A MORPHOLOGICAL, CINEFLUOROGRAPHIC STUDY OF TRISOMY 21 INDIVIDUALS

A Thesis

Presented to

the Faculty of Graduate Studies and Research

University of Manitoba

In Partial Fulfillment

of the Requirements for the Degree

Master of Science

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by

Leonard Roy Queen

Department of Dental Science

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A MORPHOLOGICAL, CINEFLUOROGRAPHIC STUDY OF TRISOMY 21 INDIVIDUALS

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ABSTRACT

The mongoloid appearance has long been recognized as being distinctively different from that of a "normal" individual.

This investigation of the craniofacial region attempts to integrate information obtained from several radiographic analyses in order to contribute to the understanding of this phenotype. These analyses consisted of an area analysis obtained from a lateral cephalogram, a co-ordinate analysis also using a lateral cephalogram and a functional analysis utilizing a cinefluorographic film sequence. The integration of information obtained from these analyses permitted the evaluation of variations in morphology along with functional activity.

The trisomy 21 group consisted of 40 individuals divided into 25 males and 15 females with an age range of 6 years to 25 years. Each individual was placed in one of 10 groups according to sex and age. A "normal" sample was used for comparison having a similar distribution.

The area analysis consisted of measurements in square millimeters of seven structural areas in the oral nasal region. These areas were computed by tabulating the number of square millimeters in these structures. The co-ordinate analysis consisted of a series of angular

and linear measurements which augmented the results of the area analysis. The functional analysis consisted of the evaluation of five selected stages in the deglutition cycle, with special emphasis being placed on the tongue, soft palate and cervical vertebrae.

The measurements were evaluated statistically by a multivariant analysis. The statistical and subjective evaluation of the results suggest the following conclusions:

1. The areas of the nasal cavity and nasopharynx were found to be significantly smaller in the trisomy 21 group.
2. The oropharynx and oral cavity tended to be larger in the trisomy 21 individuals especially at the adult age level.
3. The soft palate was consistently larger in the trisomy 21 sample.
4. The tongue tended to be smaller in the trisomy 21 group as compared to the normal sample.
5. The pharyngeal tonsil in the trisomy 21 group discontinued involution earlier than the normal group, resulting in a smaller nasopharyngeal airway. As the growth in the nasopharyngeal airway decreased, the plane of the cervical vertebrae to the palatal plane was found to become more obtuse. The resulting

progressive extension of the head increased the distance from the posterior nasal spine to the anterior tubercle of the atlas, thus effectively increasing the size of the oropharyngeal airway in the trisomy 21 group.

6. The tongue in the trisomy 21 group was found to occupy a lower and more protruded position than the comparable normal sample.
7. The tongue posture may be a contributing factor to the high incidence of palatal constriction, posterior crossbites and proclined maxillary incisors found in the trisomy 21 group.
8. The high incidence of anterior open bites may be in part attributable to the obtuse cranial base found in the trisomy 21 group.
9. The apparent mandibular prognathism observed in the trisomy 21 group appears to be related to the differential growth of the maxilla and mandible, as well as, hypoplasia of the anterior cranial base.
10. The typical mongoloid appearance was felt to be due not only to the basic genetic defect but also to adaptive mechanisms involving the tongue and the cervical vertebrae which function in response to a constricted airway. The most important site of this constriction appeared to be the nasopharynx.

11. This thesis presented evidence to support treatment therapy directed at increasing and maintaining a patent airway in the trisomy 21 individual.

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DEDICATION

This thesis is dedicated to the memory of my late mother who gave so unselfishly of herself throughout her life.

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INTRODUCTION

CHAPTER I

INTRODUCTION

During the last two decades a number of studies have analyzed the craniofacial characteristics of trisomy 21 individuals, thereby contributing much to the understanding of this phenotype. Radiographic studies of this area have been an important part of this information. These studies, to date, have been primarily concerned with morphological descriptions and the findings in most parameters measured have illustrated significant differences from accepted norms. The abundance of literature on the craniofacial morphology of the trisomy 21 individuals make them excellent subjects to determine the effect of this structural variation on function.

Adaptability of the oral nasal region has long been discussed in the context of respiration, speech and deglutition; however, the exact mechanisms and the effect of the adaptation on the adjacent structures have been little studied. Furthermore, a review of the literature reveals that few studies have attempted to relate morphological size of structures within the oral nasal region to functions, both vital (respiration) and necessary (deglutition).

An attempt was therefore made to integrate information derived from a series of analyses, both morphological and functional in order to develop a better understanding of the abnormalities and the functional adaptation to them in the trisomy 21 individuals. The morphological analyses consisted of an area analysis and a co-ordinate analysis both obtained from a lateral cephalometric radiograph. The functional aspect of the study consisted of a cine-fluorographic analysis of the function of deglutition which was analysed both subjectively and statistically from five selected stages within each swallow.

The objectives of this study were:

1. To evaluate and compare the morphological size (area) of structures in the oral nasal region in the trisomy 21 group and in a comparable normal group.
2. To evaluate the function of deglutition, from a cine-fluorographic film of the event in both trisomy 21 and normal subjects.
3. To determine whether the trisomy 21 sample studied illustrated the same phenotypic features as did similar studies previously documented.
4. To attempt to integrate information from the various analyses in order to determine possible mechanisms of adaptation to altered morphology.

5. To attempt to interpret the effect of altered function on existing morphology.
6. To examine the patency of the airway in terms of morphology and function.

It is the opinion of the author that the findings in this thesis may be applicable to normal individuals who suffer from similar disorders to those found in the trisomy 21 individuals. As a collorary to this statement, any treatment utilized in trisomy 21 subjects may also be applicable to 'normal' subjects.

REVIEW OF THE LITERATURE

CHAPTER II

REVIEW OF THE LITERATURE

HISTORY OF MONGOLISM

The earliest reported description of mongolism was made by Esquirol (1838) who measured the heads of these individuals and made several astute observations in regard to their facial features. Esquirol's atlas of selected cases illustrated the small heads, depressed nasal root and oblique palpebral fissure. Mention was made of associated mental retardation.

Sequin (1846), in his definitive work, refers to mongoloids as "futuraceous cretins", however, he appeared to sense that he was dealing with a definite syndrome as his description included such clinical features as white peeling skin, unfinished truncated fingers, cracked lips and tongue, red ectopic conjunctiva and unusual susceptibility to respiratory infections.

John Landon Downs (1866) has been credited with the first complete clinical description which defined Downs disease as a separate identity. Emphasis was placed on race degeneration and on the consistant physiognomy of the disease, such as broad flat faces, oblique eyes, epicanthic folds, cracked lips, thick fissured tongues, and small nose. Downs also noted the characteristic thick indistinct speech of the mongoloid.

Shuttleworth (1866), who was considered to be an authority, referred to mongols as "unfinished children", that is, development was incomplete due to "depression of maternal powers".

Frasier and Mitchel (1876) presented the first scientific report on the disease; this was followed by Jones (1890); Oliver (1891); Smith (1896); Garrod (1899). Since that time a considerable volume of material has accumulated relating to Downs syndrome. Brousseau and Brainerd (1928) published a comprehensive review of the literature, Benda's (1946), (1969) monographs included basic research, clinical observations, and pathology, and Kisling (1966) published a comprehensive craniofacial roentgenographic study.

There are many other reported specific investigations; these will be included where they are considered relevant.

Epidemiology

In recent years there has been a renewed interest in the pathogenesis of mongolism. The most recent reports have been colloborated with cytogenetic analyses. In "The Manitoba Study" Uchida (1970), the reported incidence of mongolism varied from 0.90 to 1.35 per 1,000 live births between the years 1961 to 1969. In this analysis, 96% of the mongoloids were trisomic. 2.9% were translocations, equally distributed among three types: t(Gp 21q), sporadic; t(Dq 21q),

sporadic; and t(Dg 21Q), inherited. The frequency of mosaics was roughly one percent of these four more common variants.

Wahrman and Fried (1970) in a comprehensive study in Jerusalem, found an incidence of 2.19 per 1,000 live births, of which 5.7% were due to translocations. These investigators attributed the higher incidence to the thoroughness of their investigation.

Collmann and Stoller (1963) reported a birth incidence of 1.4 per 1,000 births, however earlier epidemiological studies indicated a lower incidence. It was suggested that this discrepancy was due to biased samples or to variation due to race and ethnic groups, Uchida (1970); Lilienfeld (1969). Penrose and Smith (1966), Lilienfeld (1969), and Edwards (1970) suggested that a decline in neonatal mortality due to use of antibiotics and/or an increase in diagnostic standards could cause discrepancies in reported incidences of mongolism.

Etiology of Mongolism

The possibility of an increasing frequency of mongolism is an obvious concern, therefore, the identification of the etiological factors involved is of paramount importance. Epidemiological investigations have been carried out on a series of parameters. One of the well-substantiated observations concerning mongolism is increased incidence with advancing maternal age, Oster (1956); Collmann and Stoller

(1962); Cohen and Lilienfeld (1970); Sever et al. (1970); Wahrman and Fried (1970); Uchida (1970). In the United States, pregnancies in women 35 years of age and older constitute 13.5% of the total number of pregnancies yet contribute to approximately 50% of the mongoloid offspring. There was some indication that there was a decline in the frequency of mongoloid births in much older women, which would suggest that environmental factors may be important in addition to maternal age, Uchida (1970); Wahrman and Fried (1970). Maternal exposure to radiation, particularly fluoroscopy and therapeutic radiation, has been found to be greater in mothers of mongols, Cohen and Lilienfeld (1970); Schuman and Gullen (1970). Uchida and Curtis (1961) concluded that the risk of mongolism in children of abdominal irradiated mothers was approximately four times as great as in the children of control mothers. Schull and Neel (1962) used the data compiled in a genetic study of the offspring of mothers exposed to radiation from the Hiroshima and Nagasaki atomic bombs. The frequency of mongoloid progeny in exposed mothers was estimated to be less than half that of the frequency of the non-exposed. The possibility that high energy radiation plays a role in the etiology of mongolism is as yet undetermined.

Other etiological factors do occur and include race, Sever et al.(1970); Lilienfeld (1969); geographic location, Collmann and Stoller (1962); Sever et al. (1970) and infectious hepatitis, Collmann and Stoller (1962). In addition Nichols (1970) suggested that viruses may be related to production of mongoloids. This was based upon epidemiological data which indicated that 'seasonal cluster' (of mongoloid births) do occur.

The direct mechanism by which the vast majority of chromosome breaking occurs is not known, and any attempt to draw conclusions from the literature concerning the possible cause of the chromosomal disorder can lead to generalizations and an over-simplification of the problem. A multiplicity of agents, rather than one agent, is likely involved in the development of mongolism.

Premature Senility and Cause of Death

The life span of mongoloids is considerably shorter than that of the general population. Much of this difference had been attributed to poor resistance to infection, since following the advent of antibiotics the life span of mongoloids doubled. Even so, the mortality is extremely high, especially in the first half year of life. Wahrman and Fried (1970) reported that only 60.8% reach the age of six months and by

the age of one year 44.7% had died. Forsman and Kesson (1965) also found the survival rate to be extremely low; 20% survive after 30 years of age, 8% after 40 years and 2.6% after 50 years. Jarvis (1970) attributed the "early senile dementia" or "Alzheimers disease" to pathological degeneration of neural tissue.

Penrose and Smith (1966) found respiratory infections to be the most common cause of death, followed by cardiac anomalies. The remainder died from other infectious diseases, tuberculosis, malignancies and accidents.

Physical Symptoms

Mongolism has been diagnosed by a collection of symptoms by several authors. Oster (1953) selected ten cardinal signs of mongolism. This was followed by Hall (1964) with his ten symptoms for diagnosis of the newborn mongoloid. Wahrmann and Fried (1970) illustrated statistically the ten most informative symptoms of mongolism in the newborn. This information was corroborated by a cytogenetic analysis. As oblique palebral fissures occurred in 97.5%, (the most frequent single contributing cause to "mongoloid facies") this feature is a major reason for suspicion of mongolism. Figure 1 , the remaining ten features are:

1. abundant neck skin (94.1%)
2. mouth corners turned down (84.0%)
3. general hypotonia (82.4%)

4. flat face (79.6%)
5. at least one dysplastic ear (78.8%)
6. epicanthus in at least one eye (76.0%)

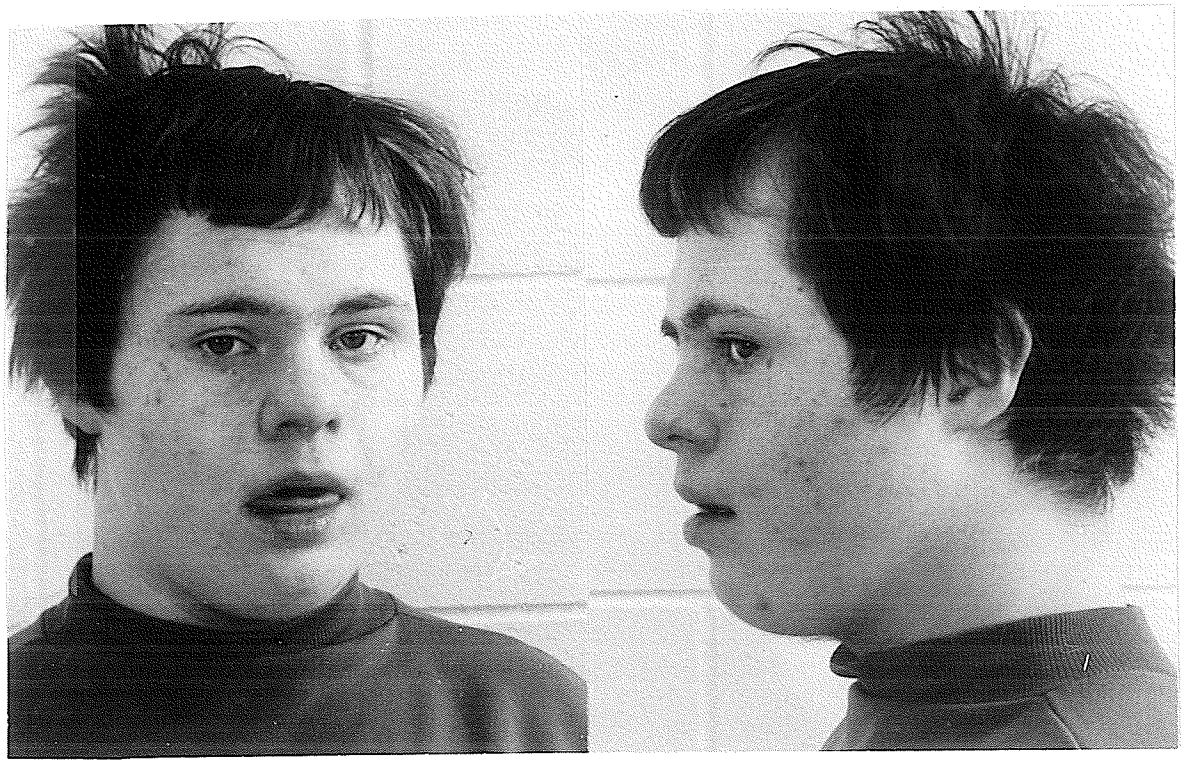

Figure 1. A photograph of a male individual with Trisomy 21 (Downs) Syndrome.

7. gap between toes one and two (67.3%)
8. tongue protruding (62.7%)
9. head circumference at birth not exceeding 32 cm. (42.6%)
10. simian crease in at least one hand (42.3%)

Diagnostically, any infant exhibiting five of these ten signs have been shown, by chromosomal analysis, to be a mongoloid.

It is apparent that an accurate diagnosis may be made on the basis of physical symptoms alone;however, for a complete diagnosis a cytogenetic analysis is essential.

Cytogenetics

The first suggestion that a chromosomal abnormality could be responsible for mongolism was made by Waardenburg (1932). However, not until Tjio and Levan (1956) established the normal chromosomal number of 46 was it possible to identify diseases of chromosome number. In Figure 2, Lejeune et al. (1959) was credited with establishing the connection between mongolism and 47 chromosomes. They described the super-numerary chromosome as small and acrocentric. These findings were subsequently confirmed by several other investigators, Jacobs et al. (1959) using 6 mongols (3 males and 3 females); Book et al. (1959) examined chromosomes of 3 mongols and Ford et al. (1959) examined a mongol with Klinefelters syndrome. All of these investigators concluded that the extra somatic chromosome was morphologically similar to chromosome 21.

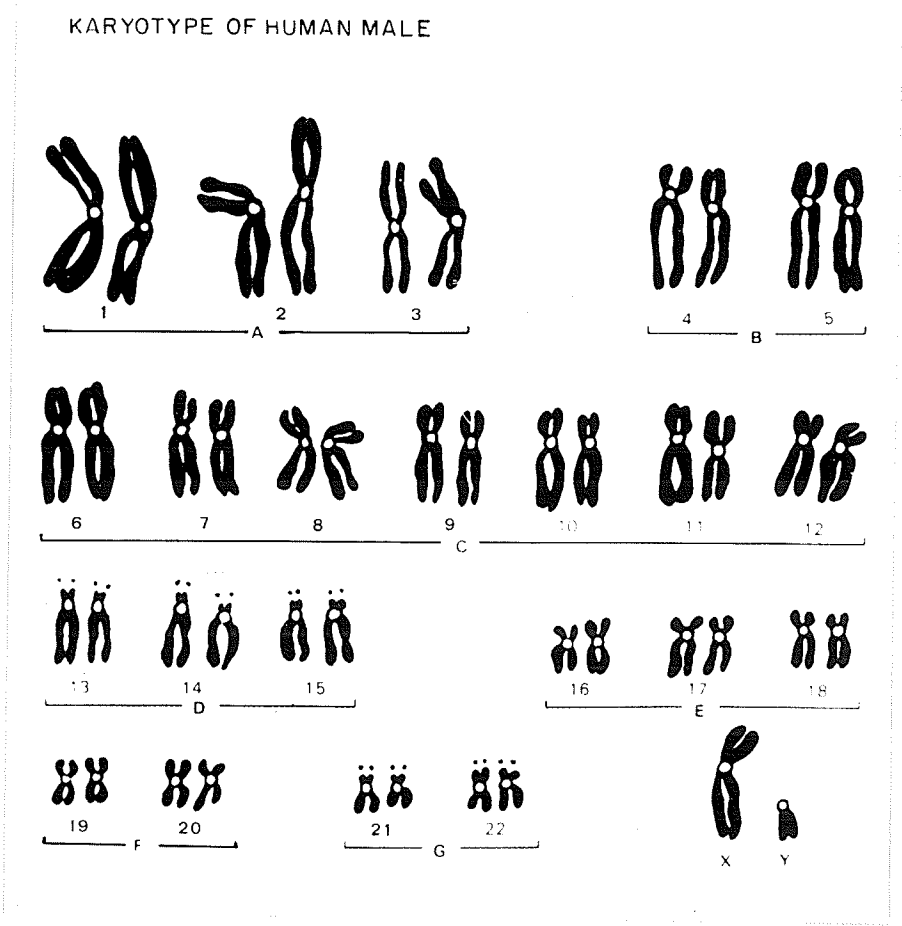

Figure 2. A normal human male Karyotype.

At the Denver Convention (1960) it was agreed that autosomes would be serially numbered 1 to 22 in descending order of length and then divided into seven gorups, each group being designated a letter from A to G, Therman et al. (1961). The presence of an extra chromosome belonging to the G (21-22) group is responsible for mongolism. It was difficult to differentiate chromosome no. 21 from chromosome no. 22 on a size or morphological basis. Yunis et al. (1965a and 1965b) in a DNA replication study using cultured leukocytes suggested that mongolism was trisomy of chromosome number 22. However, by convention, mongolism is assumed to be trisomy of chromosome number 21 (Figure 3).

Mongolism is not a uniform cytogenetic entity. Cases of translocation of chromosomes, as well as normal translocation carriers have been described by Polani et al. (1960); Fracearo et al. (1960) and by Penrose et al. (1960).

Clarke et al. (1961) described another cytogenetic entity, that of an individual presenting both normal and trisomic cellular tissue.

Hamerton et al. (1965) established a classification of the cytogenetic entities responsible for Downs syndrome.

