw.

5. In unilateral clefts, the number of missing teeth in the posterior region was higher on the cleft side than on the non-cleft side.

6. The most frequently missing teeth were: the upper second premolar, the lower second premolar and the upper lateral incisor.

7. Missing teeth were more frequent in clefts of the primary and secondary palates than in clefts of the primary palate alone.
8. In patients with clefts of the primary and secondary palates, the majority had either one or two teeth missing (maximum six teeth missing).

9. Patients with isolated clefts of the secondary palate also had significantly higher incidence of missing teeth than normal cases. The majority of missing teeth were lower second premolars, upper lateral incisors and upper second premolars.

Olin (1964) made a study of missing bicuspids in cleft patients and arrived at the following results:

1. Twenty-four percent of 175 patients examined had missing bicuspids. The most frequently missing was maxillary second bicuspid.

2. A total of eighty-two missing bicuspids were noted, sixty in the upper arch and twenty-two in the lower arch. Some patients had upper and lower bicuspids missing.

3. Missing bicuspids are more frequent in patients with clefts of the primary and secondary palates than in other types of clefts.

4. Incidence of missing bicuspids has been reported to be 6.6 percent in the general population while this study showed 24 percent of cleft lip and palate patients had missing bicuspids.
Harvold (1954, p. 494) states: “In the permanent dentition absence of teeth is found in many cases (of cleft lip and palate), especially when anterior part of cleft in the palate is closed within the first few months after birth.”

Kirkham (1931, p. 1076) points out that as the defect in the alveolus occurs at or near the lateral incisor, “it is obvious that this tooth shows more embryologic irregularity than all the others.” He mentions absence of the lateral incisor on the cleft side as one of these irregularities. He thinks that after plastic surgery, “a larger percentage of missing teeth, misplaced teeth is found than in those who have no operation.”

Dixon (1966, p. 84) reports on the tendency for absence of the permanent lateral incisor in relationship to a complete alveolar cleft as well as in incomplete alveolar clefts. Absence of lateral incisors was also noted in clefts of secondary palate only. His figure for absence of bicuspids is 26 percent.
CLEFT MICROFORMS AS REVEALED BY THE TEETH

Studies by Böhn (1963), Dixon (1966, Kraus, Jordan and Neptune (1966) have shown that the dental anomalies seen in patients with clefts fall into a particular pattern. Very similar dental anomalies are seen in the absence of clefts, being generally regarded as a minor form of the same disorder, known as "dental microforms".

The most frequent anomalies in patients with clefts are absent teeth and supernumerary teeth.

Böhn (1963, p.88) observed the whole range of dental anomalies on the unaffected side in unilateral clefts and showed that the size of the lateral incisor on the non-cleft side is to some extent dependant on the dental anomaly on the cleft side. If the affected lateral is absent, the corresponding tooth on the normal side is absent or small. Similarly, if supernumerary teeth are present on the cleft side, the lateral on the normal side is larger than average.

Kraus et al (1966, p.51) in a study of foetal material showed that the dental malformations seen in cleft material are occasionally seen in foetuses without clefts, although the number and severity in these cases are much reduced.
Several attempts have been made in the literature to show that dental anomalies are evidence of genetic transmission of cleft palate. Dixon (1966, p.185) cites Lucas (1888) who reported an increased incidence of absent lateral incisors in the families of cleft palate patients. On the other hand, a recent study of Woolf et al (1965, p.547) showed statistically that the incidence of lateral incisor anomalies was not greater in families with a history of cleft defects than in a normal population. In Dixon's study (1966, p.185) the incidence of lateral incisor absence in isolated cleft palate and on the contralateral side in unilateral clefts, suggested that "partial anodontia is a genetically associated anomaly, and not a microform cleft tendency". As Dixon states further, "the presence of occasional instances of supernumerary teeth, geminated teeth and abnormalities of tooth form, such as tuberculated teeth, on the contralateral side to a unilateral cleft have yet to be explained".

**SUPERNUMERARY TEETH (HYPERDONTIA).**

Levine (1962:p.297) defines a supernumerary tooth as an extra tooth which does not resemble any normal tooth from a morphological standpoint.
He distinguishes the above from a "supplemental tooth" which he describes as an extra tooth which does resemble a normal tooth in shape and size.

Supernumerary teeth can occur anywhere in the dental arch; however, there are certain places where the majority of them are found.

PATHOGENESIS.

As Gardiner (1961, p.65) states, various theories have been put forward to explain the origin of these extra teeth.

1. Atavism - recurrence of ancestral forms of teeth which have become extinct.
   This view is not strongly held now (Gardiner 1961).

2. Excessive growth of the dental lamina.
   This is the most favoured theory and was put forward by G.V. Black in 1909.

3. Proliferation of remnants of the dental lamina.

4. Dichotomy of tooth germ. If the division is equal, the result is a supplemental tooth but if unequal, the additional tooth may be malformed and conical.