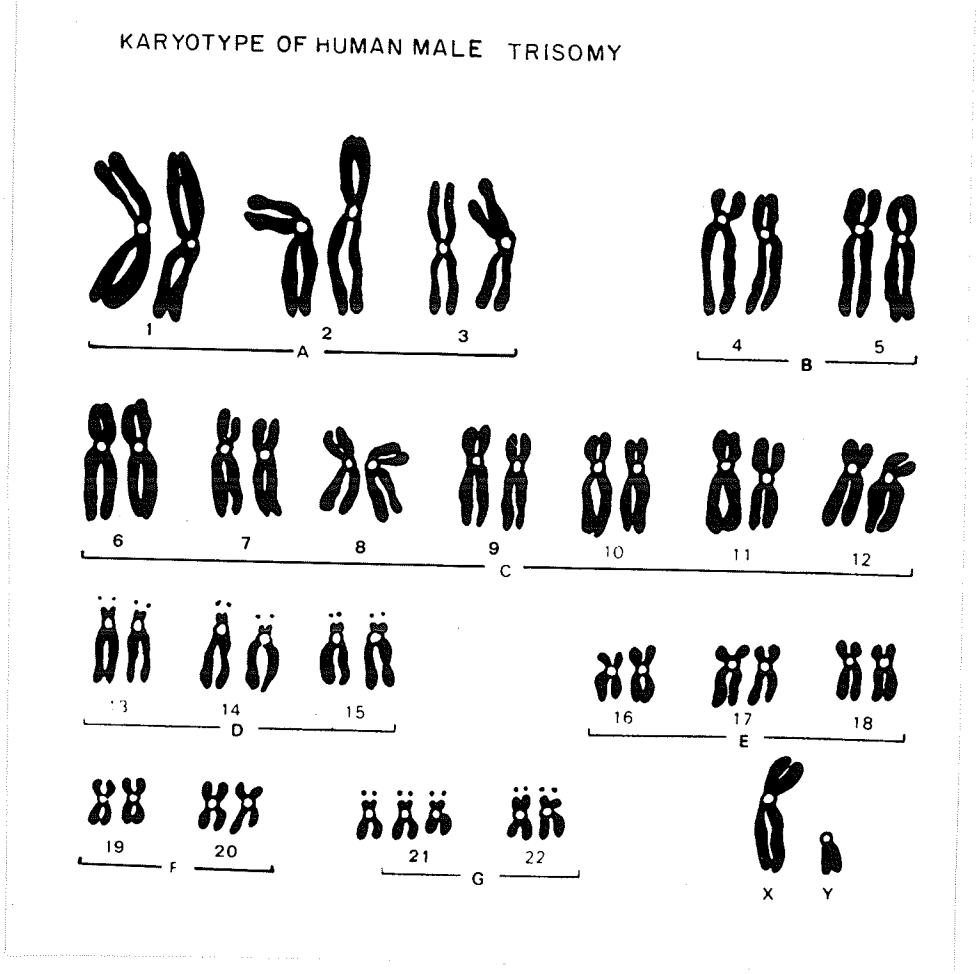

Figure 3. Karyotype of a human male affected by the Trisomy 21 Syndrome.

1. Primary Trisomies

This group has 47 chromosomes, one of which is an additional chromosome in group G, arbitrarily defined as no. 21. The primary trisomies constitute the majority of the mongoloid population (94.5%), Hamerton (1971). This type of abnormality is due to a phenomenon referred to as non-disjunction, which occurs either as a result of failure of hormologous chromosomes to separate during the first of the two meiotic divisions or failure of the chromatid to separate during the first of the two meiotic divisions or failure of the chromatid to separate during the second meiotic division, Penrose and Smith (1966).

2. Secondary Trisomies

Secondary trisomies include mongoloids with 46 chromosomes and in addition a translocation or isochromosome. Mongolism resulting from translocations occurs due to a break near the centromeric region of two acrocentric chromosomes; with the simultaneous fusion of the long arms and the short arms of the two chromosomes. This reorganization leads to two homologous chromosomes, one containing the long arms, the other the short arms and the satellites of the original chromosomes. In Figure 4, the small translocated chromosome was lost in subsequent cell division, Mikkelson (1971). This type of acrocentric translocation was termed "Robertsonian translocation", Robertson (1916).

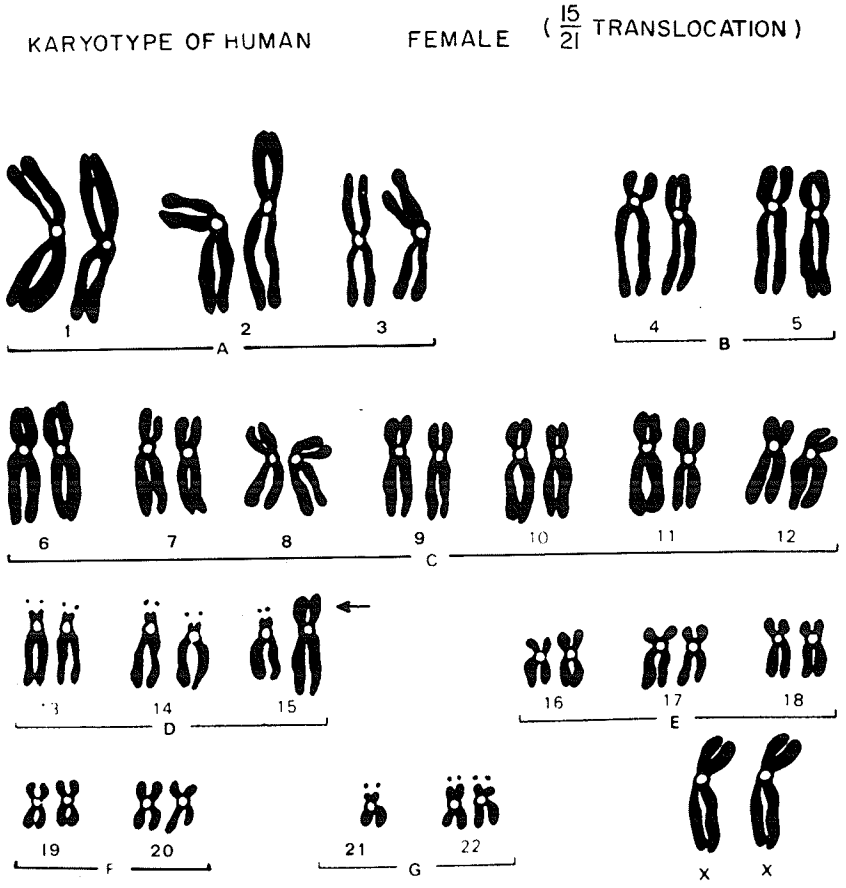

Figure 4. Karyotype of a normal human carrier of the D/G or 15/21 Translocation.

Several types of translocations have been documented, D/G (DgGg) and G/G (GgGg) according to the Chicago nomenclature (1966) were the most common. This latter defect can involve two 21 (homologous) chromosomes, in which case it was termed an isochromosome, Wright et al. (1967). Secondary trisomies constitute about 5.47% of mongoloids; of these 44% were the D/G type and 56% the G/G type. 49% of the D/G were inherited and 51% occurred sporadically, whereas in the G/G translocations 5.6% were inherited and 94% occurred sporadically, Hamerton (1961) (1971). Translocations appear to be more common among mongols born to young mothers — about 8% compared to only 1.5% born to mothers over thirty, Mikkelson (1971).

Mixoploid (Mosaics)

Mosaics referred to individuals who present two or more cell types as a result of mitotic non-dysjunction, Ford (1969).

The most common type reported in mongolism was a mosaic of normal cells and cells with trisomy 21, however, triple type mosaics have been reported. Mitotic non-dysjunction can give rise to mosaics from a normal zygote or from a zygote trisomic for chromosome no. 21. In addition, anaphase "lagging" of a 21 chromosome at the first mitotic division can result in a mosaic individual, Richards (1960).

Mosaics constitute 1% of mongoloids and 10% of atypical cases, Hamerton et al. (1965). The atypical mosaics represented that group which has two chromosomal defects, including trisomy 21 plus Klinefelters (xxy) Ford et al. (1959); Hamerton et al. (1962).

GROWTH OF THE ORAL NASAL REGION

The oral nasal region is an extremely complex region closely aligned with several vital functions: respiration, deglutition, speech and mastication. The stimulation of respiration occurs at birth at which time all of the related organs must be stimulated in a co-ordinated manner to ensure a patent airway, Bosma (1960).

Nasal Area

This area has been little studied. Rosenberger (1934) did a quantitative growth study of this area using the Broadbent Bolton cephalometric sample. He suggested that the downward and forward drifting of the cranium and palate at an early age was responsible for enlarging the nasal and nasopharynx areas. Brodie (1941), using the same cephalometric sample, found similar results. Rosenberger (1934) reported that by the age of 5 years the pattern for upper

facial development was established. Therefore, defects in early childhood persisted throughout life. Brodie (1941) supported this thesis and suggested that facial development was established as early as 3 months of age. Moss (1967), utilizing Van du Klaau's functional matrix concept, placed less emphasis on the early predetermination of the growth process.

Scott (1957) believed that the height of the nasal cavity was determined by growth of the cartilagenous nasal septum.

Rosenberger (1934) using lateral cephalograms, stated that the nasal growth, both antero-posterior and vertical, was completed in females by the age of 14 years, whereas there was a latent vertical growth in males from 16 to 18 years. Scott (1957) stated that the width of the upper portion of the nasal cavity was 75% of its adult dimension by 1 year of age and reached adult portions at 10 years. The lower portion of the nasal cavity increased beyond the tenth year due to remodelling of the lateral nasal wall and the antrum.

Alimchandani (1973) found a linear increase in anterior nasal width at ages 3 to 5 years and 12 to 15 years. Little change was noted thereafter. He also found that the width was smaller at all ages in his trisomy 21 sample as compared to his normal sample.

Nasopharynx

The nasopharynx, in contrast to the nasal area, has been extensively studied.

Schuller (1929) asserted that "the shape and size of the nasopharyngeal cavity is determined to a great extent through the configuration of the surrounding bony structures" and that "anomalies of the shape and size of the nasopharynx arise through congenital or acquired deformities of the base of the skull or of the upper cervical vertebrae".

Antero-Posterior Dimension

Keith and Campion (1922) concluded that growth at the spheno-occipital synchondrosis was important in increasing the area of this space. Krogman (1929) in an anthropological study, attributed the antero-posterior pharyngeal growth to growth of the body and greater wings of the sphenoid bone.

McCarthy (1925) suggested that the anterior tubercle of the first cervical vertebra contributes to the antero-posterior dimension of the nasopharynx. This was supported by Osborne (1968) who found that patients with congenital palatopharyngeal incompetence possessed a significantly greater prevalence of anomalies of the upper cervical vertebrae. Similarily, patients with upper cervical vertebrae anomalies were found to have a greater osseous nasopharyngeal depth. King (1952), using longitudinal lateral cephalometric

material of subjects from 3 months to 16 years, found that forward growth of maxilla and the growth at the spheno-occipital junction antero-posteriorly after the first two years was minimized by forward growth of the anterior tubercle of the atlas. Thus, from this time on there was no appreciable increase in the depth of the nasopharynx.

Ricketts (1954), using lateral laminograph in radiographs and measuring from posterior nasal spine (PNS) to anterior tubercle of atlas (AA), found the osseous nasopharynx to be 42 mm in subjects 12.5 years of age. Brader (1957) measured between anterior tubercle of atlas and pterygomaxillary fissure (PMF) and found the mean distance to be 32 mm in subjects 13 years of age. He also reported the soft tissue depth, as determined by the distance between PMF and the posterior pharyngeal wall, to be 21.4 mm in the controls and 13.6 mm in a corresponding cleft palate group. The difference was attributed to an excess of adenoid tissue in the cleft palate group.

Subtelny (1957) in a similar study, reported considerable fluctuation in the soft tissue depth of the nasopharynx up to eleven years of age. This fluctuation was attributed to adenoid growth and concomitant growth of the upper face. His measurement between the posterior pharyngeal wall and PNS was found to average 22.0 mm at 14 years of age.

Engman (1965) reported the distance between PMF and posterior pharyngeal wall to be 23.7 mm. There was no significant difference between normal subjects and a corresponding cleft group.

Nasopharyngeal Height

The increase in the height of the nasopharynx with age is relatively greater than the increase in depth. King (1952) attributed this to the increased height of cervical vertebrae (posteriorly) and to the descent of the hard palate, pterygoid processes, mandible and hyoid bone (anteriorly).

Ricketts (1954) and Subtelny (1957), using similar analyses for height, reported values of 27 mm and 26.2 mm for ages 12.5 years and 14 years respectively.

Bergland (1963), using anthropological measurements of skull material, reported that the nasopharynx was lower in Lapps than in Norwegians and considerably lower in females than in males in both samples. This data suggests the importance of comparisons between different racial populations in determining general trends.

Pharyngeal Width

Subtelny (1955), utilizing an oriented coronal laminagraph, was able to establish the nasopharyngeal width by

measuring between the medial pterygoid plates. Normal subjects measured 24.61 mm at age 3 or less, and width increase found to plateau during the second year of life.

Bergland (1963) reported that at the age of six an increase in choanal width occurred. This was found in both the Lapps and Norwegian skull samples and was attributed to an adolescent "growth spurt", and to a remodelling of the medial pterygoid laminae. Similar results had previously been reported by Keith and Campion (1922), however these investigators measured only three skulls.

Pharyngeal Tonsil (Adenoid Tissue)

Scammon (1953), studying the growth of lymphatics on cadaver material, found that the quantity of lymphatic tissue increased rapidly in infancy and early childhood. The increase was at a slow rate until it reached a peak at about 10-11 years of age only to be followed by a gradual decline thereafter.

Subtelny et al. (1956), employing both serial cephalometric roentgenography and cephalometric laminagraphy, described the adenoid tissue as following a definite growth cycle. By two years the adenoid tissue was well-defined and could occupy as much as one-half the nasopharyngeal cavity. Growth of this tissue was in a downward and

forward direction and reached its peak as early as 9 to 10 years or as late as 14 to 15 years. In some instances, this tissue essentially obliterated the entire nasopharynx, only to degenerate with subsequent increase in age.

Ricketts (1954), in a study of 'cleft children', indicated that there was the extreme variability in the amount of adenoid tissue in different subjects. Subtelny et al. (1956), illustrated a case where the nasopharynx was completely constricted by adenoid tissue, resulting in a chronic mouth breather with impaired speech. These cases were also extremely susceptibile to pharyngeal infections.

The size of the adenoids and the size of the nasopharynx have been cited as decisive factors for oral or nasal breathing, Ricketts (1954), Subtelny (1954), Lubarth (1960), Moyers (1963).

Linder-Aronson (1970) found a high correlation between nasal air flow and adenoid size, as measured on a lateral cephalometric radiograph. There was also a fairly high correlation between mouth breathing and adenoid size. Dunn et al. (1973) reported that the bigonial width and gonial angle were inversely proportional to the nasopharyngeal airway size.

Linder-Aronson (1974) reported that children who underwent adenoidectomy had significantly greater increases in the angle of the upper and lower incisors to sella-nasion and mandibular plane respectively, than did non-treated children.

The consequences of upper respiratory obstruction are varied. Luscher (1930) reported that the stimulus produced by air currents on the trigeminal nerve in the nasal mucosa played an important part in the movement of the thorax-lung system. Furthermore, Unno et al. (1969) observed decreased elasticity of the lungs and increased pulmonary resistance in 97 subjects with relatively high nasal obstruction. These changes in pulmonary function, which were due to a reflex action, appeared to be reversible.

Chronic upper airway obstruction as a cause of heart failure was first reported by Menasche et al. (1965) followed by Noonan (1965), Luke et al. (1966) and Massumi et al. (1969). In all cases adenoidectomy and tonsillectomy were followed by substantial improvement.

Oropharyngeal Area

This area has been studied only in its association with adjacent areas. Bosma et al. (1960) termed the process of respiration in the new born infant "glossopharyngeal

respiration", since it involved not only the entire pharynx but also the trunk. This is similar to the respiration of patients suffering paralysis of the respiratory muscles due to poliomyelitis and this phenomenon is often associated with extension of the head in order to establish a more patent airway.

Negus (1940) suggested that the tongue, the hyoid and the larynx grew downwards and backwards in the throat. King (1952) found that growth in length of the pharynx occurred from age 3 months to 16 years.

Subtelny (1956) subdivided this area into an anterior cavity (oral cavity) and a posterior cavity (pharyngeal cavity). The pharyngeal cavity was analysed for nasalized vowels in cleft subjects.

Costelli et al. (1973) found that the length of the pharyngeal cavity had a significantly faster rate of growth in boys than in girls.

Soft Palate

Much research has been done on soft palate function but little in terms of its size. Subtelny (1957) conducted a serial cephalometric study on subjects from infancy to early adulthood. Growth in length was most rapid up until 1½ to 2 years of age. This was followed by a levelling off

period where there was a little increase in the soft palate until 4 to 5 years of age. Thereafter there was evidence of a steady, though slower increase, up to late adolescence or early adulthood. The increase in thickness of the soft palate was greatest in the first year of life. The amount of growth thereafter was in small increments, with the maximum being reached at age 14 to 16 years. The angulation of the soft palate to the hard palate became progressively more acute with increase in age.

Subtelny (1956) suggested that removal of adenoid tissue had a considerable effect on subsequent soft palate function. Following adenoidectomy the soft palate must extend a greater distance, necessitating greater muscular activity in order to affect a posterior pharyngeal seal.

Ricketts (1954), utilizing mid-sagittal laminagraphs of children age 12½ years with Class 11 malocclusions, concluded that the size and shape of a complement of structures determined the functional adequacy of the soft palate. These structures included the angulation of the cranial base, the position of the posterior nasal spine, the size of soft palate, the range of function of soft palate and the amount of adenoid tissue. The length of the soft palate varied from 31 mm to 42 mm and the width from 7 mm to 11 mm.

Tongue Size

Many observations have been made regarding the clinical appearance of the mongoloid tongue. Thomson (1907) stated that "It's apparent largeness may, indeed, often be noticed soon after birth; but this appearance is probably attributable rather to an abnormal shortness of the mouth than to any real longness of the tongue.". Benda (1956) also suggested that protrusion of the tongue was due to a small oral cavity rather than a large tongue. Gillis et al. (1968) and Frostad (1970) both described a protruding tongue, however, Gosman (1951) described the mongoloid tongue as being large in a small oral cavity.

The position of the tongue and its relation to other oral structures has been studied by several investigators; Hixon (1949), Peat (1968), Fishman (1969), Bandy (1969). There are, however, few studies which involved the actual measurement of tongue size.

Jelonek (1967) measured the size, weight and volume of tongues from cadavers. An impression was taken of the tongue of the cadaver and then the tongue was excised. It was concluded that, in all cases (fetal to adult) tongue size and arch size were highly correlated.

Bandy (1969), using live subjects and the principle of fluid displacement, devised a method to measure the volume of the anterior portion of the tongue. In this report the volume and length of the tongue showed little correlation with the width and length of the lower dental arch or the position of the incisor teeth.

Vig (1974), utilizing lateral cephalometric radiograms, measured the area of the tongue using a planimeter and suggested that the tongue becomes relatively smaller from 10 years of age to adulthood.

Hixon (1949) studied the tongue as it pertained to speech production. Peat (1968) did a cephalometric study of tongue position as it related to dentoalevolar structures. Fishman (1969) did a postural and dimensional study of the tongue in individuals with normal and abnormal occlusions, as well as, in individuals with and without speech defects.

Cranial Base

Since the cranial base is intimately related to both maxilla and mandible, its angulation and indeed its length, must influence the spatial position of these structures. Moss (1955) stated that there was a direct correlation between the type of malocclusion and cranial base flexure. Fletcher (1957), in a study of growth disturbances and

physiological activity, felt that unusually obtuse cranial angles seemed to be reflected in enlarged antero-posterior dimensions of the nasopharynx. Kisling (1966), in an extensive study of mongolism, reported that the cranial bases of affected individuals were more obtuse and shorter than a comparable control sample. In addition, the anterior cranial base was significantly more affected than the posterior cranial base. The sella was higher and narrower in mongoloids than in the control sample. Similar findings were reported by Ghiz (1969), who reported that the mean angle between nasion-sella-basion in the trisomy 21 to be 138.53^{o} and in the controls to be 131.57^{o}. The distance from sella to nasion in trisomy 21 was 58.10 mm and in the controls 65.02 mm. The distance between sella basion in the trisomy 21 was 36.56 mm and in the controls 40.13 mm. These mean differences were all significant at the 1% level of confidence. Benda (1940), utilizing prepared slides of the cranial base, described insufficient proliferation of cartilagenous tissue in both the sphenooccipital and the sphenoethnoidal synchondrosis, as well as early fusion of these areas in mongoloid subjects.

A more recent study by Nevile (1973) supports Benda's finding that epiphyses of the trisomy 21 fused earlier. Alimchandani (1973) showed similar findings in regard to the cartilage of the nasal septum and the inferior conchae

and suggested that the cranial base abnormalities of the trisomy 21 may, in part, be due to an endochondral growth deficiency, since most bones developing from cartilage appear to be seriously affected.

Head Posture (Craniocervical relationship)

Head posture has long been recognized as a vital physiological feature of man. Schmidt (1876) suggested the need for an anatomic plane within the skull to correspond to the physiological horizontal. This was described - defined at a conference in Frankfurt am Main (1884).

Lundstrom (1955) discussed the variation of the Frankfurt horizontal with the true horizontal as a result of head posture.

Bosna (1960), in a roentgenologic investigation, illustrated that head posture of the new born infant was vital for respiration.

Cleall et al. (1966), in a cinefluorographic investigation of head posture, measured Frankfurt horizontal to true vertical and found the relationship to be essentially perpendicular, being 90.1 degrees with a standard deviation of 3.0 degrees.

Solow and Tallgren (1971), in a cephalometric investigation, utilized the cervical vertebrae and their relationship to the sella nasion plane in a determination of head posture. They suggested that there was an interrelationship between facial morphology and head balancing position.

MATERIALS AND METHODS

CHAPTER III

MATERIALS AND METHODS

I. SAMPLE

The sample consisted of two groups, one a sample of 40 trisomy 21 individuals while the other consisted of 40 normal individuals. The trisomy 21 group were all residents of various institutions and homes in Manitoba and had an age range of 6-24 years. Each of the mongoloid subjects had been karyotyped and found to have an extra chromosome in the G group*. The normal group consisted of 40 Caucasian individuals from the City of Winnipeg and neighbouring area. The age range was 6-24 years.

Each group consisted of 15 females and 25 males, divided by sex and age into 10 subgroups, five for the females and five for the males. The age groups were: (1) 6-10 years of age, (2) 11-12 years of age, (3) 13-14 years of age, (4) 15-17 years of age, (5) 18-24 years of age. A complete outline of subject distribution is illustrated in Table 1.

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*The karyotyping was done by Dr. Irene Uchida, formally of the Department of Medical Genetics, Winnipeg Children's Hospital, Winnipeg, Manitoba, Canada.

TABLE 1

AGE AND SEX DISTRIBUTION OF TRISOMY 21 AND NORMAL GROUPS

SAMPLE SIZE					
TRISOMY	AGE	NUMBER	NORMAL	AGE	NUMBER
MALE			MALE		
GROUP 1	UNDER 10 YRS	2	GROUP 1	UNDER 10 YRS	2
2	11–12	3	2	11–12	3
3	13–14	7	3	13–14	7
4	15–17	6	4	15–17	6
5	18–24	7	5	18–24	7
FEMALE			FEMALE		
GROUP 1	UNDER 10 YRS	2	GROUP 1	UNDER 10 YRS	2
2	11–12	2	2	11–12	2
3	13–14	3	3	13–14	3
4	15–17	2	4	15–17	2
5	18–24	6	5	18–24	6

II. RECORDS

The records obtained on both trisomy 21 and the normal groups were:

1. A lateral cephalometric radiograph in maximum occlusion utilizing a cephalometrix* cephalometer.
2. A cinefluorographic sequence, which included speech and swallowing cycles was obtained by utilizing a Picker** cinefluorographic unit.
3. Panorex radiographs***.
4. Plaster models made from an alginate impression of both arches.

Cephalometric Records

All lateral cephalometric records were obtained by using the cephalometrix cephalometer in the Orthodontic Department at The University of Manitoba.

The subject's head was placed in the cephalostat adjacent to the film cassette, the ear rods were firmly placed in the external auditory meatus and the subjects

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* Moss Corporations, Chicago, Illinois.

** Picker X-Rays Engineering Ltd., 1174 Sanford Road, Winnipeg, Manitoba.

*** S.S. White, Dental Products Division, Philadelphia, Pennsylvania.

were asked to close firmly on their posterior teeth. The Frankfort horizontal was made approximately parallel to the floor. The X-ray beam passed through the head perpendicular to the mid sagittal plane and through the transmeatal axis. The films were produced using an exposure time of 15 millampere seconds and a kilovolt potential of 90 K.V.P.

Cinefluorographic Records

All of the cinefluorographic records were obtained in the Orthodontic Department, Faculty of Dentistry, The University of Manitoba. The Picker cinefluorographic unit utilized an 8-3/4" image intensifier to which was attached to a 16 mm camera. The cinefluorographic record could be monitored throughout the filming sequence on an attached 12" television screen. The film speed was set at 30 frames per second. A Kodak Ectachrome daylight film EF 449 was used to obtain maximum contrast of hard and soft tissue.

Subjects were coached on the cinefluorographic sequence used; however, the trisomy 21 individuals often found the sequence too difficult in which case only an adequate swallow was obtained. The X-ray beam was monitored so that it passed approximately through the maxillary first molar. The restraining ear rods were only used to position the subject and at no time were they used when taking the

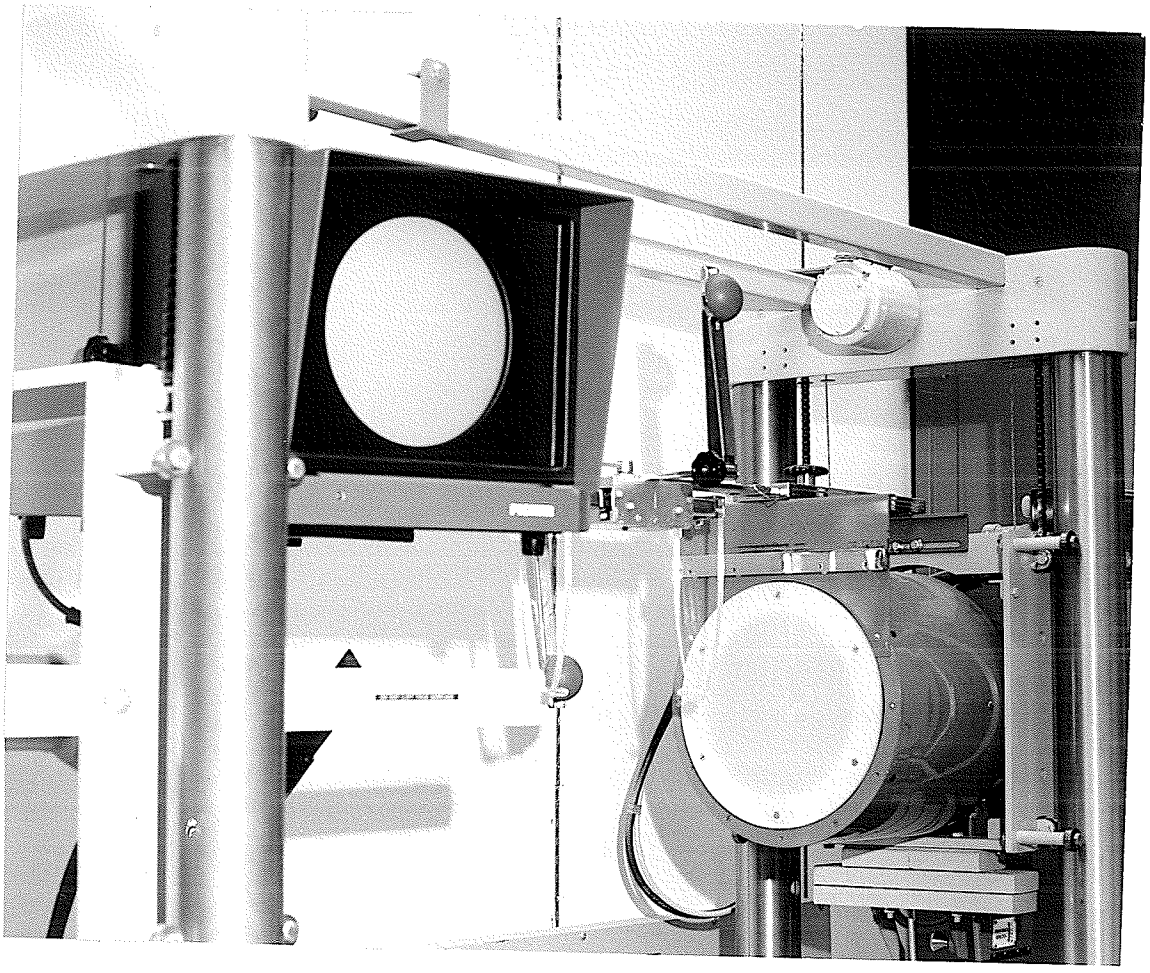

Figure 5. The Picker cinefluorographic unit.

cinefluorographic sequence. In order to better differentiate soft tissue structures, Picker microtrast barium paste was placed in the midline on the tongue and lips. Each subject was given a cup with 5 ml of water and were given the following instructions:

1. Swallow the water.
2. Close on the back teeth.
3. Say "Mississippi" (pause); close on the back teeth.
4. Say "Peter looks silly swimming" (pause); close on the back teeth.
5. Slide lower jaw forward with teeth together, now open wide, close on the back teeth.
6. Slide lower jaw back as far as possible keeping teeth together.
7. Relax and swallow.

III. ANALYSIS OF RECORDS

Cephalometric Area Analyses

Two distinctly different types of analyses were carried out on the lateral cephalogram.

The cephalometric area analysis consisted of two dimensional area measurements of seven specific areas within the cephalogram. The areas were calculated by tabulation of the number of millimeter squares that were in the specific structural areas. For this purpose highly accurate millimeter segmented graft paper was used.

To establish some criteria to determine the accuracy of the tongue area measurement a pilot study was carried out on 7 normal individuals. Special trays were fabricated from hard base plate wax such that they followed the soft tissue contour of the tongue with the teeth in occlusion. Soft alginate was placed under the tongue and the subject was instructed to place the tip of the tongue lightly against the lower incisors. The loaded impression tray was placed on the dorsal surface of the tongue and the subject instructed to close. Nearly complete closure was obtained since the tray was preformed to direct the teeth into maximum inter cuspation.

The impression was poured in dental stone. The stone tongue was then sectioned down its midline and zeroxed. This zeroxed copy of the tongue was then compared with the tracing obtained from the lateral cephalogram. Since the impression invariably did not go posteriorly further than the pterygomaxillary fissure a line was drawn on the zerox copy and the tracing to mark this as the posterior border of the tongue. The areas of both zerox copy and cephalometric tracing were then tabulated. The findings indicated that there was very little difference between either technique in measuring the anterior portion of the tongue and suggested that the lateral cephalogram accurately measured the two dimensional area of the tongue.

The areas, as illustrated in Figure 6, are:

1. Area 1 - This area is referred to as the nasal area. It includes all the area above the maxilla enclosed posteriorly by a perpendicular from the palatal plane upward from the posterior nasal spine and anteriorly by a line from an extension of A point to nasion.
2. Area 2 - This area represents the nasopharynx. It consists of the air space above the palatal plane and posterior to the vertical extension from the posterior nasal spine.
3. Area 3 - This area represents the soft tissue portion of the nasopharynx superior to the palatal plane and enclosed posterior superiorly by the cervical vertebrae and the cranial base, as well as, area number 2.
4. Area 4 - This area is referred to as the oropharynx area and represents the air space inferior to the palatal plane down to the level of the third vertebra . It is enclosed posteriorly by the posterior pharyngeal wall and anteriorly by the tongue and soft palate.

5. Area 5 - This area is referred to as the oral area and is the area superior to the tongue and inferior to the hard palate.
6. Area 6 - This is the area enclosed by the soft palate.
7. Area 7 - This is the area of the tongue; it includes the dorsum surface down to the third cervical vertebra across to the most anterior superior point of the hyoid bone, then anteriorly to the most inferior anterior position of the tongue.

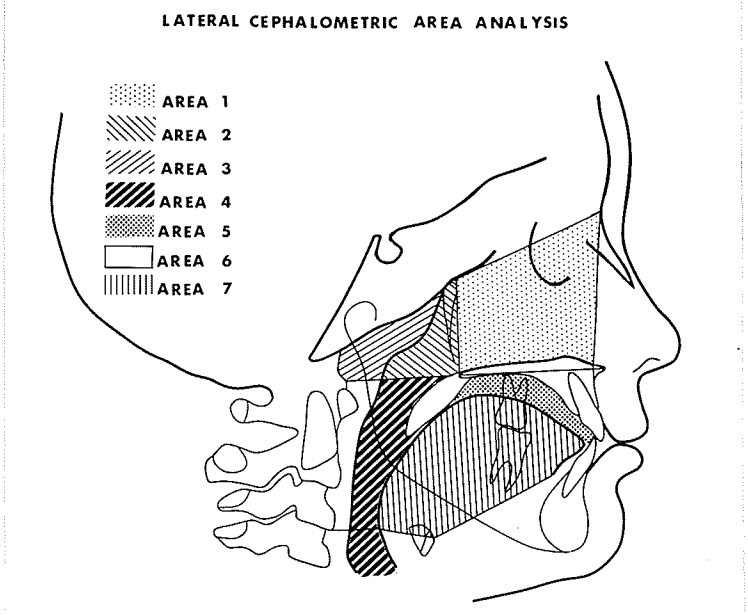

Figure 6. Lateral cephalometric area analysis diagramatic illustration.

Cephalometric Co-Ordinate Analysis

Fifty-nine landmarks were selected to be digitized from the radiographs of the normal and trisomy 21 samples.

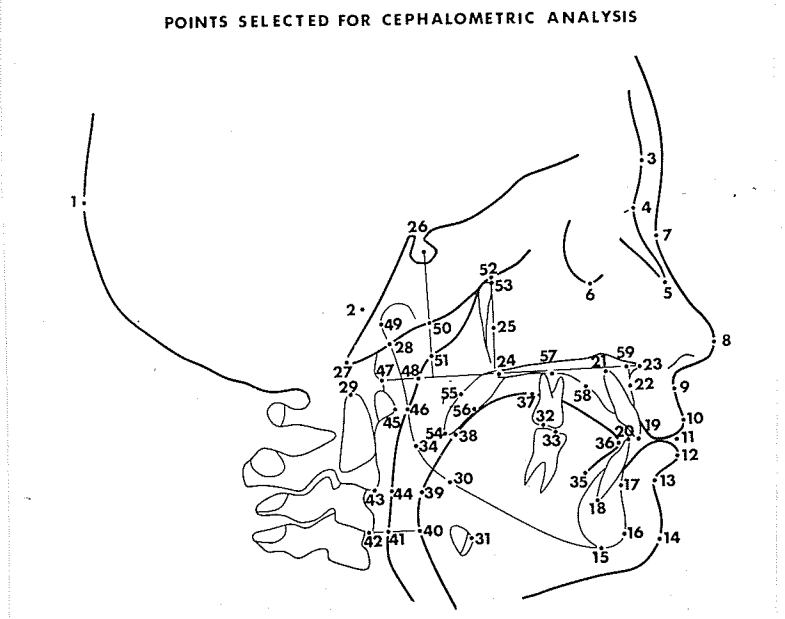

Figure 7. An illustration of the 59 points used in the cephalometric analysis.

The detailed description of these landmarks are defined in the Glossary. The co-ordinates were digitized by utilizing the illuminated screen of a Tagarno motion analyzer to which was attached a Ruscom* strip chart digitizer which recorded

* Ruscom Logics Limited, Rexdale, Ontario, Canada.

the co-ordinates on IBM cards utilizing an IBM 26 Printing Card Punch*.

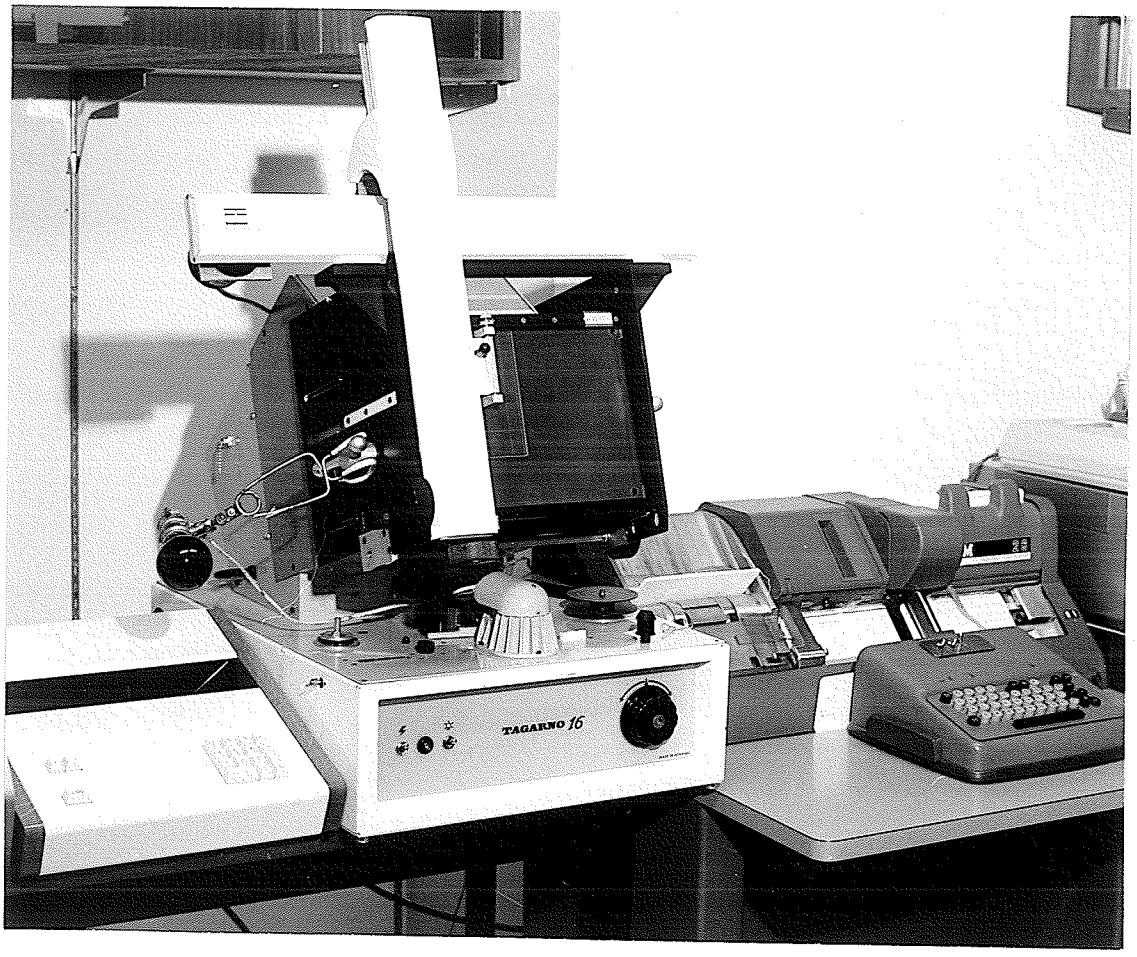

Figure 8. Photograph of Tagarno 16 movie projector and attached Ruscom strip chart digitizer and IBM Printing Card Punch.

* IBM, Don Mills, Ontario, Canada.

The data was subsequently statistically processed by the Computer Department of the Health Sciences Complex, The University of Manitoba.

Co-Ordinate Analysis

The co-ordinate analysis, as developed by Cleall and Chebib (1970), was used as a prelude to the statistical evaluation of the data. The initial analysis consisted of the mathematical calculation of means, standard deviations and standard errors for the individuals within the 5 age groups. The origin for the cephalometric co-ordinate analysis was point 26 (sella) with the direction to point 4 (nasion). In the cinefluorographic analysis the origin point was formed by dropping a perpendicular from the pterygo-maxillary fissure to the palatal plane and the direction was defined by a point formed by a perpendicular extended from A point to the palatal plane, Cleall and Chebib (1971). The utilization of the data following the initial calculations was to subject it to a 3-way mixed analysis of variance involving group, sex and age. A determination of the significant F values was then made. When more than one factor was involved and the F values were significant, these specific factors were submitted to a Duncan significance test. In those involving only one factor the F values were used as determinations of the level of significance.

The measurements that were treated in this manner will be described in three groups; angular, linear vertical and linear horizontal.

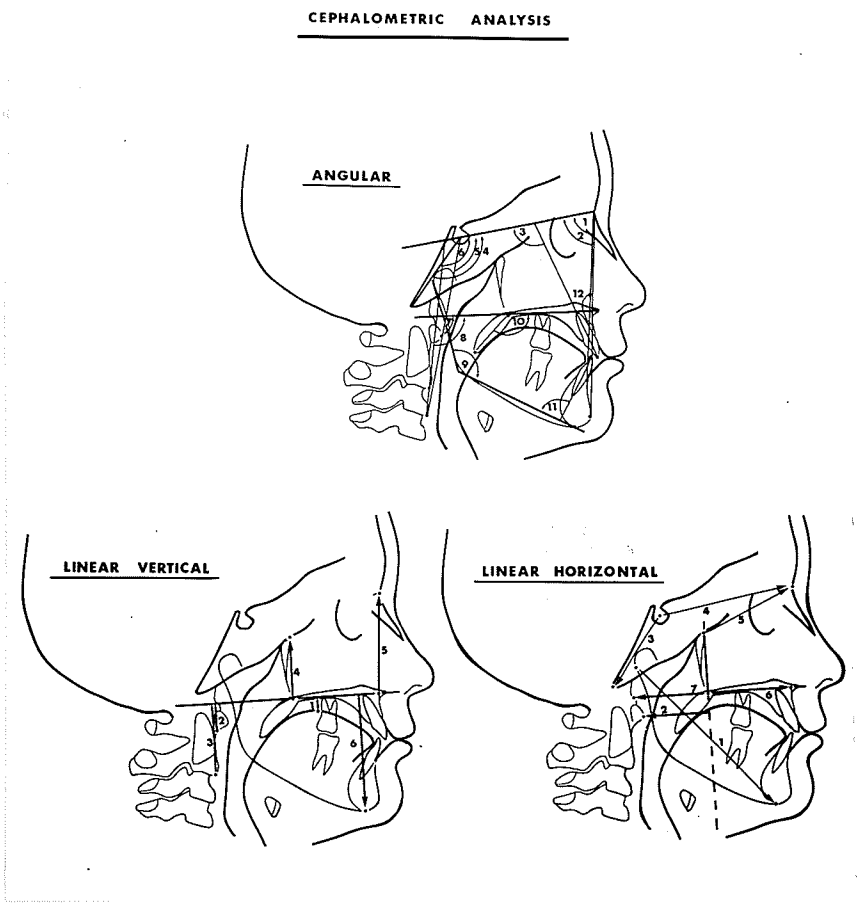

Figure 9. Diagrammatic illustration of the lateral cephalometric co-ordinate analysis which include angular, linear vertical and linear horizontal measurements.