5. Heredity.

6. General conditions, for example, cleft palate.

Saarenmaa (1950, p.298) states, "the most probable explanation appears to be that supernumerary teeth
in all parts of the arch are the result of proliferation of the dental lamina from independent dental elements, seeing that they are often of normal size as compared with adjoining teeth". He states further that supernumerary teeth are not inherited according to any definite formula.

**CLINICAL FINDINGS.**

The most common site of supernumerary teeth is the upper central incisor region, followed by the upper third molar and lower bicusp region.


Supernumerary teeth can take a variety of shapes and sizes. Often they assume the form of the normal teeth in the region in which they erupt; more often, they exhibit imperfect forms with single or multiple cusps, but usually single rooted.

(Flint, 1939, p.135). Worth (1963, p.101) mentions two common shapes of supernumerary teeth:—that of a cone or that of a poorly formed bicuspid, with the former far more numerous. The common site is behind the upper incisors or between the two or, less commonly, in place of the central incisors, which is either displaced from normal position or prevented from erupting. Since the dental lamina produces the permanent teeth on the palatal aspect of the deciduous predecessors, it might be anticipated that supernumerary teeth may develop palatally.
A single supernumerary is common, but two are also frequently noted, one behind each central incisor.

Gardiner (1961, p.64) gives an incidence figure of 4½ percent for supernumerary teeth in a normal population. Saarenmaa (1950, p.296) quotes an incidence of 0.64 percent in young adults and 0.28 percent in schoolchildren. He noted a difference between the sexes; boys had over three times as many supernumeraries as girls. He did not note any side preference and stated the incidence as "practically equal" in upper and lower arches. Millhon and Stafne (1941, p.604) conclude, after a study of eighty-one cases that supernumerary teeth occur more often with cleft palate than with harelip. The cleft occurred more frequently between the lateral incisor and the cuspid. If both a lateral incisor and a supernumerary tooth were present, the cleft occurred between them with the supernumerary tooth distal to the cleft. Millhon and Stafne suggest further that when the cleft separates the lateral incisor and supernumerary, the latter is the result of division of the tooth germ by the cleft.

Bohn (1963, p.65) makes some general observations on supernumerary teeth in patients with clefts. If the cleft is less severe, the supernumerary teeth
are longer and more of an incisor form. The incisor form of supernumerary teeth in the lateral incisor area in normal individuals has also been noted by Gardiner (1961, p.64), frequently preceded by deciduous supernumerary teeth, indicating that these are derived from odontogenic epithelium with specific morphogenetic properties. (Johnson 1967, p.442).

The view has been put forward (Keith 1933; cited by Millhon and Stafne 1941, p.599) that a normal lateral incisor is originated from two growth centres, one derived from the frontonasal process and the other from the maxillary process. A transient delay in the dissolution of the epithelial wall will give an explanation of how these supernumerary teeth originate.

The presence of severe displacements and rotations in association with supernumerary teeth has been noted. It is, however, possible that these irregularities may be a manifestation of a more general disturbance and not the direct effect of the presence of a supernumerary tooth. (Johnson, 1967, p.442).
FISSURAL TEETH

(Bohn, 1963, p.12, p.99, p.101 and p.103)

Bohn (1963, p.98) describes fissural teeth as "the dental formations that occur instead of the upper lateral incisor in the cleft area."

PATHOGENESIS.

The fissure in the alveolar process runs lateral to the upper central incisor, mesial to the canine and through the normal site of the lateral incisor. This always causes abnormal dental conditions and it has been found expedient to call them mesial fissural tooth and distal fissural tooth. Previously both teeth have been regarded and designated as equal to the lateral incisor; or one has been regarded as the representative of the normal lateral incisor and the other as a supernumerary tooth. (See figure on page 53).

INCIDENCE.

A mesial fissural tooth developed in 47 percent of the clefts in the deciduous dentition and in 25.6 percent in the permanent dentition. A distal fissural tooth developed in 75.6 percent of the cases in the deciduous dentition and in 44.3 percent in the permanent dentition. Both fissural teeth were present in 36.9 percent in the deciduous and in 14.3 percent in the permanent dentition.
The number of fissural teeth usually decreases with increasing size of cleft, up to complete bilateral clefts, when there is another rise. Sex and side of cleft did not significantly influence the fissural teeth.

**Morphology.**

The mesial fissural tooth has incisor shape as its basic pattern. In the permanent dentition, this tooth often shows anomalies in the form of hypoplasia, malformation and reduction in size.

The distal fissural tooth shows canine shape nearly as often as incisor shape in the deciduous dentition. In the permanent dentition, canine-shape predominates but usually in markedly reduced sizes that degenerate into peg shape. Malformation and hypoplasia are also seen but not as frequently as in the mesial fissural tooth.

Measurements show that the fissural teeth of the secondary dentition are on the average smaller than the corresponding lateral incisor on the non-cleft side. The distal fissural tooth is generally smaller than the mesial. The size of the fissural teeth decreases with increase in size of cleft.