Lateral Cephalometric Angular Measurements

These measurements are illustrated in figure 10.

Angle 1 - The angle formed from sella-nasion to pogonion.

Angle 2 - The angle formed from sella-nasion to B point.

Angle 3 - The angle formed by the intersection of the long axis of upper central incisor to sella-nasion plane.

Angle 4 - The angle formed by basion sella-nasion.

Angle 5 - The angle formed by the intersection of the plane formed by the joining of the inferior anterior border of the first and third cervical vertebrae with the sella-nasion plane.

Angle 6 - The angle formed by the intersection of the plane formed by the joining of the inferior anterior border of the second and third cervical vertebrae with the sella-nasion plane.

Angle 7 - The angle formed by the intersection of the plane formed by the first and third cervical vertebrae with the palatal plane.

Angle 8 - The angle formed by the intersection of the plane formed by the second and third cervical vertebrae with the palatal plane.

Angle 9 - The angle formed by the intersection of the ramus with the body of the mandible.

Angle 10 - The angle formed by the intersection of the soft palate to the palatal plane.

Angle 11 - The angle of the lower incisor to the mandibular plane.

Angle 12 - The angle formed by joining Nasion-A point pogonior (Angle of Convexity)

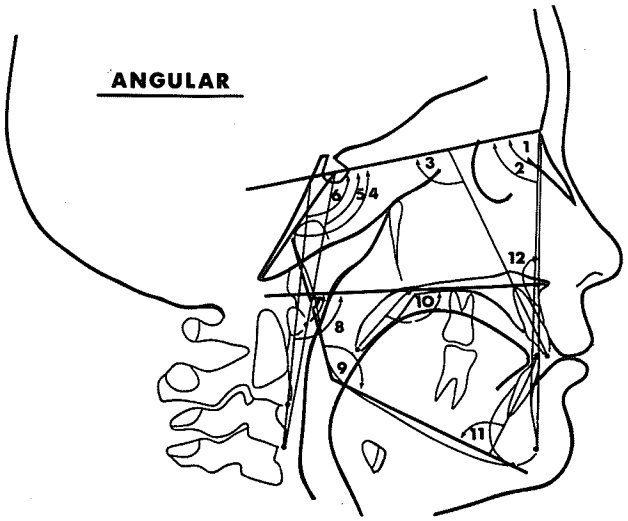

Figure 10. Diagramatic illustration of the lateral cephalometric angular measurements.

Cephalometric Linear Vertical Measurements

These measurements are illustrated by figures 9 and 11.

Linear vert. 1 - The distance from the dorsum of the tongue to the palatal plane.

Linear vert. 2 - The distance from the inferior anterior border of the anterior tubercle of the atlas to the palatal plane.

Linear vert. 3 - The inferior anterior border of the second cervical vertebra to the palatal plane.

Linear vert. 4 - The perpendicular distance from the posterior nasal spine to the anterior cranial base.

Linear vert. 5 - The distance from an extension of point A to nasion.

Linear vert. 6 - The perpendicular distance from the palatal plane to menton.

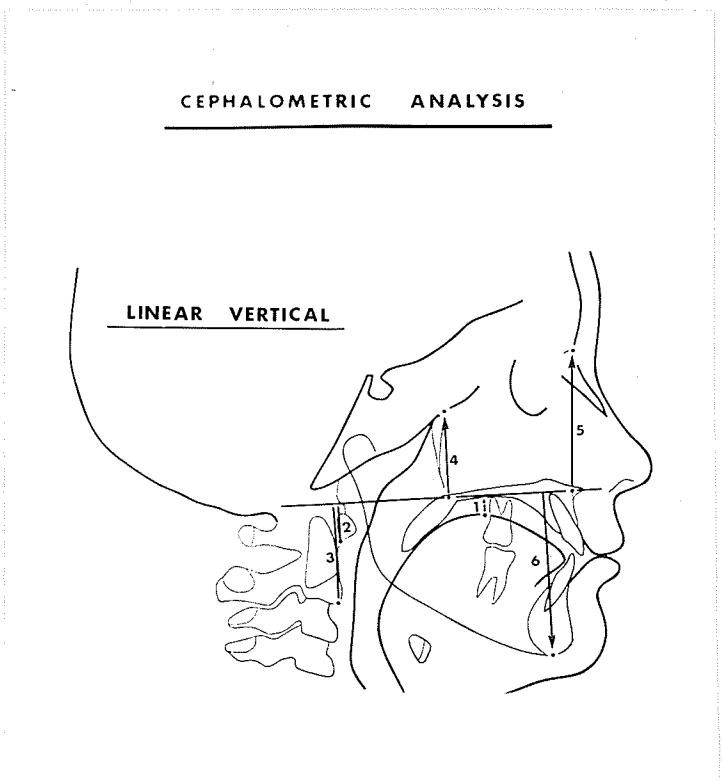

Figure 11. The diagramatic illustration of the lateral cephalometric linear vertical measurements.

Cephalometric Linear Horizontal Measurements

These measurements are illustrated in figures 9 and 12.

Linear Horiz. 1 - This measures the distance from the most posterior position on the condyle to menton.

Linear Horiz. 2 - The horizontal distance from the posterior nasal spine to the anterior tubercle of the atlas.

Linear Horiz. 3 - The distance from basion to sella.

Linear Horiz. 4 - The distance from sella to nasion.

Linear Horiz. 5 - The distance from landmark number 52 to nasion.

Linear Horiz. 6 - The horizontal length of the hard palate measured from posterior nasal spine to a vertical extension of A point.

Linear Horiz. 7 - The horizontal distance from posterior nasal spine to the bony surface of the first cervical vertebra .

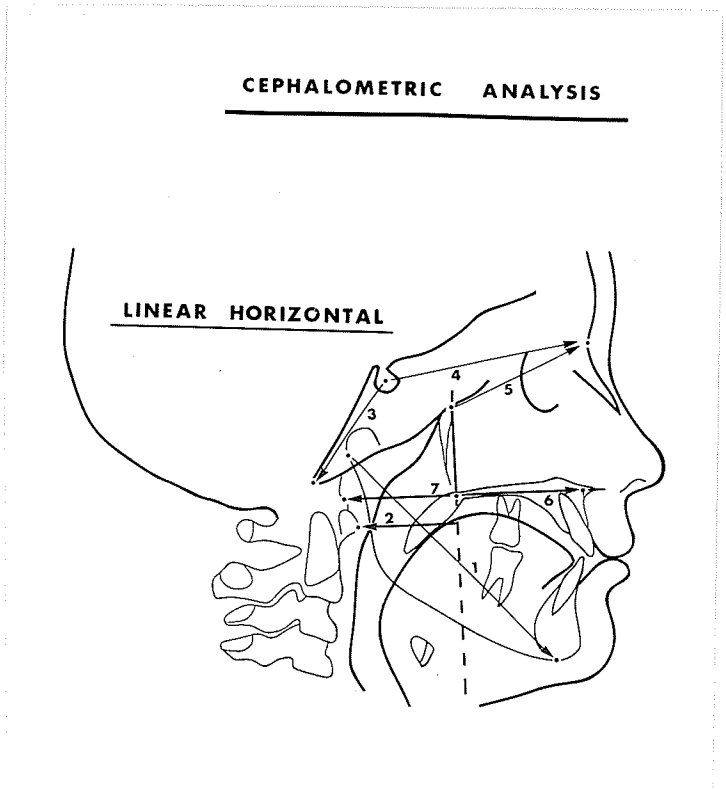

Figure 12. The diagramatic illustration of the lateral cephalometric linear horizontal measurements.

Cinefluorographic Analysis

The Tagarno motion analyzer with its various additional components was used to digitize the selected landmarks.

Five stages were chosen for analysis within the deglutition cycle. These stages were selected on the basis that they represented the different stages of deglutition and also on the basis of ease of identification. The stages selected are similar to those used by Cleall (1965). They are as illustrated in figure 13:

Stage 1 - The rest position preceding deglutition.

Stage 2 - The position when the tongue makes contact with the lingual surface of the upper incisor and the palatal mucosa.

Stage 3 - The position of the tongue when it has reached the junction of the hard and soft palate.

Stage 4 - The position when the hyoid bone is at its most superior anterior position.

Stage 5 - The rest position at the end of deglutition.

These stages are represented separately in figures 14, 15, 16 and 17.

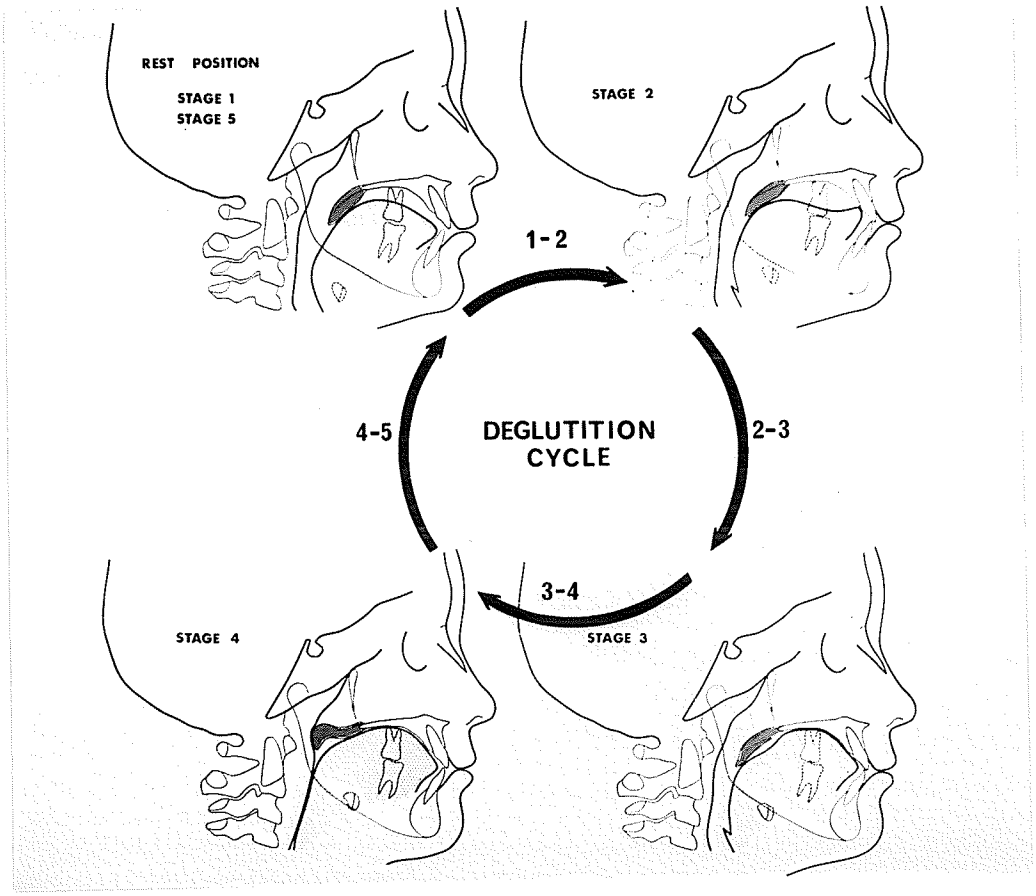

Figure 13. A diagramatic illustration of the 5 stages of deglutition selected for analysis.

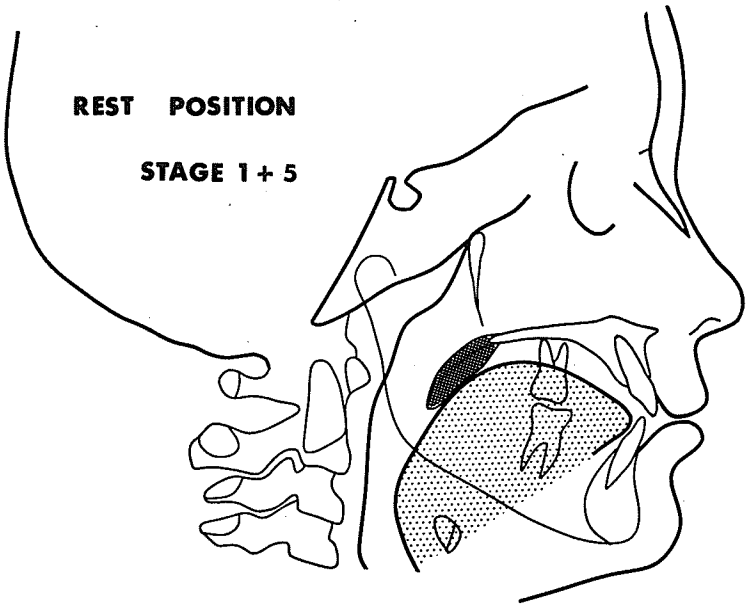

Figure 14. A diagramatic illustration of rest position, stage 1 and 5 of deglutition.

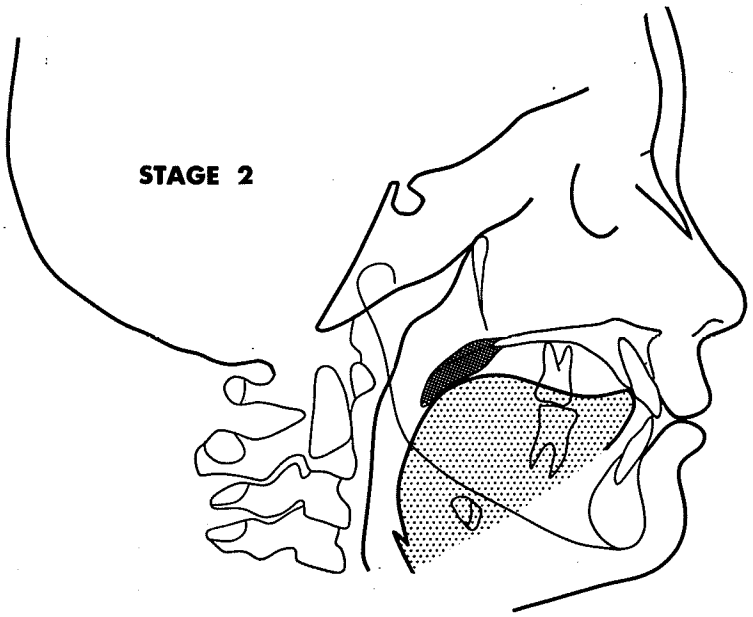

Figure 15. A diagramatic illustration of stage 2 of deglutition.

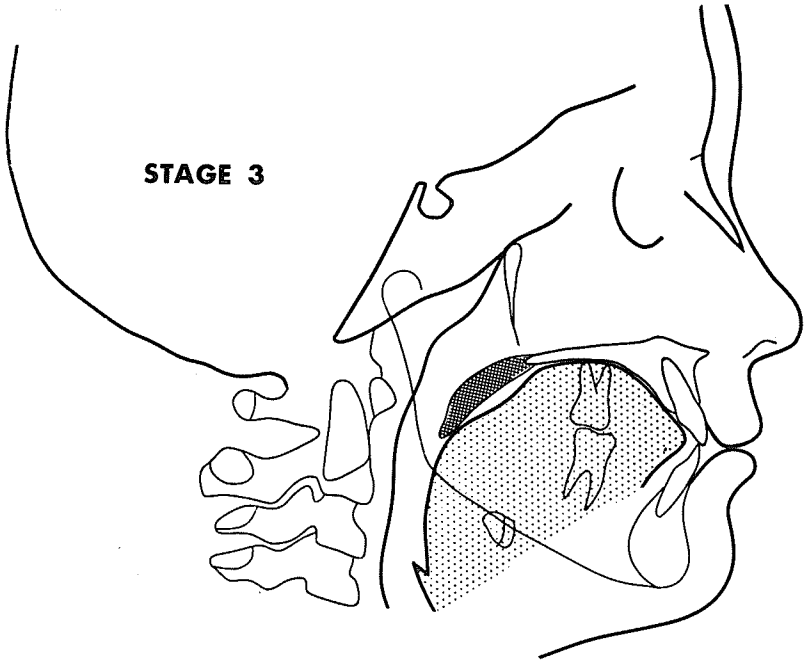

Figure 16. A diagramatic illustration of stage 3 of deglutition.

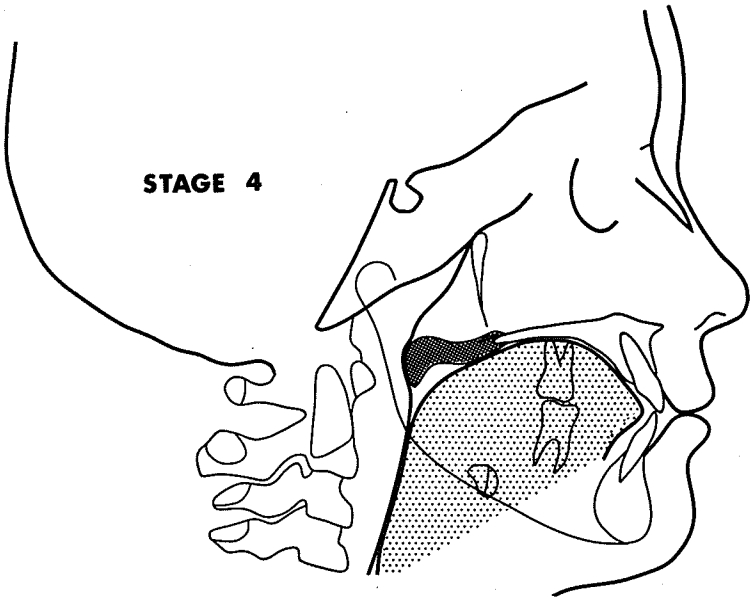

Figure 17. A diagramatic illustration of stage 4 of deglutition.

The template method of transferring from stage to stage was used to insure maximum accuracy, Stone (1971).

Within each stage 3 landmarks were stationary and 13 landmarks were moving points. The location of these landmarks are illustrated in figure 18 and their definitions appear in the Glossary.

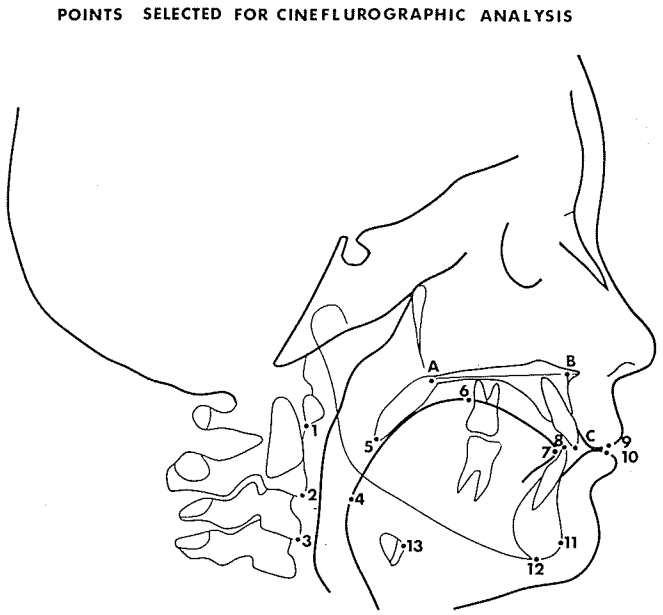

Figure 18. A diagramatic illustration of the 3 stationary points A, B and C and 13 moving points selected for cinefluorographic analysis in each of the stages of deglutition.

A standardized set of angular and linear measurements were used to analyze each stage. These measurements, for ease of presentation, will be divided into angular, linear vertical and linear horizontal measurements as represented in figure 19.

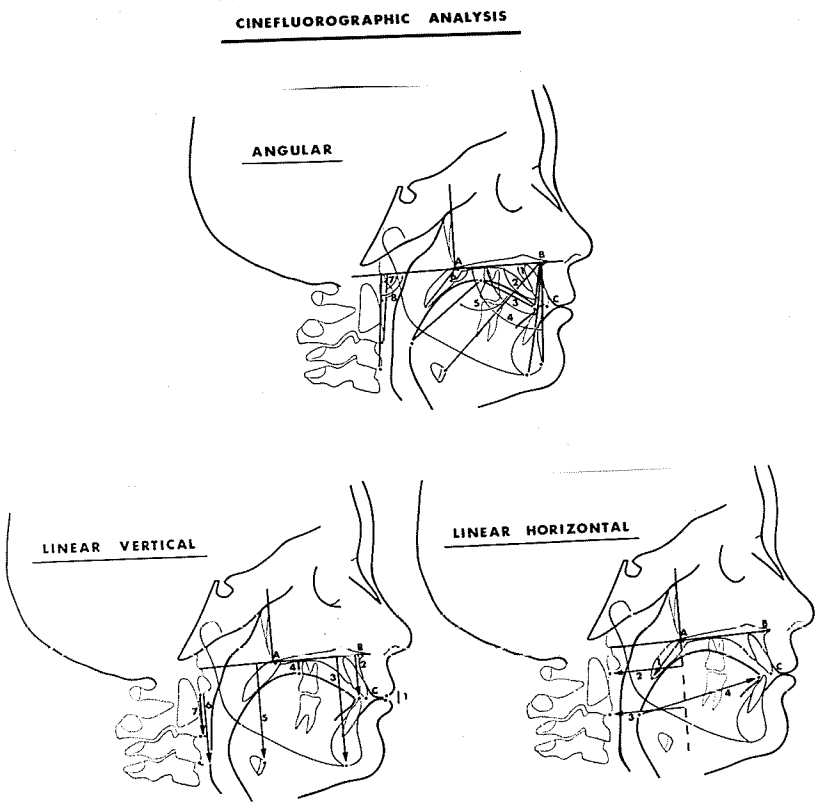

Figure 19. A diagramatic illustration of the cinefluorographic analyses which include angular, linear vertical and linear horizontal measurements.

Cinefluorographic angular measurements as illustrated in figures 19 and 20 are:

Angular 1 - The angle formed by the palatal plane to the hyoid bone with the vertex at B point.

Angular 2 - The angle formed by palatal plane to tongue tip with the vertex at B point.

Angular 3 - The angle formed by palatal plane to menton with the vertex at B point.

Angular 4 - The angle formed by palatal plane to pogonion with the vertex at B point.

Angular 5 - The angle of the dorsum of the tongue. Landmarks 4 - 6 - 8.

Angular 6 - The angle of the soft palate to the palatal plane with the vertex at A point.

Angular 7 - The angle formed by the intersection of the first and second vertebrae with the palatal plane.

Angular 8 - The angle formed by the intersection of the second and third vertebrae with the palatal plane.

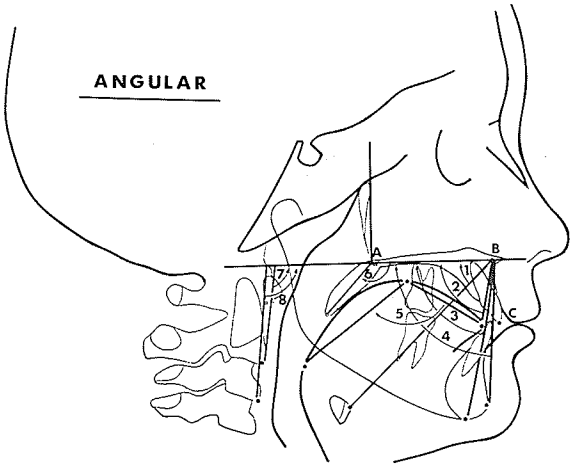

Figure 20. A diagramatic illustration of the cinefluorographic angular measurements.

Cinefluorographic linear vertical measurements as illustrated by figures 19 and 21 are:

Linear vert. 1 - The closest distance between the upper and lower lips.

Linear vert. 2 - The perpendicular distance from the palatal plane to the tip of the lower incisor.

Linear vert. 3 - The perpendicular distance from the palatal plane to menton.

Linear vert. 4 - The perpendicular distance from the palatal plane to the highest point on the dorsum of the tongue.

Linear vert. 5 - The perpendicular distance from the palatal plane to the most anterior superior position on the hyoid bone.

Linear vert. 6 - The distance from the inferior surface of the anterior tubercle of the atlas to the most anterior inferior position on the third cervical vertebra .

Linear vert. 7 - The distance from the inferior surface of the anterior tubercle of the atlas to the most anterior inferior position on the second cervical vertebra .

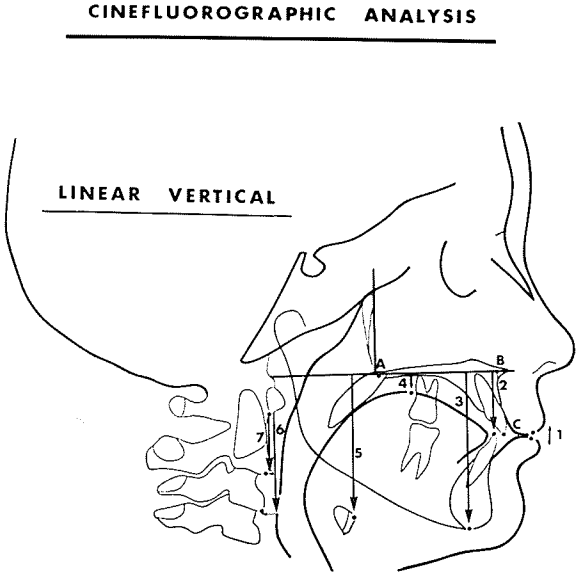

Figure 21. A diagramatic illustration of the cinefluorographic linear vertical measurements.

Cinefluorographic linear horizontal measurements as illustrated by figures 19 and 22 are:

Linear Horiz. 1 - The distance from the posterior nasal spine to the tip of the soft palate.

Linear Horiz. 2 - The horizontal distance from the posterior nasal spine to a point on the inferior surface of the anterior tubercle of the atlas.

Linear Horiz. 3 - The horizontal distance from the posterior nasal spine to the most anterior inferior point on the second cervical vertebra .

Linear Horiz. 4 - The length of the tongue as measured from anterior tongue tip to a point adjacent to the most anterior inferior position on the second cervical vertebra .

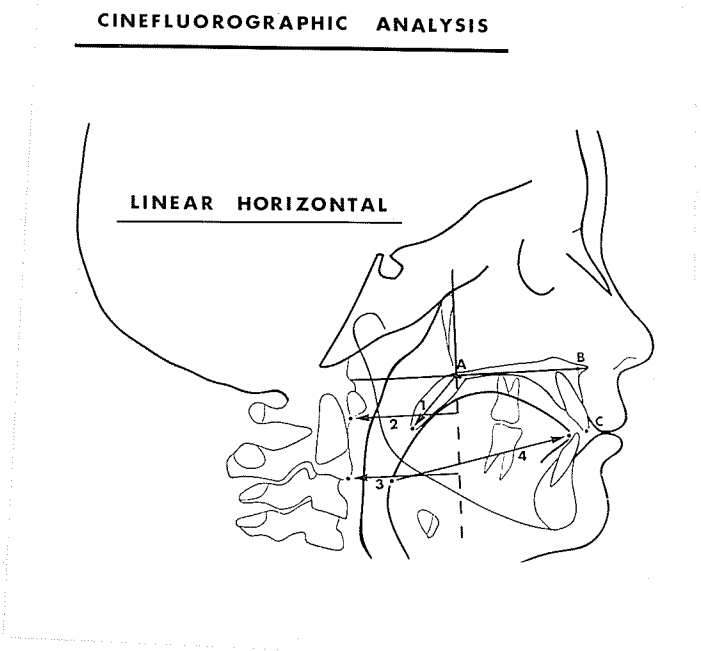

Figure 22. A diagramatic illustration of the cinefluorographic linear horizontal measurements.

RESULTS

CHAPTER IV

RESULTS

The results shall be presented in three sections, as follows:

1. The results from the area analysis as obtained from lateral cephalometric radiographs. (square millimeters)
2. The results of the lateral cephalometric co-ordinate analysis also obtained from lateral cephalometric radiographs. (angles, linear measurements in centimeters)
3. The results from the cinefluorographic analysis obtained from the deglutition cycle.

The presentation and interpretation of the data has been aided by the use of diagrams, graphs and tables in either complete or abridged forms. The tabulated statistical results are presented in the Appendix.

I. CEPHALOMETRIC AREA ANALYSIS

Seven separate area measurements were made on each of the forty trisomy 21 and forty normal individuals, these measurements will be dealt with separately.

Area 1 - Nasal Area

The results of this analysis indicated that the nasal area was smaller in the trisomy 21 subjects in all age groups, however, this discrepancy between the groups became more pronounced with age. This is illustrated in figure 23 and represented in tabular form in table 11.

It should be noted that the increase in size, in both groups, occurred primarily before the age of 14 years and that there was little increase thereafter.

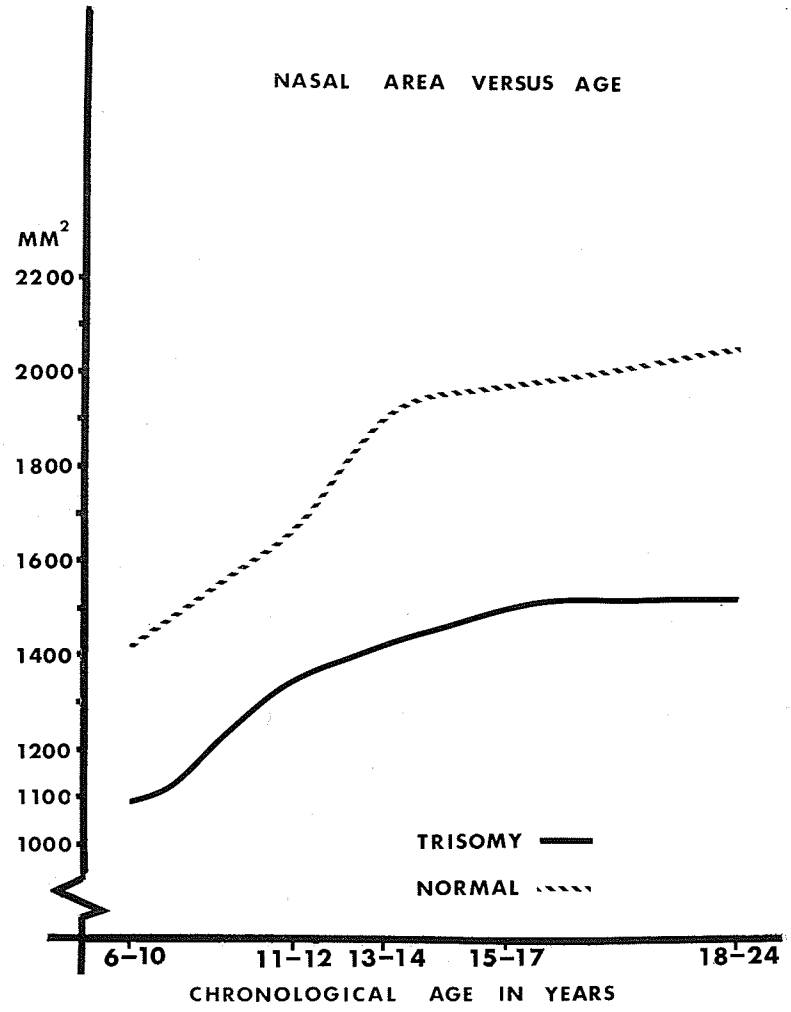

Figure 23. A graphic illustration of nasal area versus groups age.

TABLE II

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES BETWEEN THE NASAL AREAS OF TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6 - 10 yrs	1153.7	87.19	1501.7	92.23	3.71
11- 12 yrs	1368.6	42.12	1677.4	82.50	3.74 *
13- 14 yrs	1467.4	58.33	1949.2	58.33	8.25 **
15- 17 yrs	1575.7	65.22	2025.0	65.22	6.88 **
18- 24 yrs	1535.6	51.16	2055.5	51.16	10.16 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

Area 2 - Nasopharynx Area

The results of these measurements indicated that the soft tissue involution was less in the trisomy 21 subjects and this discrepancy increased with age. It may be noted from the graph (figure 24) that after age 12 years the trisomy nasopharynx changed very little, whereas in the normal group the nasopharynx increased rapidly up to 14 years of age. The growth after this age was slow but continuous to adulthood. This is represented in tabular form in table III.

TABLE III

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF NASOPHARYNX AREA BETWEEN THE TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	177.7	49.67	204.5	49.67	0.53
11-12 yrs	275.0	44.43	269.2	44.43	0.47
13-14 yrs	290.1	31.41	446.4	31.41	4.97 **
15-17 yrs	276.2	35.12	384.3	35.12	3.27 *
18-24 yrs	323.7	27.55	491.3	27.55	6.08 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

Area 3 - Bony Nasopharynx Area

The measurement of the bony nasopharynx suggested that there was no significant difference in this area between the trisomy 21 and normal subjects. This is illustrated in tabular form in table IV.

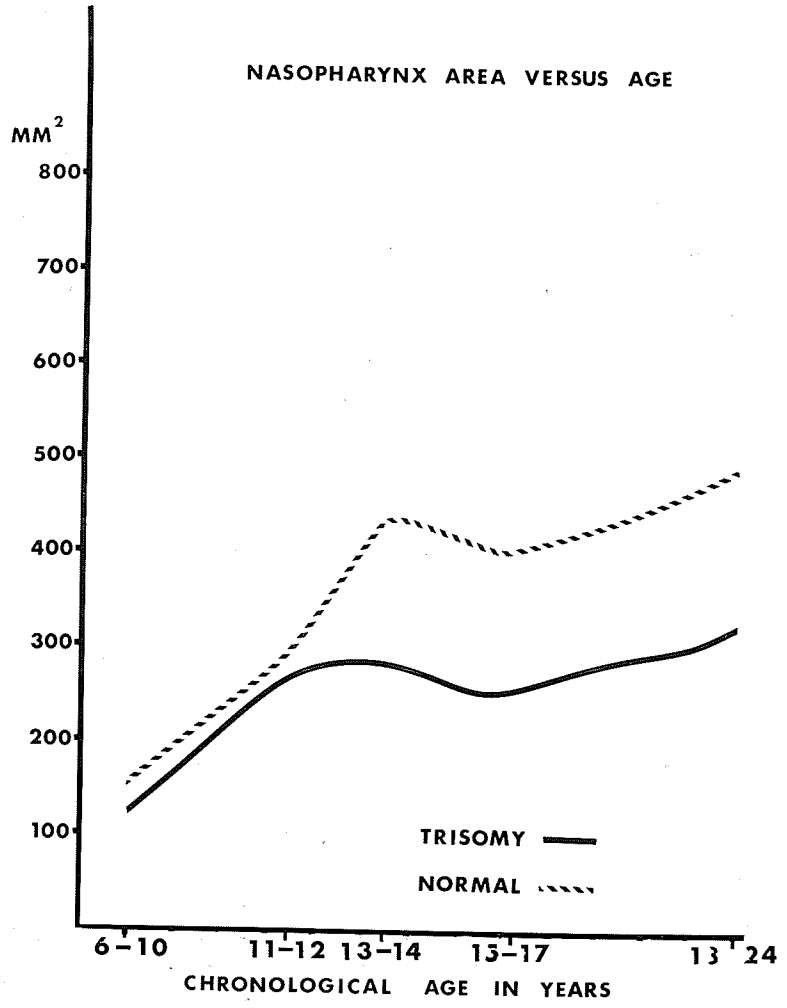

Figure 24. A graphic illustration of the nasopharynx area as it relates to the five age groups.

TABLE IV

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF THE BONY NASOPHARYNX AREA BETWEEN THE TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	581.7	93.62	623.2	20.65	0.11
11-12 yrs	602.0	51.44	733.0	36.50	0.41
13-14 yrs	759.2	42.94	798.8	45.70	0.17
15-17 yrs	1003.1	75.20	828.5	38.90	0.32
18-24 yrs	1203.2	45.00	865.2	25.01	1.72

Areas 4 and 5 - Oropharynx Area and Oral Cavity Area

Both of these areas illustrated a large degree of variability and as a result none of the areas was significantly different in the younger age groups. The adult age groups showed a significant discrepancy at the 0.05 level of confidence, due to a larger area in the trisomy group. The trisomy 21 oropharynx and the oropharynx plus oral cavity area had increased considerably more than those of the normal group by this age, as illustrated in figures 25 and 26 respectively. The graphs indicate that the area discrepancy began between 11 and 14 years of age and that when the areas of the oropharynx and oral cavity are combined the difference became much more discrete. These findings are also represented in tabular form in tables V and VI.

TABLE V

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF THE OROPHARYNX AREAS BETWEEN THE TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	494.5	44.06	416.0	51.58	0.29
11-12 yrs	516.4	28.85	538.4	112.90	0.09
13-14 yrs	604.5	61.80	490.4	52.81	0.377
15-17 yrs	636.3	53.40	540.7	63.16	0.36
18-24 yrs	967.8	75.20	715.0	45.70	2.89 *

* Significant at the 0.05 level of confidence.

TABLE VI

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF THE ORAL CAVITY AREA BETWEEN THE TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	449.5	55.93	278.5	77.58	0.56
11-12 yrs	305.6	81.05	352.4	79.16	0.17
13-14 yrs	368.7	33.91	503.8	93.88	0.70
15-17 yrs	602.7	51.19	411.7	55.90	0.89
18-24 yrs	800.6	80.80	387.8	53.04	2.87 *

* Significant at the 0.05 level of confidence.

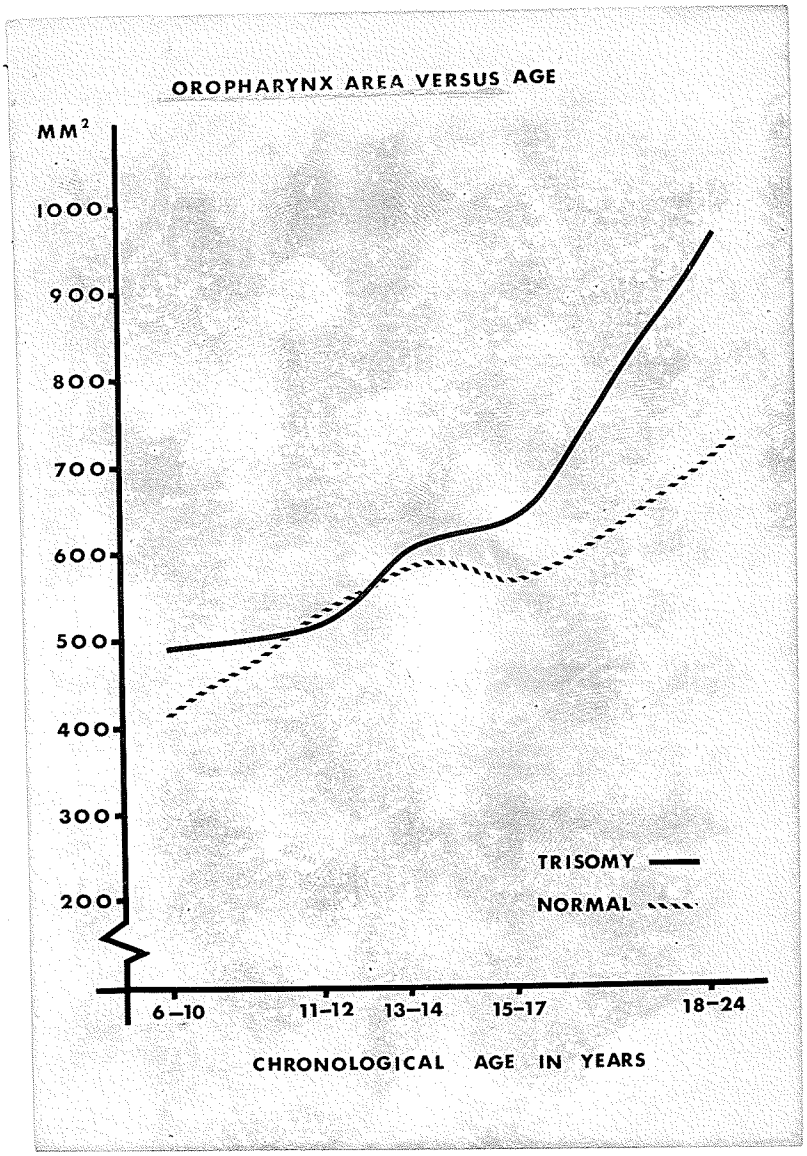

Figure 25. A graphic illustration of the oropharynx area as it relates to the five age groups.

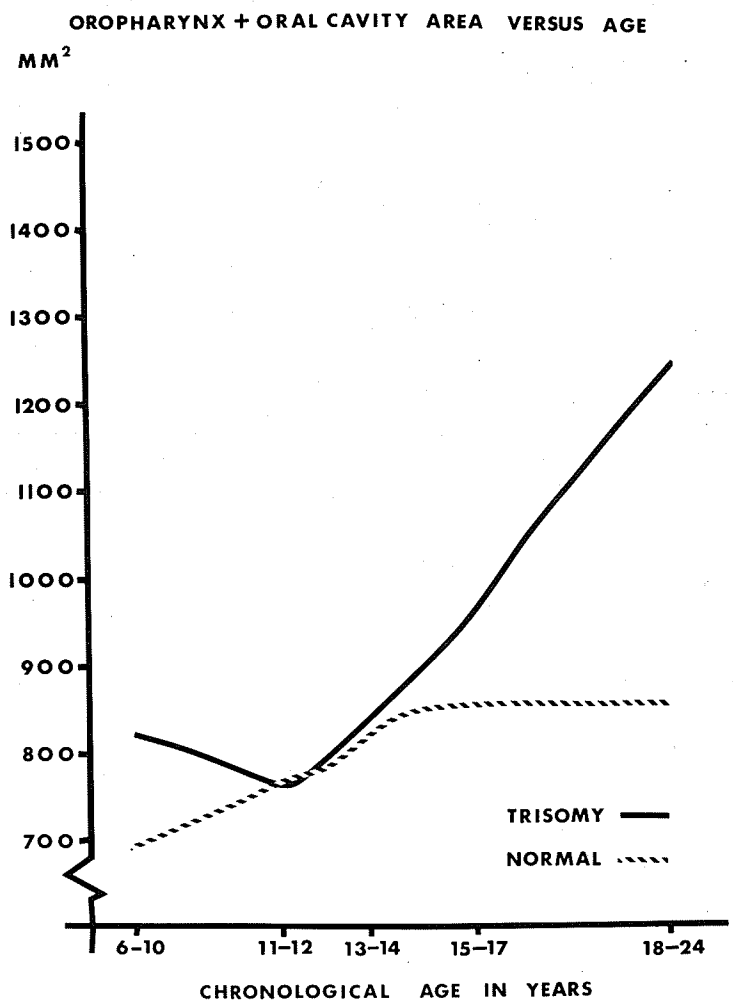

Figure 26. A graphic illustration of the oropharynx area plus oral cavity as they relate to the five age groups.