When a mesial fissural tooth is present, the size of the permanent central incisor is reduced.

When a distal fissural tooth is present, the
permanent canine is usually larger than when the fissural tooth is missing.

PEG TEETH. (Jordan et al 1966, p.32)

Normally the maxillary lateral incisors present three distinct lobes on their incisal edge. The middle or central lobe, the most conspicuous in terms of incisal prominence, is flanked respectively by a less prominent mesial lobe and a distal lobe which slopes cervically from the central lobe to end in a rounded distoincisal shoulder. Incisor crowns (particularly laterals) showing a prominent central lobe with little or no apparent development of the mesial and distal lobes appear conical in outline and are generally referred to as "peg-shaped".

Peg-shaped incisors may be a conspicuous feature of erupted cleft dentitions. Here they may feature a prominent lingual tubercle in addition to the apparent lack of development of mesial and distal lobes.

INCIDENCE.

Castaldi et al (1966, p.156) in a survey of permanent tooth anomalies reported that they did not find any peg teeth in their sample. They cite Clayton as having found an incidence of 0.3 percent.
Jordan et al (1966, p.49) obtained an incidence figure of 11 percent for cleft models and 12.5 percent for cleft foetuses. Moskin and Gorlin (1963, p.1477) found an incidence of 0.88 percent. They present a table of frequency of occurrence of peg teeth and the figures vary from a maximum of 2.5 percent to a minimum of 0.52 percent. The same authors demonstrated (p.1479) that females had a higher frequency of peg-shaping of lateral incisors. There was also a predominance of left sided occurrence.
ABNORMAL CROWN FORM

Thoma and Goldman (1960, p. 80) state that both local and systemic conditions may affect the form and structure of the developing teeth. In some cases only the gross appearance of the tooth is affected, the structure remaining normal; in others, the structure itself is changed or both form and structure may be involved.

Boyle (1955, p. 19) mentions that the crown of a tooth may lack a cusp that is present normally or have additional cusps. The supplementary cusp on the lingual surface of upper molars is known as the cusp of Carabelli. The maxillary incisors may develop a tubercle or accessory cusp arising from the cervicolingual ridge.

It has already been stated that supernumerary teeth in the margins of an alveolar cleft show variations in crown form. Sir Frank Colyer (1926, p. 614), in discussing abnormally shaped teeth from the region of the premaxilla, states that abnormally shaped teeth are divided into conical and tubercular shape. He describes the variations in form that can occur.
A frequent variation in crown form is one affecting the incisors, when the palatal ridge is elevated and nearly on a level with incisal margin and connected with this by a crest. This has been described as "T" form by Bolk and Bohn (1963, p.14). The labial surface of the tooth has a more or less median vertical groove, giving the incisal edge an inward angle. Colyer (1926, p.614) has described a similar type with slight variation from the T-form. Bohn has suggested that this might be a specific form of abnormality of teeth found with cleft defects but the numerous variations of form reported by Colyer (1926) suggests that this is not so. Teeth of similar form have also been found in the dentition of normal children by Beresford (1965, p.123) and De Jonge (1954, p.154). It is possible that this elevation of the cingulum is one stage in the formation of a supernumerary incisor tooth. (Dixon 1966, p.182). In the permanent dentition the lateral incisor was frequently conical in form and occasionally an x-ray revealed the condition of dens-in-dente which also may be considered as a tendency towards the formation of an extra tooth (Dixon 1966).
Photograph shows hyperdevelopment of lingual tubercle on lateral incisor.

Photograph shows excessive development of lingual cusp on first bicuspid.

Dixon (1966, p.182) has noted the tendency for a supernumerary lateral incisor on the mesial margin of a cleft in both dentitions to resemble the adjacent central incisor. The development of a cleft possibly influences the dental lamina at a very early stage, possibly before the development of the individual teeth, as Glasstone's (1952) study has shown that
tooth germs are independent of external mechanical conditions in the developing jaw in respect of their morphology.

Jordan et al (1966, p.43) describes deviant forms of the mandibular first bicuspid. Three marked deviations from normal morphology are found in cleft dentitions. One is marked by reduction of the lingual cusp, absence of the transverse ridge joining both cusps and a deep, exaggerated mesiolingual groove. Another is marked by a deep invagination of the occlusal surface in the area normally occupied by the lingual cusp. The third features four cusps.

Irregular lobes in the incisors may be another feature. The malformation often takes the form of a pronounced incisal fissure especially in the lateral incisor adjacent to the cleft. (Jordan et al 1966, p.46).

**ABNORMAL ROOT FORM.**

The aetiology of this condition has already been described.

Boyle (1955, p.20) states that the root, like the crown, may be unusually large or small. Accessory roots are also very common. In cleft conditions, the additional feature of "dilaceration" has already been noted.
ABNORMALITIES OF TOOTH STRUCTURE

HYPOPLASIA.