Area 6 - Soft Palate Area

The measurements of this area indicated that the soft palate was significantly larger in the trisomy 21 group at all age levels. This was significant at the 0.05 level of confidence in the first two age groups and at the 0.01 level of confidence thereafter. The data suggests, as illustrated in figure 27, that there was little change in the trisomy 21 group throughout the experimental period. The normal subjects, however, showed an appreciable increase in size up to age 14 years with little growth thereafter. These results are in tabular form in table VII.

TABLE VII

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF THE SOFT PALATE AREA BETWEEN THE TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	280.0	20.12	173.0	8.39	4.57 *
11-12 yrs	277.0	25.04	184.0	11.18	4.44 *
13-14 yrs	277.9	19.82	237.0	7.68	3.71 **
15-17 yrs	316.8	22.51	334.0	11.11	4.95 **
18-24 yrs	309.5	17.17	228.0	11.44	6.24 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

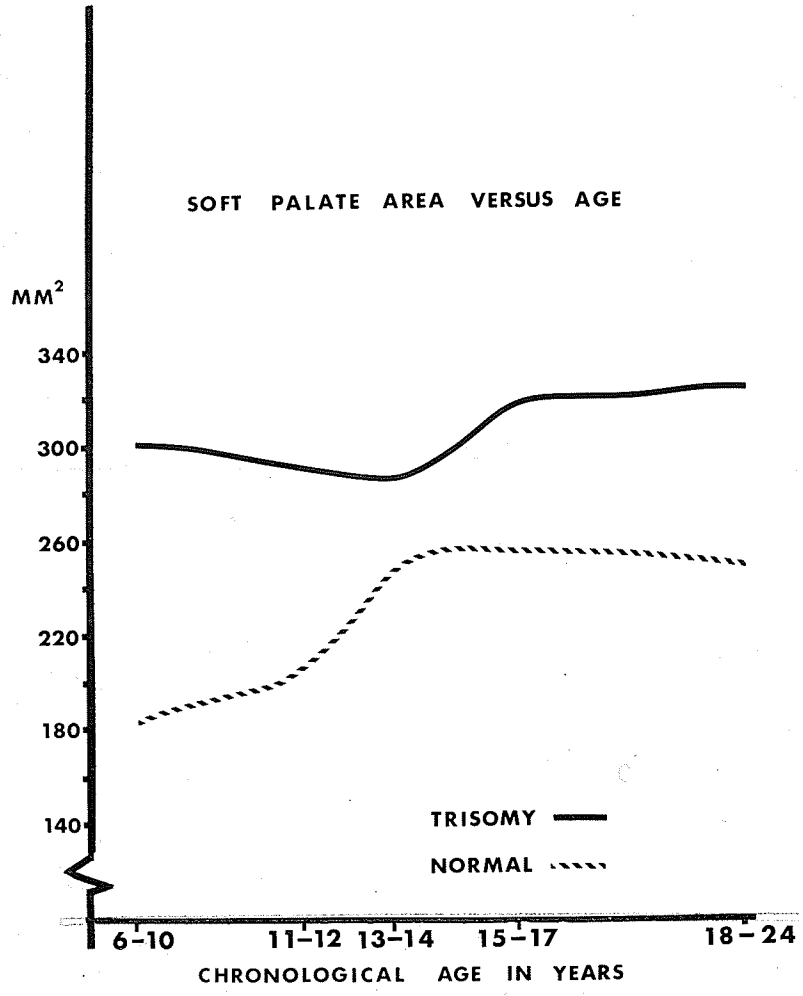

Figure 27. A graphic illustration of the soft palate area as it relates to the five age groups.

Area 7 - Tongue area

There was no significant difference in tongue area between the two groups except in the 15-17 year age group. Here the normal group showed larger tongues significant at the 0.01% level of confidence. In the normal group, the tongue size increased to 13-14 years and did not increase appreciably thereafter. In the trisomy 21 group, the tongue size increased rapidly to age 11-12 years and then increased slowly but continuously to adulthood. These finding are illustrated graphically in figure 28 and in tabular form in table VIII.

TABLE VIII

MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES OF THE TONGUE AREAS BETWEEN THE TRISOMY AND NORMAL SUBJECTS

	Trisomy		Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	1434.7	98.46	1654.2	87.34	1.54
11-12 yrs	1946.6	155.91	1827.0	101.59	0.94
13-14 yrs	2065.4	67.26	2300.0	89.31	2.62
15-17 yrs	1993.7	46.58	2464.0	99.85	4.77 *
18-24 yrs	2211.6	65.95	2240.5	78.33	0.36

* Significant at the 0.01 level of confidence.

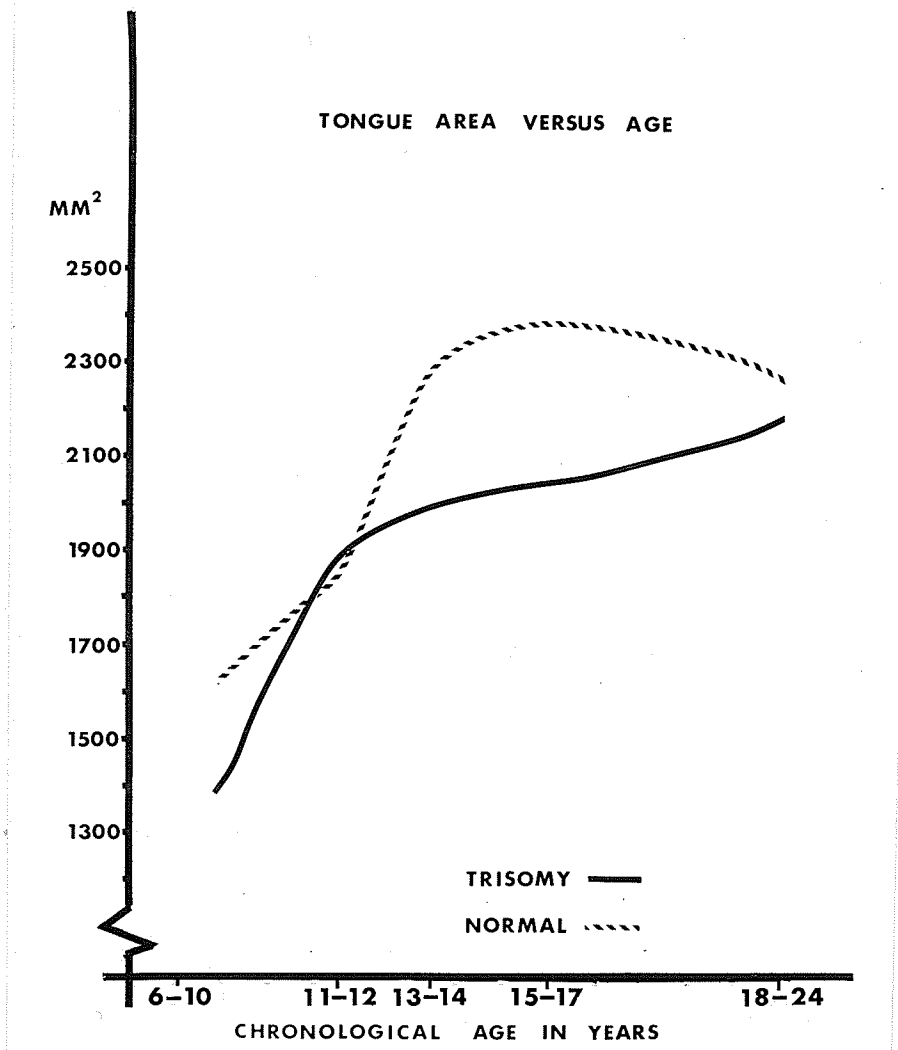

Figure 28. A graphic illustration of the tongue area as it relates to the five age groups.

In evaluating the measurements it was found that the trisomy 21 group was consistently smaller in all areas except the soft palate, oropharynx and oral cavity areas. Of these measurements, only the soft palate area was significantly larger than the normals, at all age levels. The oral cavity and oropharynx areas were only significally larger in the adult age groups.

The nasal cavity was consistently larger in the normal subjects at all age levels, while the nasopharynx established a discrepancy only after the 11-12 year age group. After this age, the discrepancy between the two groups became progressively greater as a result of rapid soft tissue involution in the normals and a retarded involution in the trisomy group. These results are consistent in both the group analysis, (table IX) and in the group age analyses.

TABLE IX

GROUP DATA FOR AREA ANALYSES, MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY 21 AND NORMAL GROUPS

	Trisomy		Normal			
Variable	Mean	Standard Error	Mean	Standard Error	F Value Significance	
Area 1 Nasal	1467.5	35.96	1920.2	42.26	108.41	**
Area 2 Nasopharynx	285.1	14.79	405.6	21.53	22.39	**
Area 3 Nasopharynx	865.4	48.90	800.5	19.80	0.01	
Area 4 Oropharynx	706.9	85.48	604.6	30.19	0.46	
Area 5 Oral Cavity	556.0	124.83	406.2	33.82	0.67	
Area 6 Soft Palate	296.0	7.39	220.4	6.02	51.75	**
Area 7 Tongue	2020.8	55.39	2189.7	65.37	6.39	*

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

II. CEPHALOMETRIC CO-ORDINATE ANALYSIS

The primary purpose of this cephalometric analysis was to establish a morphological basis for the preceding area analyses and for the functional analyses. Previous studies by Ghiz (1969), Frostad (1970), Jensen (1971) and Nevile (1973) have extensively investigated many craniofacial parameters relating to this trisomy group; thus the cephalometric phenotypic characteristics are well established. However, in view of the fact that the sample used by the author was smaller than the previously mentioned studies, a cephalometric analysis was found to be imperative, not only to insure that the sample was representative, but to extend the analyses into the nasal-oral region.

The results of the cephalometric analysis shall be presented as morphological characteristics of various structures within the craniofacial complex.

Cranial Base

The angle of the cranial base, as measured by basion-sella-nasion, was found to be more obtuse in the trisomy 21 subjects at all age levels. This shown in tabular form in table X and table XI and is illustrated graphically in figure 29. These findings are consistent with those of Kisling (1966) and Ghiz (1969).

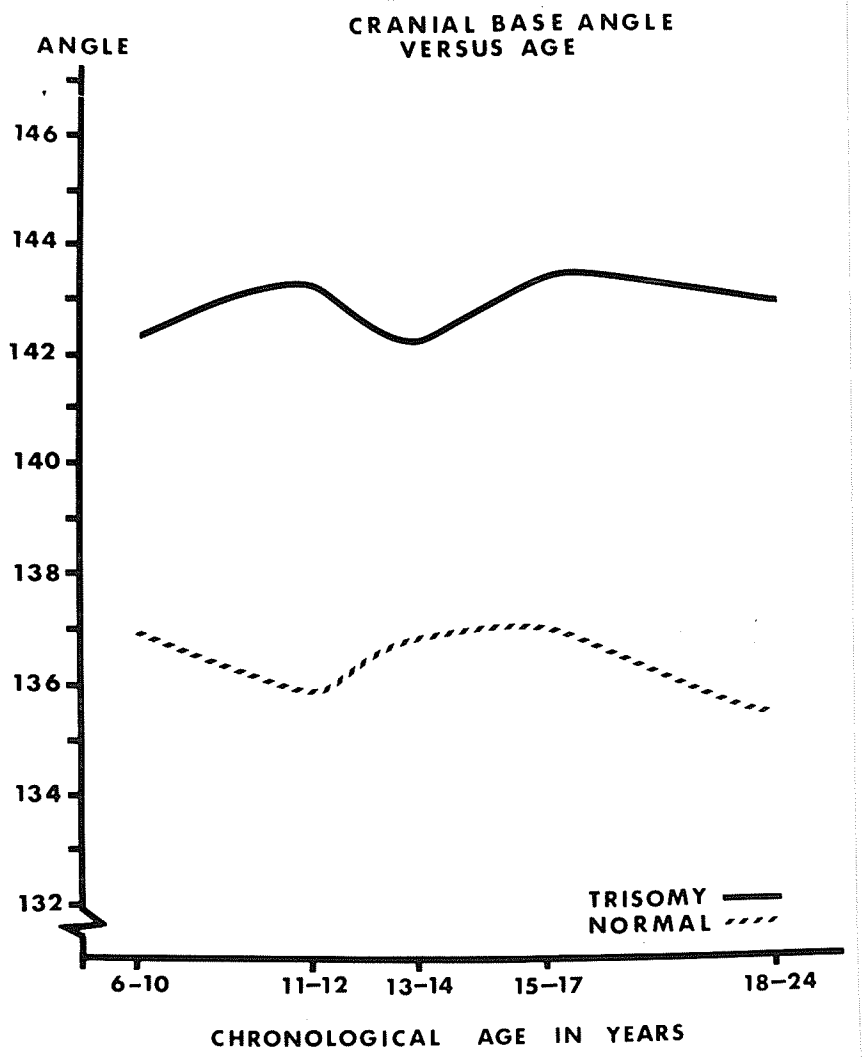

Figure 29. A graphic illustration of the cranial base angle as compared to group ages in the trisomy and normal groups.

TABLE X

MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES OF THE CEPHALOMETRIC MEASUREMENTS BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal			
Variable	Mean	Standard Error	Mean	Standard Error	F Value	Significance
Angular						
Lower 1 to Md. Plane	95.84	0.962	95.90	1.239	0.83	
Cranial Base Angle	142.800	0.764	136.700	0.921	26.26	**
Angle of Mandible	126.70	1.033	129.600	1.216	8.75	**
SN PO Angle	82.16	0.653	80.52	0.470	5.77	*
Angle of Convexity	180.29	1.072	172.87	1.104	42.08	**
SNA	81.06	0.677	82.00	0.601	30.90	**
SNB	81.42	0.649	72.24	0.633	8.56	**
Upper 1 To SN	110.09	0.899	105.40	1.101	10.78	**
Linear Palatal Length 24-29	4.085	0.046	4.578	0.067	42.24	**
Ant. Cranial Base 26-4	6.05	0.050	6.76	0.064	85.66	**
Post Cranial Base 26-27	3.81	0.040	4.00	0.054	3.17	
Mand. Length 49-15	10.01	0.274	10.552	0.151	3.10	

TABLE X (continued)

	Trisomy		Normal			
Variable	Mean	Standard Error	Mean	Standard Error	F Value Significance	
Nasopharynx Width	3.268	0.052	3.018	0.056	9.05	**
Nasopharynx Height	2.52	0.059	2.845	0.064	20.26	**
Nasal Length 52-4	4.50	0.072	4.99	0.168	9.32	**
Ant. Nasal Ht. 59-4	4.23	0.074	4.836	0.082	40.31	**
Palatal Plane to SN Angle	7.68	0.632	5.08	0.893	3.66	

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

TABLE XI

CRANIAL BASE ANGLE (BASION-SELLA-NASION), MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

Trisomy			Normal		
Age Group	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	143.39	3.76	137.39	2.14	3.24
11-12 yrs	143.24	2.94	135.62	2.91	3.40
13-14 yrs	142.071	1.11	136.86	2.06	3.29 *
15-17 yrs	143.49	1.44	137.78	1.89	3.22 *
18-24 yrs	143.12	1.45	135.42	1.64	5.54 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

The length of the anterior cranial base, as measured from sella to nasion, was significantly smaller ($p < 0.01$) in the trisomy 21 group. A similar finding was obtained on a group-age basis, however, the level of significance was slightly less. This is illustrated in tabular form in table XII in Appendix A. The posterior cranial base was found to be less affected which was consistent with the findings of Ghizoni (1969).

Nasal and Nasopharynx

The measurements of the anterior nasal height, nasal length and the posterior nasal height concur with the findings of the area analysis. The group analysis demonstrated that these parameters were significantly smaller in the trisomy 21 subjects ($p < 0.01$). This is illustrated in table X. The group-age analysis suggested that there was considerable variation as the differences became greater with increased age (see Appendix, tables XIII, XIV and XV). The naso-pharynx width, as measured from the posterior nasal spine to the anterior tubercle of the atlas, tended to be larger in the trisomy 21 as compared to the normal subjects. This is illustrated in the Appendix, table XVI.

Palatal Analyses

The length of the palate, as measured from posterior nasal spine to an extension of A point to the palatal plane (Cleall and Chebib, 1971) indicated that the palatal length was significantly smaller in the trisomy 21 group ($p < 0.01$) This is illustrated in table X. The group-age analysis indicated that the trisomy palate was significantly shorter in the adult age groups. This is illustrated graphically in figure 30 and presented in tabular form in table XVII. The upper incisor to the sella nasion plane angle indicated that the incisors were significantly more proclined in the trisomy 21 group ($p < 0.01$), table X. The group-age analysis of this same measurement is illustrated in figure 31 and is presented in tabular form in table XVIII of the Appendix. The antero-posterior position of the maxilla was more retruded in the trisomy 21 group (see table X), however, the mean difference was only 1^{o} and this value falls well within the range of normal variation. This can in part be attributed to the hypoplasia of the anterior cranial base which, in effect, offsets any hypoplasia which may be present in the maxilla.

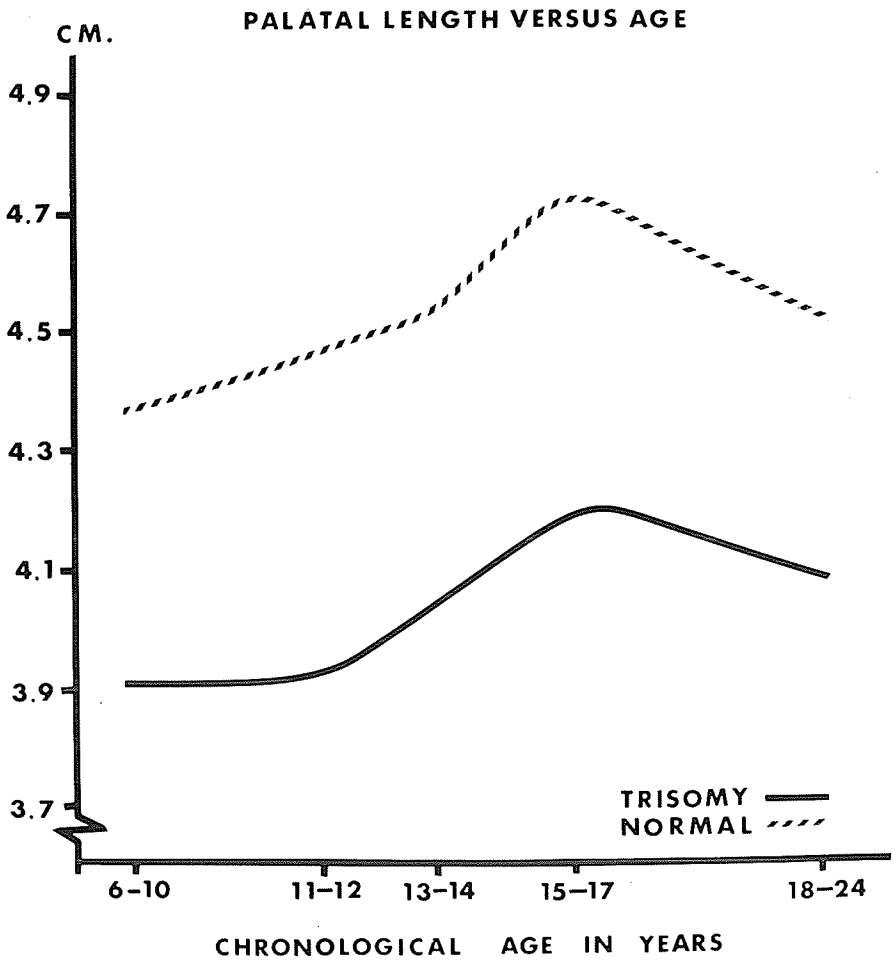

Figure 30. A graphic illustration of the palate length as compared to the group ages in the trisomy and normal groups.

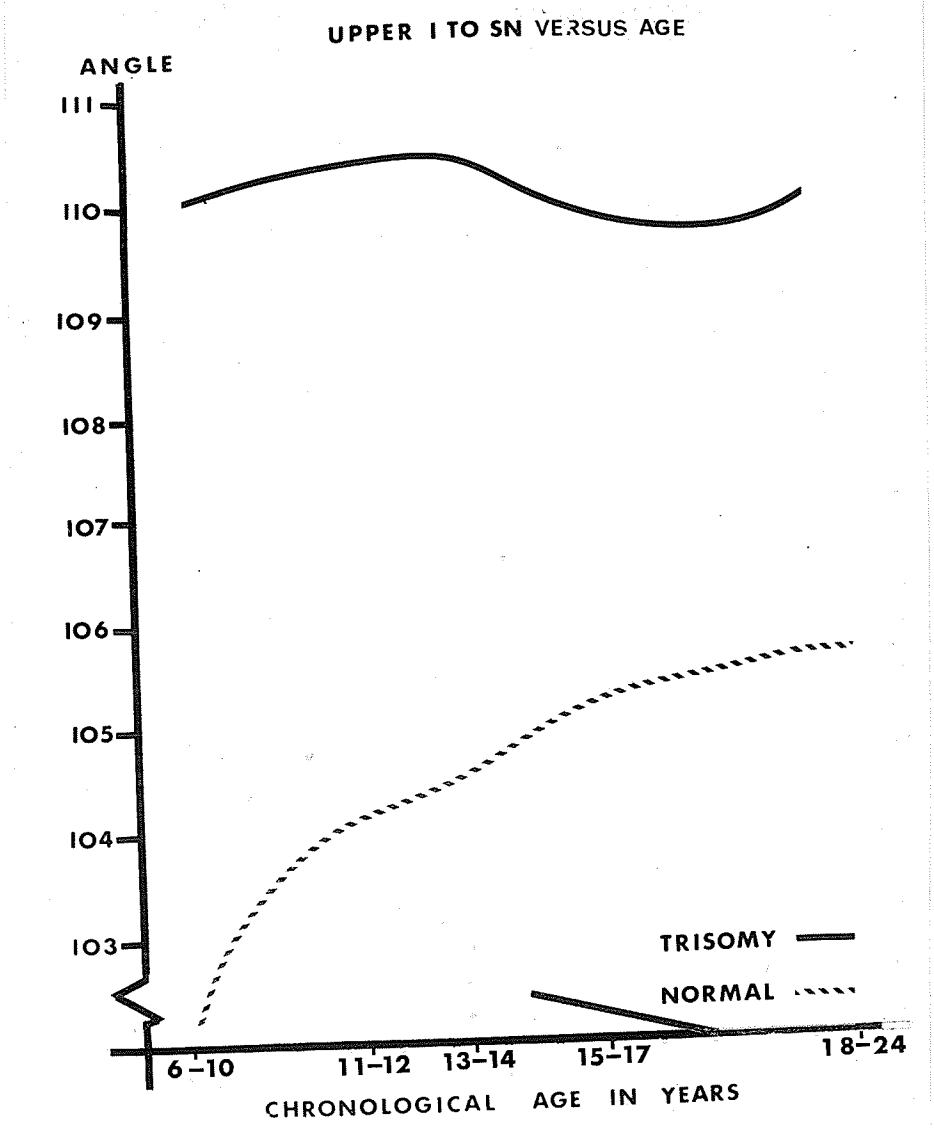

Figure 31. A graphic illustration of upper incisor to the SN plane as compared to the group ages in the trisomy and normal groups.

Mandibular Analyses

The data related to mandibular length indicated that there was no significant difference between the trisomy 21 and the normal groups (table X and figure 32). This agreed with the results obtained by Nevile (1973). The angle of the ramus to the body of the mandible was significantly more acute ($p < 0.01$) in the trisomy 21 group (table X).

The measurements related to the antero-posterior positions of the cranial base, maxilla and mandible indicated that these relationships were significantly different in the trisomy 21 group as compared to the normal sample. Sella-nasion-B point in the trisomy 21 group was significantly smaller at the 0.01 level of confidence (table X). The group-age analyses showed that this discrepancy between groups was primarily in the earlier age groups, as illustrated in the Appendix, table XIX. The angle of convexity was signifcantly greater in the trisomy 21 group ($p < 0.01$), table X. The group-age comparison indicated that the angle of convexity of younger age groups tended to be straighter or more prognathic than that of the older age group. This difference was significant ($p < 0.05$) up to the 13-14 age group. This measurement is illustrated in figure 33 and table XX. The 'lower incisor to mandibular plane measurement' suggested that there was no significant difference in its angulation between the two groups (table X).

The findings with regard to sella-nasion-pogonion angle concurred with previous findings, this angle being significantly ($p < 0.05$) more prognathic in the trisomy 21 group (table IX). The group-age analysis of this parameter again suggested more prognathism in the younger age groups (table XXI of the Appendix).

The results obtained from the cephalometric analyses were generally consistent with the findings of the preceding investigators: Kisling (1966), Ghiz (1969), Frostad (1969) and Nevile (1973). This indicated that the sample used was indeed representative of the trisomy 21 population. The cephalometric measurements made in the oral-nasal region also tended to confirm the results obtained in the lateral cephalometric area analysis.

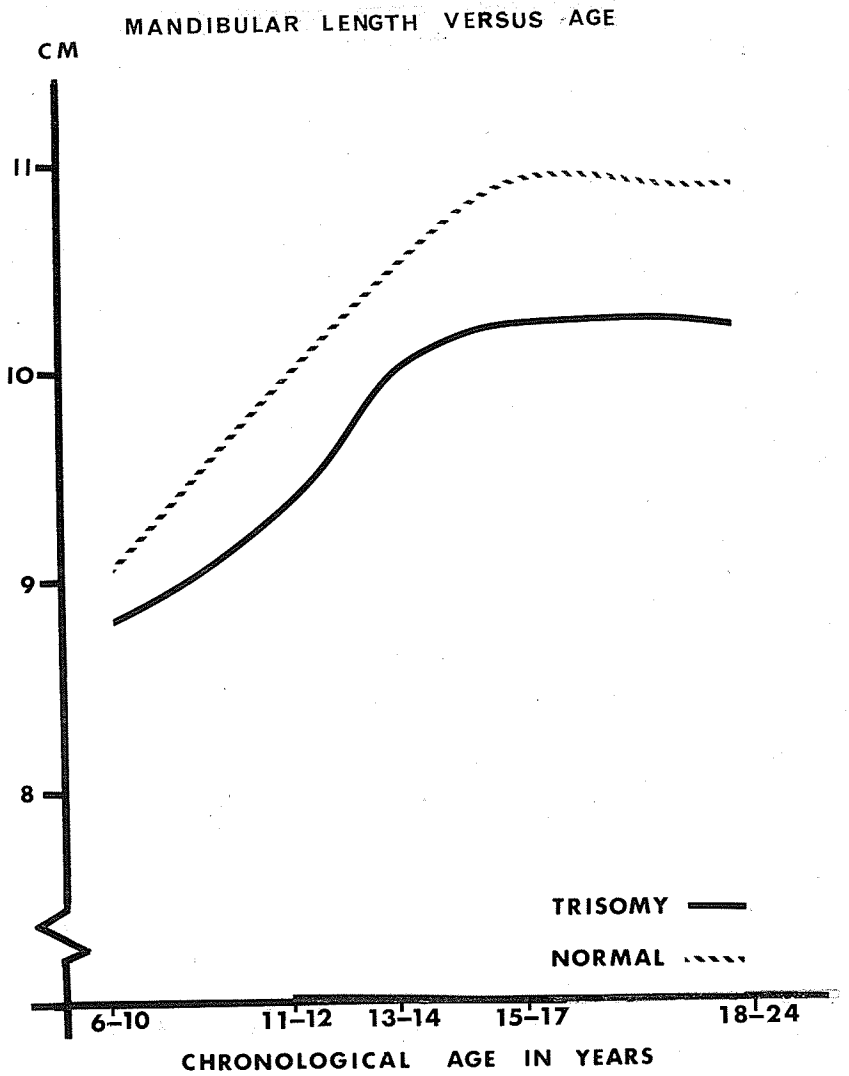

Figure 32. A graphic illustration of the mandibular length as it compares to the group ages in the trisomy and normal groups.

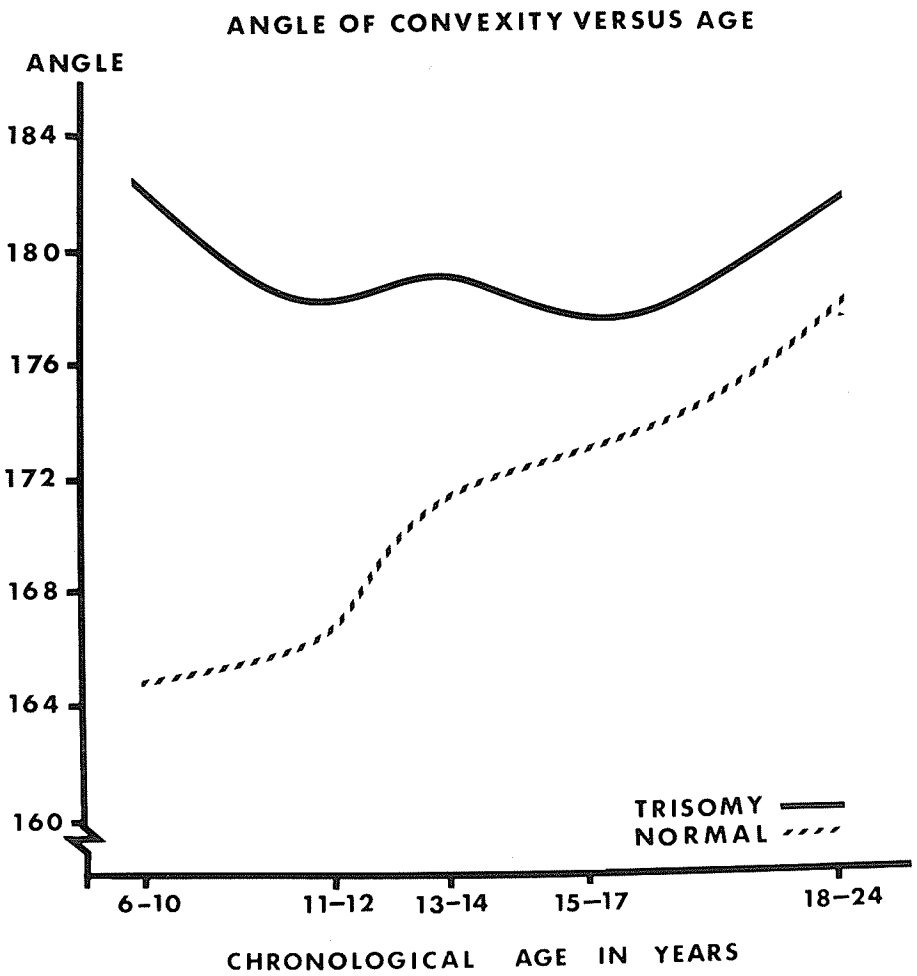

Figure 33. A graphic illustration of the angle of convexity as it compares to the group ages in the trisomy and normal groups.

Cinefluorographic Analyses

The results of the cinefluorographic analysis will be described as they pertain to the function of deglutition. Special emphasis will be placed on the functional analysis of the tongue posture and position, hyoid position and the vertebral size and its angular alterations in respect to the trisomy 21 and normal groups.

1. Tongue

The tongue contour was determined by the angle formed from points selected on the dorsum of the tongue at the various stages of deglutition. This angle was established from the following points: the tip of the tongue, the dorsum of the tongue and the position of the tongue in relation to the second cervical vertebra . The data showed no significant difference between the trisomy and normal groups. This would suggest that neither the contour nor shape of the tongue were appreciably different. This is illustrated in tabular form in table XXII of the Appendix.

The position of the tongue at the various stages of deglutition was determined by two measurements: the angle formed by the tongue tip to the palatal plane, and the vertical distance from the highest point on the dorsum of the tongue to the palatal plane.

The angle formed by the tongue tip to the palatal plane clearly indicated that the tongue was more protruded in the trisomy 21 group; this finding was significant at the 0.01 level of confidence in all stages of deglutition. This is illustrated graphically in figure 34 and in tabular form in table XXIII.

TABLE XXIII

TONGUE TIP TO PALATAL PLANE ANGLE: MEANS, STANDARD ERRORS AND SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Deglutition	Mean	Standard Error	Mean	Standard Error	F Value Difference
Stage 1	88.46	1.592	81.50	1.452	23.86 **
Stage 2	90.94	1.495	81.55	1.013	35.49 **
Stage 3	92.32	1.346	83.29	0.946	34.50 **
Stage 4	93.54	1.345	82.71	0.927	43.53 **
Stage 5	94.89	1.185	81.00	1.087	16.75 **

** Significant at the 0.01 level of confidence.

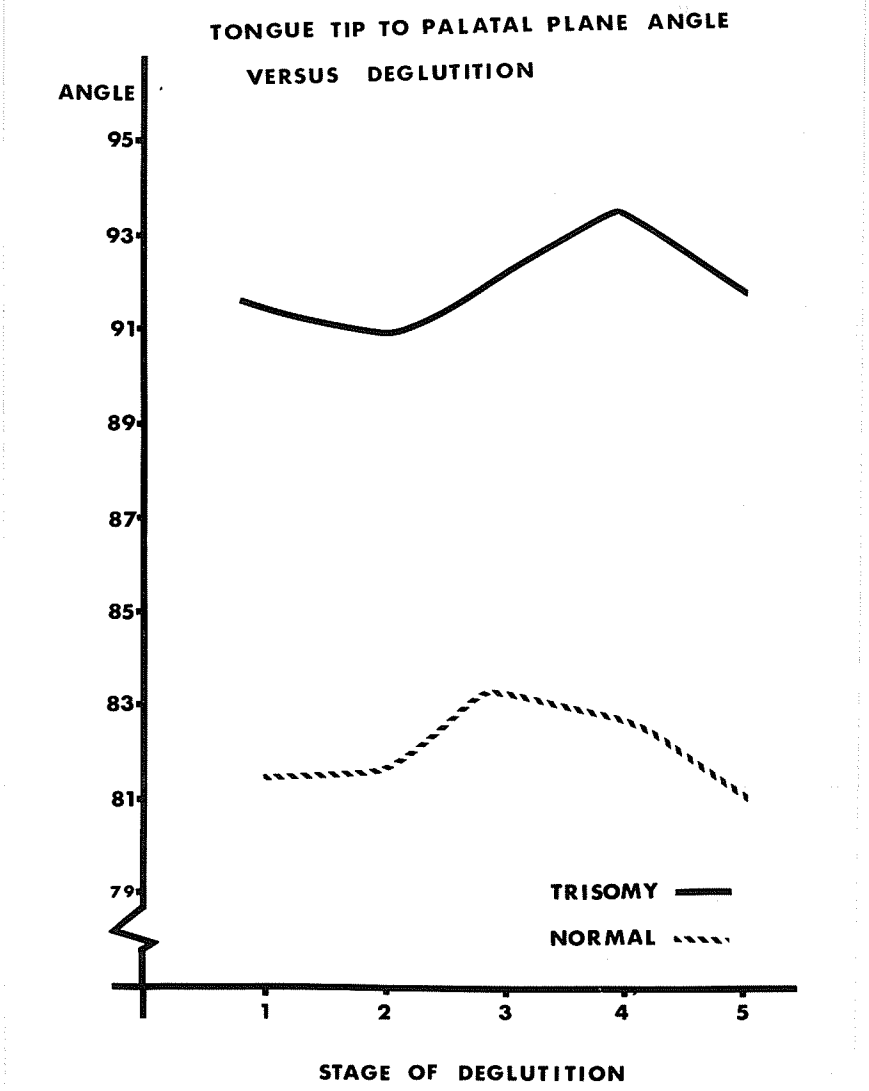

Figure 34. A graphic illustration of the tongue tip to palatal plane angle as it relates to the 5 stages of deglutition in the trisomy and normal groups.

The vertical tongue position was found to be significantly lower in the trisomy 21 at the rest position; this was significant at the 0.01 level. During the second, third and fourth stages of deglutition, the tongue position was comparable to that of the normal group, there being no significant difference at these stages. This is illustrated graphically in figure 35 and also demonstrated in table XXIV.

TABLE XXIV

VERTICAL TONGUE HEIGHT: MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Deglutition	Mean	Standard Error	Mean	Standard Error	F Value Difference
Stage 1	1.452	0.097	1.158	0.060	9.33 **
Stage 2	1.275	0.056	1.274	0.066	0.03
Stage 3	0.371	0.020	0.387	0.024	0.60
Stage 4	0.371	0.025	0.379	0.025	0.29
Stage 5	1.474	0.088	1.124	0.061	10.85 **

** Significant at the 0.01 level of confidence.

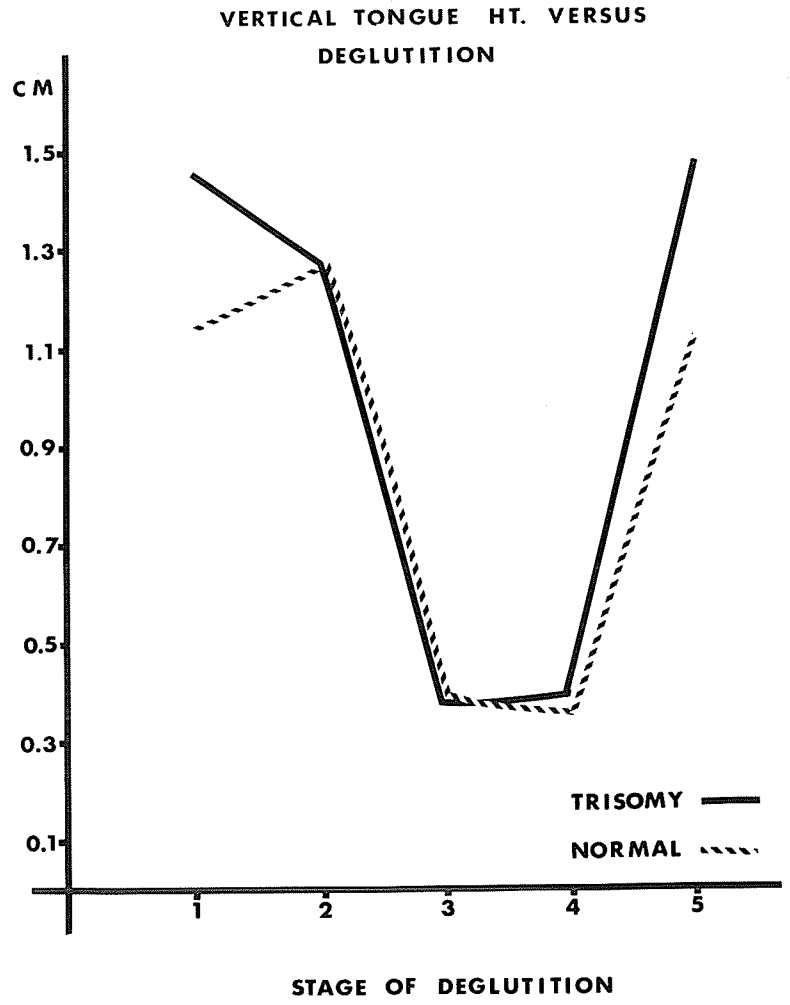

Figure 35. A graphic illustration of the vertical tongue height in centimeters as measured from the dorsum of the tongue to the palatal plane in the 5 stages of deglutition.

These results suggest the contour or shape of the tongue was essentially the same in the two groups, with no significant difference in the functional aspects of the deglutition cycle. However, the positional analyses indicated that the tongue tended to be lower and more protruded in the trisomy 21 group.

2. Hyoid Movement

The angular measurement of the anterior superior position of the hyoid to the palatal plane suggested that there was no significant difference in its movement between the two groups during the various stages of deglutition. This is illustrated in tabular form in table XXV of the Appendix.

3. Soft Palate Movement

The angle formed by the soft palate to the palatal plane suggested that the soft palate in the trisomy 21 was more obtuse than in the comparable normal group. Although the values were consistently higher in the trisomy 21 group, the difference was significant ($p < 0.05$) only in the second and fifth stages of deglutition. This is illustrated in table XXVI of the Appendix.

4. Vertebral Analyses

These analyses consisted of two vertical measurements within the first three cervical vertebrae; two angular measurements, as determined by the angle formed by the plane of the cervical vertebrae to the palatal plane, and the measurement of the distance between the posterior nasal spine and the anterior tubercle of the atlas. The distance was measured between the first and third and the first and second vertebrae. These measurements suggested that there was very little movement of these vertebrae during the deglutition cycle. The only significant difference was the overall distance between the vertebrae. This distance was significantly smaller in the trisomy 21 group ($p < 0.01$) at all stages of deglutition, indicating that this vertical length was smaller. This is illustrated in figure 36 and table XXVII, as well as in table XXVIII of the Appendix.