A marked abnormality of tooth structure which is very often seen is imperfect formation of the teeth as shown by the presence of enamel hypoplasia. (Dixon 1966, p.183). This condition shows defects of formation of the enamel and dentine at stages coincident with times of surgical repairs to the lip and palate.

The orthodontic treatment of children with cleft conditions is often rendered impracticable by the condition of the teeth. This is partly due to dental caries and partly to defective structure of the teeth.

Dixon (1963, p.183) conducted a study of seventy-six cleft children and showed that a significantly high incidence of enamel hypoplasia was found in the cleft group as compared with a normal control group.

Clinical and histological material provides some evidence of a connection existing between this defect and the trauma of surgical repair of cleft lip and palate. Ground sections have been prepared of a number of incisor teeth extracted from the margins of clefts. These revealed defects in the cervical part of the crown in the deciduous incisors and the
permanent incisors showed defects of the tip of the crown and further defects in the central portion of the crown (Dixon 1963, p.183). Location of such defects could be related to the times of reparative surgery carried out in the individual patient.

Mink (1959, p.652) tested the degree of hypoplasia in a group of cleft patients and then related it to the original cleft lip or palate classification. He found that the more severe the cleft, the greater the incidence of hypoplasia.

Dixon (1966, p.183) carried out another incidence study of hypoplasia. The result is shown in tabular form below.

Enamel hypoplasia of permanent teeth in patient study group and control group (upper teeth at top; lower teeth below).
Compared with the teeth of children in the control groups, children with cleft defects showed a very high incidence of enamel hypoplasia on the incisor teeth of both dentitions and a somewhat higher incidence in the deciduous molars and first permanent molars. The only teeth which did not show a significant increase in hypoplasia were premolars, which start to calcify at 1½ to 2½ years of age. There is also a close relationship of hypoplasia with the side of cleft involvement and also with the type of operative repair the patient has undergone (1966, p.183).

These findings suggest surgical repairs of the lip can indirectly or directly, through circulatory change, influence the formation of underlying teeth, especially as these are often superficially placed; while the palatal repair may also influence tooth development indirectly due to vascular upsets and systemic upsets.

It is worth mentioning that enamel hypoplasia has also been associated with other developmental disturbances, particularly by Grahnen and Larsen (1958) who showed a similar but symmetrical pattern in patients with a history of prematurity.
In cleft cases the hypoplasia is usually localized to the area of the disturbance (Johnson 1967, p. 442).

It is of interest to note that Barnard (1966) found the prevalence of hypoplasia in Sydney children to increase with age to twelve or thirteen years. It is 6 ± 8 per cent at twelve years of age.
MATERIALS AND METHODS

(a) Purpose of Study.

Malformations and malpositions of the teeth are common in children afflicted with the cleft lip or palate deformity. Estimates of the frequency of cleft lip and palate taken from various sources is tabulated below. (Modified after Greene, 1963).

<table>
<thead>
<tr>
<th>Year</th>
<th>Location</th>
<th>Number of Cases</th>
<th>Sample Size</th>
<th>Cases per One Thousand Births</th>
</tr>
</thead>
<tbody>
<tr>
<td>1928~37</td>
<td>Gothenberg, Sweden</td>
<td>28</td>
<td>27,000</td>
<td>1.04</td>
</tr>
<tr>
<td>1910~40</td>
<td>Denmark</td>
<td>193</td>
<td>128,306</td>
<td>1.50</td>
</tr>
<tr>
<td>1935~44</td>
<td>Wisconsin</td>
<td>736</td>
<td>567,509</td>
<td>1.30</td>
</tr>
<tr>
<td>1943~49</td>
<td>Ontario, Canada</td>
<td>695</td>
<td>655,322</td>
<td>1.06</td>
</tr>
<tr>
<td>1948~50</td>
<td>Pennsylvania</td>
<td>766</td>
<td>583,690</td>
<td>1.31</td>
</tr>
<tr>
<td>1948~55</td>
<td>New York</td>
<td>1414</td>
<td>1,242,744</td>
<td>1.14</td>
</tr>
<tr>
<td>1940~50</td>
<td>Birmingham, England</td>
<td>285</td>
<td>218,693</td>
<td>1.30</td>
</tr>
<tr>
<td>1951~55</td>
<td>Pennsylvania</td>
<td>1592</td>
<td>1,201,976</td>
<td>1.32</td>
</tr>
<tr>
<td>1955</td>
<td>California</td>
<td>368</td>
<td>313,164</td>
<td>1.18</td>
</tr>
<tr>
<td>1953~57</td>
<td>Denmark</td>
<td>644</td>
<td>393,457</td>
<td>1.64</td>
</tr>
<tr>
<td>1945~57</td>
<td>Tasmania</td>
<td>160</td>
<td>96,510</td>
<td>1.66</td>
</tr>
<tr>
<td>1956~60</td>
<td>California</td>
<td>2185</td>
<td>1,765,746</td>
<td>1.24</td>
</tr>
<tr>
<td>1956~60</td>
<td>Hawaii</td>
<td>128</td>
<td>85,180</td>
<td>1.50</td>
</tr>
<tr>
<td>1956~60</td>
<td>Pennsylvania</td>
<td>1446</td>
<td>1,242,908</td>
<td>1.16</td>
</tr>
<tr>
<td>1956~60</td>
<td>Wisconsin</td>
<td>692</td>
<td>485,104</td>
<td>1.43</td>
</tr>
</tbody>
</table>
Fogh-Andersen (1963) asserts that the incidence of cleft lip and palate has almost doubled in the past century rising from 0.7 per 1,000 live births one hundred years ago to at least 1.5 today. This takes into account the decreased infant mortality rate nowadays, as well as the negligible operation mortality. The table below gives the distribution of the various types of clefts by sex and percentage of total cases. (Modified from Greene, 1963).

<table>
<thead>
<tr>
<th>Malformation and Study Location</th>
<th>Number of Cases</th>
<th>Percentage of total cases in study</th>
<th>Males</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft Lip</td>
<td>873</td>
<td>22.3</td>
<td>564</td>
<td>64.6</td>
</tr>
<tr>
<td>Tasmania</td>
<td>50</td>
<td>22.6</td>
<td>32</td>
<td>64</td>
</tr>
<tr>
<td>Cleft Lip and Cleft Palate 1893</td>
<td>1893</td>
<td>48.5</td>
<td>1243</td>
<td>65.7</td>
</tr>
<tr>
<td>Tasmania</td>
<td>97</td>
<td>43.9</td>
<td>74</td>
<td>76.3</td>
</tr>
<tr>
<td>Cleft Palate 1141</td>
<td>1141</td>
<td>29.2</td>
<td>465</td>
<td>40.8</td>
</tr>
<tr>
<td>Tasmania</td>
<td>74</td>
<td>33.5</td>
<td>32</td>
<td>43.2</td>
</tr>
</tbody>
</table>

The following conclusions can be drawn:

1. Cleft lip and cleft palate occur more frequently together than separately.
2. Clefts of the lip with or without associated clefts of the palate are more common in the male than in the female (65:35)
3. Cleft palate cases are more common in the female (60:40).

4. When all types of clefts are considered together, males are more frequently affected (58:42).

Many investigators have noted the preponderance of clefts of the left side of the lip. From 61 to 77.9 percent of the unilateral clefts with or without associated cleft palate are reported to occur on the left side. No satisfactory explanation for this is available.

The purpose of the study has already been noted in the introduction. This is essentially a pilot study. With this reservation in mind, results obtained cannot be strictly comparable to those of other reported studies, nor is there any point in statistical analyses of the data to evaluate the significance of the occurrence of various associations of anomalies.

(b) Material Used.

All the children studied were afflicted with the cleft deformity. The majority were patients attending the Cleft Palate Clinic at the Royal Alexandra Hospital for Children at Camperdown. Eleven patients were from the files of the University Orthodontic Clinic at the United Dental Hospital, Sydney.
The ages of the sample ranged from seven

to fourteen years.

Only anomalies of the permanent dentition

were recorded.

One hundred and ninety children were studied.

This total was made up of one hundred and ten males

and eighty females.

(c) Methods Used.

A mouth mirror, plaster casts, full mouth and

occlusal radiographs were used.

A recording chart was devised to record the

patient's name, sex, age, address and hospital

registration number, type of cleft and anomalies

present. A "remarks" column was inserted to record

any additional features that warranted inclusion.

All missing teeth, supernumerary teeth, fissural

teeth and abnormal root forms were verified with

radiographs. In the recording of peg teeth, abnormal

crown and root forms and hypoplasia, only the gross

discrepancies were noted.

Inadequacies of available radiographs and

differences in interpretations will contribute to

errors in recording.

RESULTS.

These are tabulated on the following pages.
**TABLE I.**

**DISTRIBUTION OF CLEFT TYPES* ACCORDING TO SEX.**

<table>
<thead>
<tr>
<th>Clefts of Primary Palate Only</th>
<th>Number</th>
<th>Males</th>
<th>Females</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Percentage of Total Group</td>
<td></td>
<td>58.3</td>
<td>41.7</td>
<td>100</td>
</tr>
</tbody>
</table>

| Cleft of Secondary Palate Only | Number | 5     | 16      | 21    |
| Percentage of Total Group     |        | 23.8  | 76.2    | 100   |

| Unilateral Clefts of Primary and Secondary Palates | Number | 71     | 36      | 107   |
| Percentage of Total Group |        | 66.4   | 33.6    | 100   |

| Bilateral Clefts of Primary and Secondary Palates | Number | 20     | 18      | 38    |
| Percentage of Total Group |        | 52.6   | 47.4    | 100   |

| Total of Clefts | Number | 110    | 80      | 190   |
| Percentage of Total Male and Female |        | 57.9   | 42.1    | 100   |

* Note: 1. There were two cases of bilateral clefts of the primary palate only. These were classified, for convenience, with bilateral clefts of the primary and secondary palates.
2. Incomplete (subtotal) clefts were classified as complete for convenience.
<table>
<thead>
<tr>
<th></th>
<th>LEFT</th>
<th>RIGHT</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Unilateral Clefts of Primary Palate</strong></td>
<td>9</td>
<td>15</td>
<td>24</td>
</tr>
<tr>
<td>Number</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Percentage of Total</td>
<td>37.5</td>
<td>62.5</td>
<td>100</td>
</tr>
<tr>
<td><strong>Unilateral Clefts of Primary and Secondary Palates</strong></td>
<td>73</td>
<td>34</td>
<td>107</td>
</tr>
<tr>
<td>Number</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Percentage of Total</td>
<td>68.2</td>
<td>31.8</td>
<td>100</td>
</tr>
<tr>
<td><strong>Total Unilateral Clefts</strong></td>
<td>82</td>
<td>49</td>
<td>131</td>
</tr>
<tr>
<td>Number</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Percentage of all Unilateral Clefts</td>
<td>62.6</td>
<td>37.4</td>
<td>100</td>
</tr>
</tbody>
</table>

Note: There were cases of cleft of the secondary palate only. Good radiographic evidence is necessary for identifying side of cleft. Hence, this identification has been omitted.
<table>
<thead>
<tr>
<th>Condition</th>
<th>MALE</th>
<th>FEMALE</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Missing Teeth</td>
<td>42</td>
<td>36</td>
<td>78</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>22.1</td>
<td>10.9</td>
<td></td>
</tr>
<tr>
<td>Supernumerary Teeth</td>
<td>20</td>
<td>5</td>
<td>25</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>10.5</td>
<td>2.6</td>
<td></td>
</tr>
<tr>
<td>Fissural Teeth</td>
<td>45</td>
<td>17</td>
<td>62</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>23.7</td>
<td>3.9</td>
<td></td>
</tr>
<tr>
<td>Peg Teeth</td>
<td>3</td>
<td>5</td>
<td>8</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>1.6</td>
<td>2.6</td>
<td></td>
</tr>
<tr>
<td>Abnormal Crown Forms</td>
<td>14</td>
<td>18</td>
<td>32</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>7.4</td>
<td>9.5</td>
<td></td>
</tr>
<tr>
<td>Abnormal Root Forms</td>
<td>7</td>
<td>5</td>
<td>12</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>3.7</td>
<td>2.6</td>
<td></td>
</tr>
<tr>
<td>Hypoplasia</td>
<td>26</td>
<td>18</td>
<td>44</td>
</tr>
<tr>
<td>Percentage of Total Cleft Cases</td>
<td>13.7</td>
<td>9.5</td>
<td></td>
</tr>
</tbody>
</table>
# TABLE IV.

## SITES OF MISSING TEETH

<table>
<thead>
<tr>
<th></th>
<th>MAXILLA</th>
<th>MANDIBLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$I_1$</td>
<td>$I_2$ C</td>
</tr>
<tr>
<td>Unilateral Clefts of Primary Palate</td>
<td>-</td>
<td>6</td>
</tr>
<tr>
<td>Clefts of Secondary Palate</td>
<td>-</td>
<td>2</td>
</tr>
<tr>
<td>Unilateral Clefts of Primary and Secondary Palates</td>
<td>2</td>
<td>37</td>
</tr>
<tr>
<td>Bilateral Clefts of Primary and Secondary Palates</td>
<td>5</td>
<td>34</td>
</tr>
</tbody>
</table>

Note: $I_1$ = Central incisor  
$I_2$ = Lateral incisor  
C = Canine  
$P_1$ = First premolar  
$P_2$ = Second Premolar.
### CASES SHOWING NUMBERS OF MISSING TEETH

<table>
<thead>
<tr>
<th>Number of Missing Teeth</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5 or More</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Per Case</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of Cases</td>
<td>44</td>
<td>27</td>
<td>8</td>
<td>5</td>
<td>2</td>
<td>152</td>
</tr>
</tbody>
</table>
### TABLE VI.

**SITES OF SUPERNUMERARY TEETH.**

<table>
<thead>
<tr>
<th></th>
<th>MAXILLA</th>
<th>MANDIBLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$I_1$</td>
<td>$I_2$</td>
</tr>
<tr>
<td>Unilateral Clefts of Primary</td>
<td>-</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clefts of Secondary</td>
<td>1</td>
<td>-</td>
</tr>
<tr>
<td>Palate</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unilateral Clefts of Primary and Secondary</td>
<td>-</td>
<td>10</td>
</tr>
<tr>
<td>Palates</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bilateral Clefts of Primary and Secondary Palates</td>
<td>-</td>
<td>6</td>
</tr>
</tbody>
</table>

**Note:** There were no cases showing more than one supernumerary tooth per case.
TABLE VII

DISTRIBUTION OF FISSURAL TEETH

<table>
<thead>
<tr>
<th></th>
<th>Mesial to Cleft</th>
<th>Distal to Cleft</th>
</tr>
</thead>
<tbody>
<tr>
<td>Clefts of Primary</td>
<td>-</td>
<td>2</td>
</tr>
<tr>
<td>Palate</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clefts of Secondary</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Palate</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unilateral Clefts</td>
<td>6</td>
<td>36</td>
</tr>
<tr>
<td>of Primary and Secondary Palates</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bilateral Clefts</td>
<td>2</td>
<td>16</td>
</tr>
<tr>
<td>of Primary and Secondary Palates</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

TABLE VIII

CASES SHOWING NUMBERS OF FISSURAL TEETH

<table>
<thead>
<tr>
<th></th>
<th>One</th>
<th>Two *</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>57</td>
<td>5</td>
<td>62</td>
</tr>
</tbody>
</table>

* Note: All cases showing two fissural teeth were bilateral clefts of primary and secondary palates.
### TABLE IX

**DISTRIBUTION OF PEG LATERAL INCISORS**

<table>
<thead>
<tr>
<th>Upper Lateral Incisor</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Clefts of Primary Palate</td>
<td>4</td>
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<tr>
<td>Clefts of Secondary Palate</td>
<td>1</td>
</tr>
<tr>
<td>Unilateral Clefts of Primary and Secondary Palates</td>
<td>3</td>
</tr>
<tr>
<td>Bilateral Clefts of Primary and Secondary Palates</td>
<td></td>
</tr>
</tbody>
</table>

**Note:**
(1) All cases of peg teeth seen were upper lateral incisors.
(2) No cases were seen which showed more than one per tooth per case.
### TABLE X

**SITES OF ABNORMAL CROWN FORMS**

<table>
<thead>
<tr>
<th></th>
<th>MAXILLA</th>
<th></th>
<th></th>
<th>MANDIBLE</th>
<th></th>
<th></th>
<th></th>
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<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>I₁</td>
<td>I₂</td>
<td>C</td>
<td>P₁</td>
<td>P₂</td>
<td>M₁</td>
<td>I₁</td>
<td>I₂</td>
<td>C</td>
<td>P₁</td>
</tr>
<tr>
<td>Clefts of Primary</td>
<td>-</td>
<td>3</td>
<td>1</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
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</tr>
<tr>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clefts of Secondary</td>
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<td>-</td>
<td>1</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
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</tr>
<tr>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unilateral Clefts of</td>
<td>5 6 10 6 3</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Primary and Secondary</td>
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</tr>
<tr>
<td>Bilateral Clefts of</td>
<td>1 1 4</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
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<td>-</td>
<td>-</td>
<td>-</td>
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</tr>
<tr>
<td>Primary and Secondary</td>
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<tr>
<td>Palates</td>
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<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
</tr>
</tbody>
</table>

**Note**: $M₁ =$ First Permanent Molar.
<table>
<thead>
<tr>
<th>Number of Teeth per Case</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4 or More</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of Cases</td>
<td>20</td>
<td>20</td>
<td>-</td>
<td>1</td>
<td>44</td>
</tr>
</tbody>
</table>
TABLE XII

SITES OF ABNORMAL ROOT FORMS

<table>
<thead>
<tr>
<th>CLEFTS OF PRIMARY PALATE</th>
<th>MAXILLA</th>
<th>MANDIBLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( I_1 )</td>
<td>( I_2 )</td>
</tr>
<tr>
<td>CLEFTS OF SECONDARY PALATE</td>
<td>-</td>
<td>1</td>
</tr>
<tr>
<td>UNILATERAL CLEFTS OF PRIMARY AND SECONDARY PALATES</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>BILATERAL CLEFTS OF PRIMARY AND SECONDARY PALATES</td>
<td>-</td>
<td>2</td>
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</tbody>
</table>
### TABLE XIII

**CASES SHOWING NUMBERS OF TEETH WITH ABNORMAL ROOT FORMS.**

<table>
<thead>
<tr>
<th>Number of Teeth</th>
<th>1</th>
<th>2</th>
<th>3 or More</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of Cases</td>
<td>11</td>
<td>2</td>
<td>-</td>
<td>13</td>
</tr>
<tr>
<td></td>
<td>MAXILLA</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>---------------------</td>
<td>---------</td>
<td>-------</td>
<td>-------</td>
<td>-------</td>
</tr>
<tr>
<td></td>
<td>I₁</td>
<td>I₂</td>
<td>C</td>
<td>P₁</td>
</tr>
<tr>
<td>Clefts of Primary</td>
<td>3</td>
<td>-</td>
<td>-</td>
<td>-</td>
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<tr>
<td>Palate</td>
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<td></td>
</tr>
<tr>
<td>Clefts of Secondary</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Palate</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unilateral Clefts of</td>
<td>27</td>
<td>3</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Primary and Secondary</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Palates</td>
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<td>12</td>
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<tr>
<td>of Primary and</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Secondary Palates.</td>
<td></td>
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</tbody>
</table>
### TABLE XV

**CASES SHOWING NUMBERS OF TEETH WITH HYPOPLASIA**

<table>
<thead>
<tr>
<th>Number of Teeth with Hypoplasia Per Case</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4 or More</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of Cases</td>
<td>28</td>
<td>7</td>
<td>1</td>
<td>1</td>
<td>37</td>
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</tbody>
</table>
DISCUSSION

These results clearly show that dental anomalies in individuals afflicted with cleft lip and/or palate occur throughout the entire dentition and not merely in the maxilla, in the immediate area of the cleft. This is in complete disagreement with Kirkham (1931, p.1078) who stated "there should be no special reason why the dentition should be disturbed except in the lateral incisor area."

This small study has borne out Greene's contentions (1964, p.391). Greene had said that cleft lip and cleft palate occur more frequently together than separately. Although the data has not been examined for statistical significance clefts of the primary palate appear more common in boys and clefts involving the secondary palate alone are more common in girls. In clefts of the primary and secondary palates, boys are affected more frequently than girls. The preponderance of clefts of the left side has also been borne out in the study.

A study of Table III reveals that the most frequently appearing anomalies were missing teeth and fissural teeth. Jordan et al (1966, p.49) found that in their study, the most frequent traits were

(1) thick curved incisors and (2) missing teeth.
With peg teeth as the next most frequent trait. In this same study, Jordan et al found that over half of the cleft individuals in their group were affected, whereas only thirteen percent of his normal group were affected. They also state that "an individual with a cleft who is affected with abnormal teeth is apt to have multiple abnormalities, whereas, an individual so affected but without a cleft, is likely to have but a single dental abnormality." Jordan et al (1966, p.51) therefore postulate that the "factor (or factors) producing the cleft has a definite bearing upon the number of dental abnormalities that occur."

A finding of Kraus et al (1966, p.1739) that was not entirely born out by the findings of this study, was their contention that in cases of cleft palate alone, there are almost as many abnormalities found in the mandibular arch as in the maxillary, but in clefts in which the maxillary ridge is involved, there are almost four times as many abnormalities in the maxillary dentition as in the mandibular. Anterior teeth are affected, in general, almost twice as much as posterior teeth (Kraus et al, 1966, p.1740).

This study agrees with Kraus et al (1966, p.1740) that in all types of clefts"the incisors are more frequently affected than other teeth."
Since many units of the dentition far removed from the site of the cleft are affected, what is the nature of the factor or factors responsible for both cleft and dental abnormalities? As Jordan et al (1966, p.52) state, "it is the fact of cleft not the nature of the cleft, which is significantly involved with the appearance of abnormalities of dental morphology."

The occurrence of supernumerary teeth or missing teeth was confined to the maxillary dentition and apparently was directly correlated with the physical fact of the cleft (Jordan et al 1966, p.52).

Jordan et al (1966, p.53) found that the higher frequency of dental anomalies associated with clefts supports the non-genetic association of diffuse bodily abnormalities in cleft populations. They say that "it is quite obvious that the picture is not that of a hereditary syndrome." They postulate the existence of a factor or factors responsible for the cleft operating also in an apparently haphazard manner, throughout the rest of the body. This must already be at work before the forty-seventh day of foetal life when the palatal shelves begin to fuse.

It must be emphasized that all the reported dental anomalies occur in non-cleft populations but with much less frequency.
SUMMARY AND CONCLUSIONS

One hundred and ninety children afflicted with cleft lip and/or cleft palate conditions were studied for seven abnormal dental traits. The distribution of these anomalies was noted. There appeared to be no correlation between type of cleft and presence or absence of dental anomalies. All anomalies except supernumerary, fissural and peg teeth were found in both maxillary and mandibular dental arches.

It can be concluded that neither the cleft itself nor the type of cleft is an aetiological factor in the occurrence of morphological abnormalities in the individual teeth. It appears that the development of the dentition along with that of the other organs and structures of the body may be affected by the same aetiological factor or factors that are responsible for the cleft lip and/or palate.

A further aetiological agent in the form of surgical trauma as a direct cause, or an indirect action via vascular disturbances to the developing tooth germs, has been put forward.

The pilot study confirmed the existence of a dental problem in the treatment of children afflicted with the cleft deformity. This problem was found to involve not only the maxillary but also the mandibular teeth. The extent of occurrence of the
various dental anomalies demonstrated in the study was such as to indicate the need for their serious consideration in the dental and orthodontic management of children affected by oral and facial clefts.
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