TABLE XXVII

DISTANCE IN CENTIMETERS BETWEEN FIRST AND THIRD VERTEBRAE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference
Stage 1	3.77	0.082	4.30	0.087	44.56 **
Stage 2	3.78	0.087	4.27	0.087	23.11 **
Stage 3	3.78	0.087	4.26	0.092	23.70 **
Stage 4	3.79	0.086	4.21	0.090	14.35 **
Stage 5	3.77	0.087	4.29	0.084	26.43 **

** Significant at the 0.01 level of confidence.

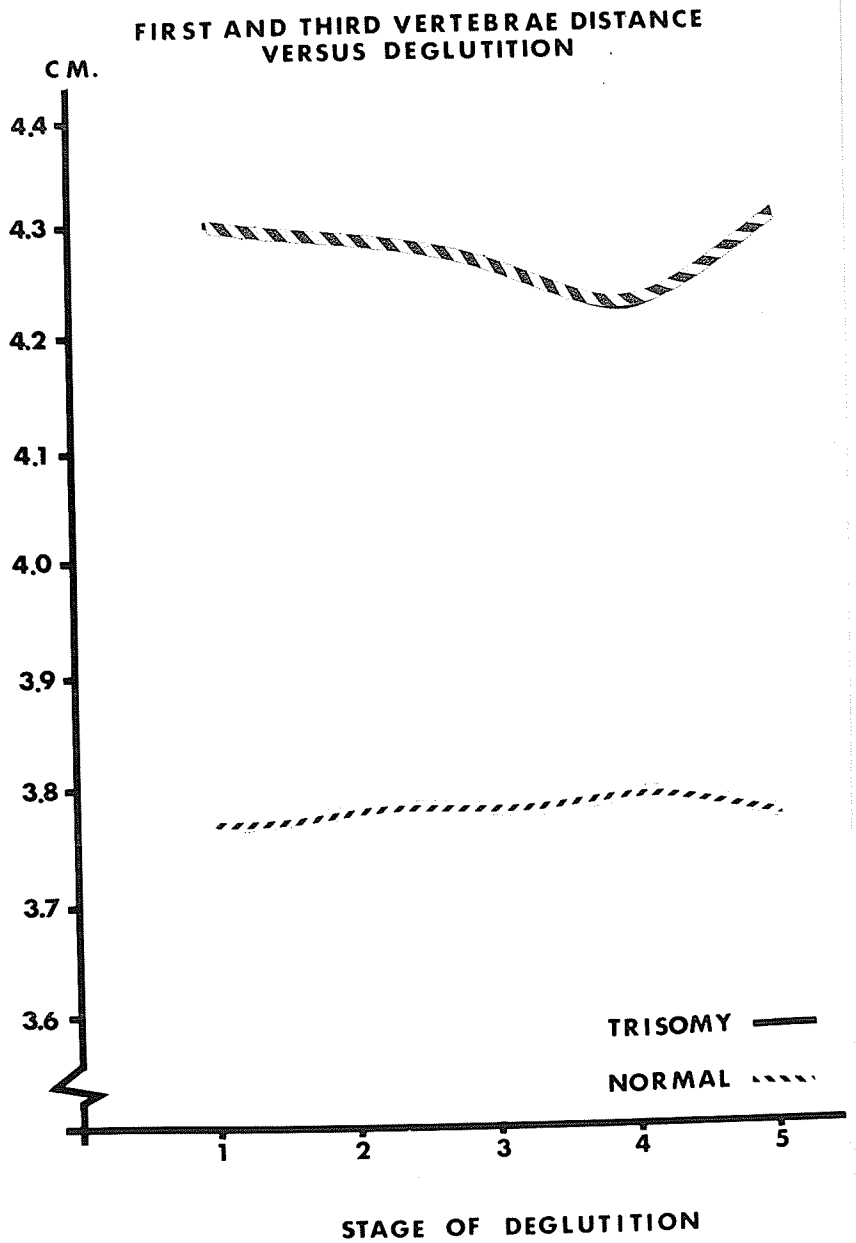

Figure 36. A graphic illustration of the distance in centimeters between the first and third vertebrae at the 5 stages of deglutition.

The two angular measurements, formed by the intersection of the vertebral planes and the palatal plane, indicated that these angles were more obtuse in the trisomy 21 group. This finding was significant at the 0.01 level of confidence at all stages of deglutition when the first and second vertebrae were used as the vertebral plane, and all stages except the fifth stage when the first and third vertebrae were used as the vertebral plane. In this latter instance the fifth stage was significantly different at the 0.05 level of confidence. This is illustrated in figures 37 and 38 and tables XXIX and XXX. These findings suggested that the head of the trisomy 21 individual was more extended than that of the comparable normal sample.

In the statistical analysis of the individual age groups, the results were invariably the same as those indicated in the grouped data (see tables XXXI through table XXXV of the Appendix). The 6-10 year age group and the adult age group demonstrated the most head extension in the trisomy sample, as illustrated in figure 39. It should be noted that in the trisomy 21 sample, from the 11-12 year age group onward, the head became progressively more extended during deglutition. The head extension of the normal subjects were comparable to that of the trisomy 21 subjects at the 11-12 age group, however, there was considerably less variability (see figure 40). It should be noted that the angulation in the normal subjects changed very little through the various age groups.

TABLE XXIX

FIRST AND SECOND VERTEBRAE TO PALATAL PLANE ANGLE: MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Deglutition	Means	Standard Error	Means	Standard Error	F Value Differences
Stage 1	97.41	1.542	90.57	1.346	25.22 **
Stage 2	97.85	1.447	90.28	1.224	32.97 **
Stage 3	97.41	1.466	90.31	1.31	31.51 **
Stage 4	98.42	1.414	92.05	1.357	25.27 **
Stage 5	97.22	1.552	90.25	1.313	23.71 **

** Significant at the 0.01 level of confidence.

TABLE XXX

FIRST AND THIRD VERTEBRAE TO PALATAL PLANE ANGLE: MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Deglutition	Mean	Standard Error	Mean	Standard Error	F Value Difference
Stage 1	99.22	1.437	93.89	1.166	8.47 **
Stage 2	99.11	1.367	93.65	1.105	9.13 **
Stage 3	98.69	1.344	93.95	1.135	7.65 **
Stage 4	100.27	1.366	94.54	1.219	9.40 **
Stage 5	98.83	1.491	93.74	1.033	6.67 *

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

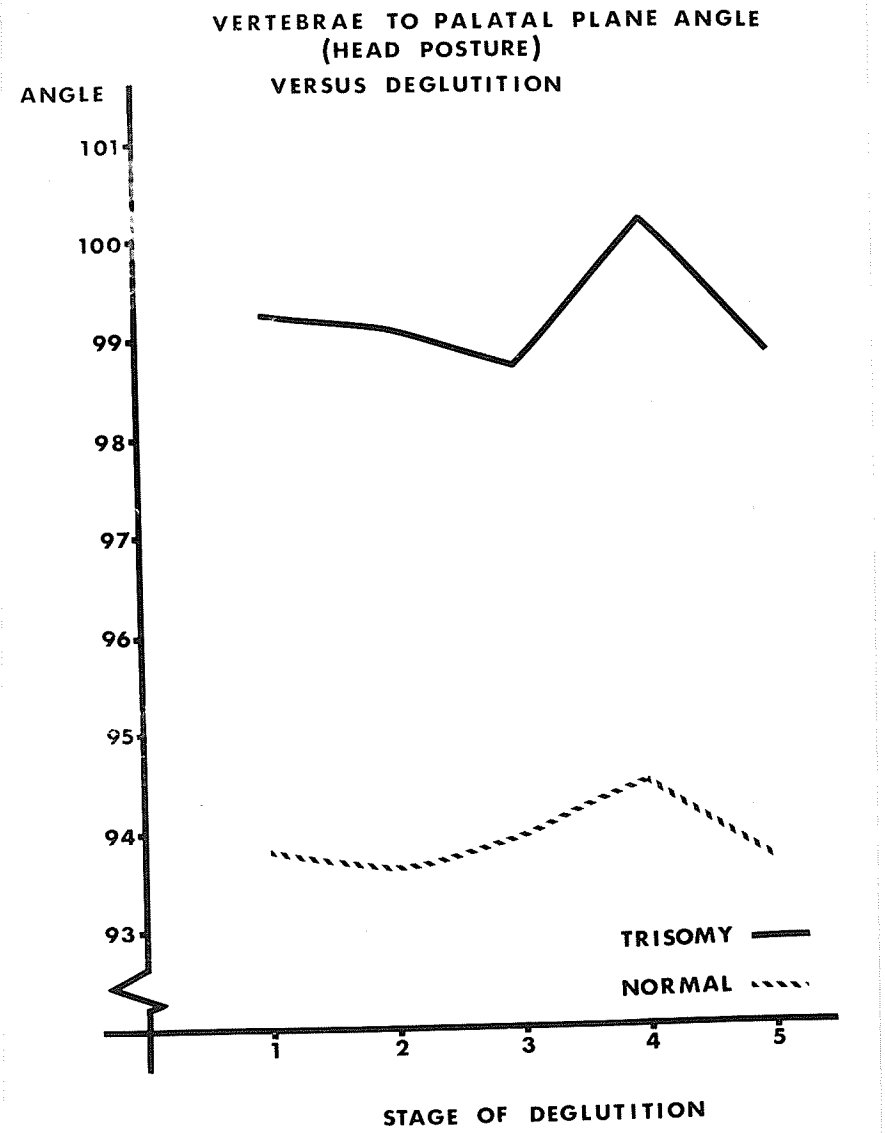

Figure 37. A graphic illustration of the angle formed by a line joining the first and second vertebrae to the palatal plane at the various stages of deglutition.

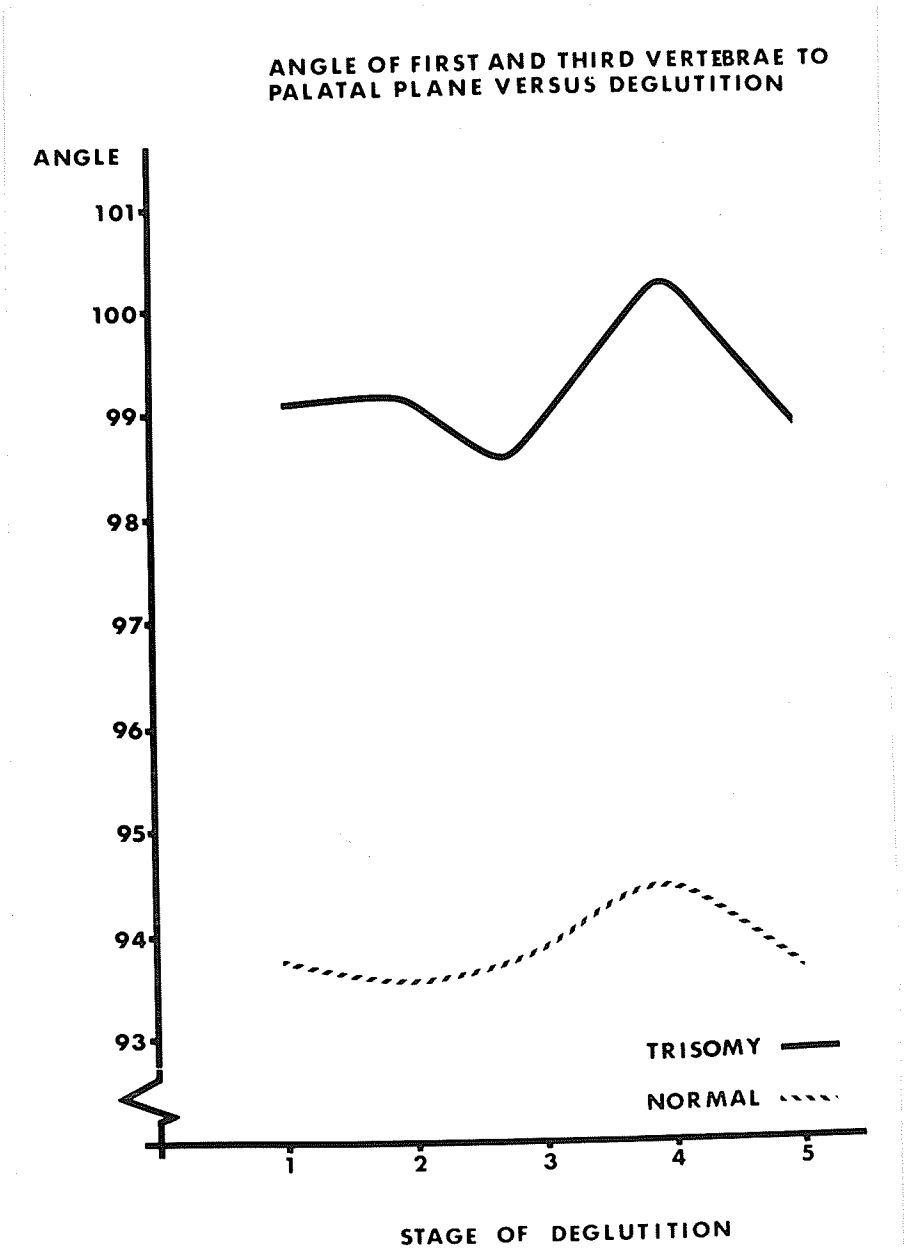

Figure 38. A graphic illustration of the angle formed by a line joining the first and third vertebrae to the palatal plane at the various stages of deglutition.

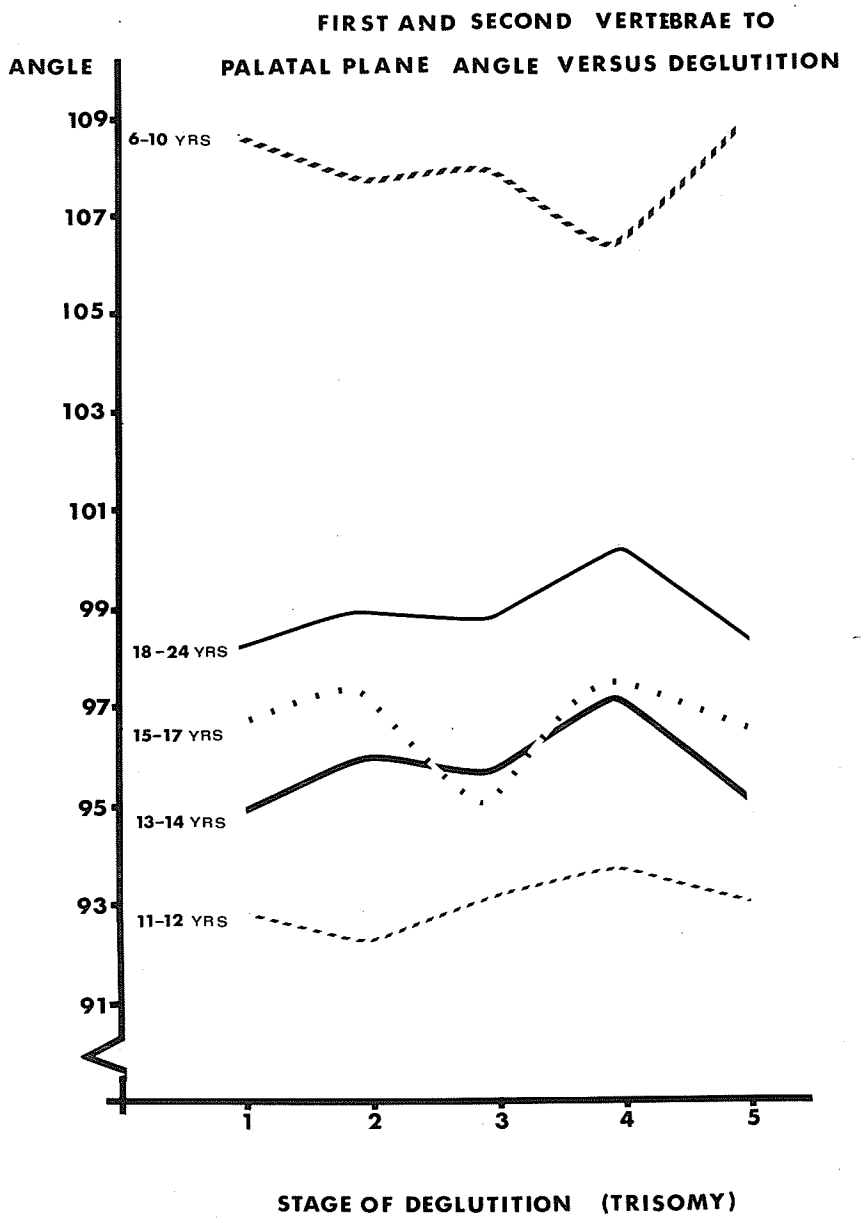

Figure 39. A graphic illustration of the first and second cervical vertebrae to palatal plane angle in the trisomy 21 group at each of the five age groups as they vary at the 5 stages of deglutition.

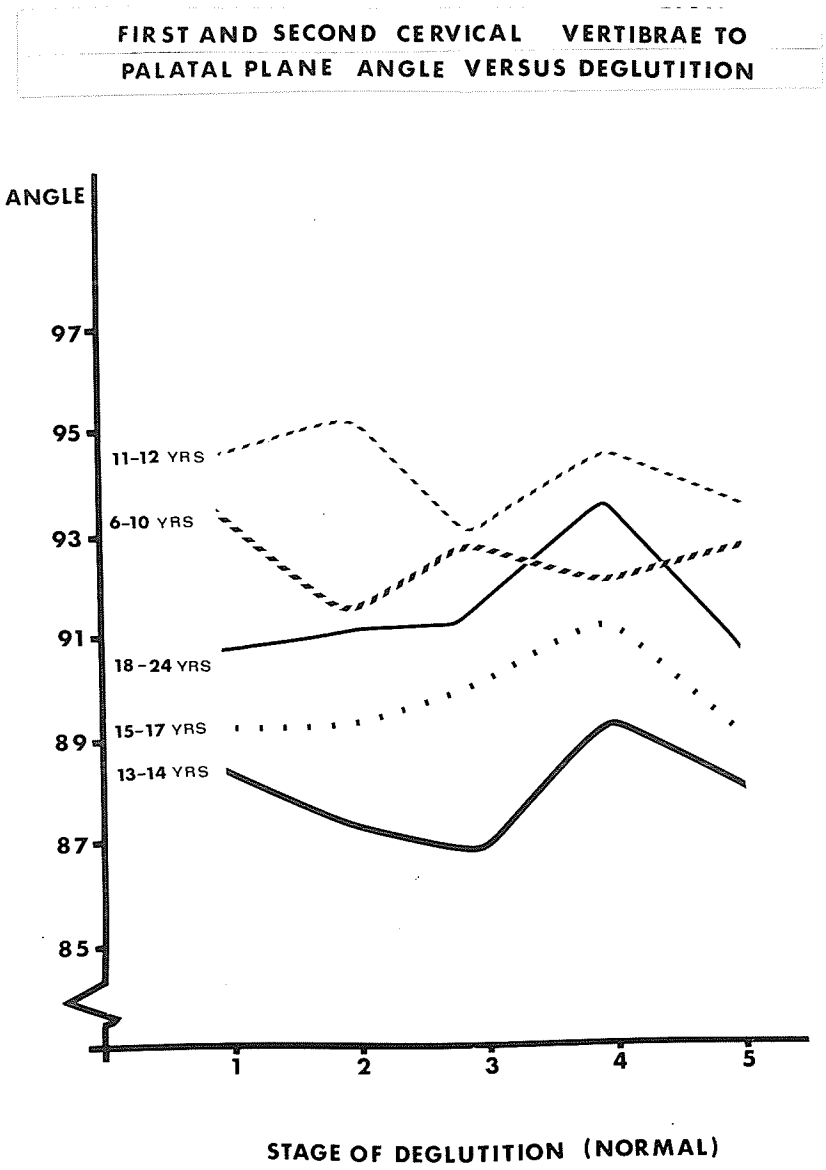

Figure 40. A graphic illustration of the first and second certical vertebrae to palatal plane angle in the normal sample at each of the five age groups as they vary at the 5 stages of deglutition.

The measurement of the distance from the posterior nasal spine to the anterior tubercle of the atlas indicated that the trisomy 21 subjects had a larger nasopharyngeal opening than the normal group. This finding was significant at the 0.01 level of confidence and is illustrated graphically in figure 41 and in tabular form in table XXXVI.

TABLE XXXVI

DISTANCE IN CENTIMETERS FROM POSTERIOR NASAL SPINE TO THE ANTERIOR TUBERCLE OF THE ATLAS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal			
Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference	
Stage 1	4.04	0.091	3.59	0.077	23.37	**
Stage 2	4.02	0.088	3.62	0.073	19.81	**
Stage 3	4.02	0.084	3.62	0.072	21.31	**
Stage 4	4.07	0.087	3.66	0.075	20.19	**
Stage 5	4.02	0.94	3.60	0.072	18.57	**

** Significant at the 0.01 level of confidence.

These analyses suggest that the vertebrae were smaller and that their angle to the palatal plane was greater in the trisomy 21 subjects. This greater angulation subsequently was associated with a larger nasopharyngeal opening, as indicated by the measurement from the posterior nasal spine to the anterior tubercle of the atlas. The analyses also indicate that the head of the trisomy 21 becomes progressively more extended from 11-12 years of age to adulthood. The normal sample demonstrated very little change although some extension was noted during this period.

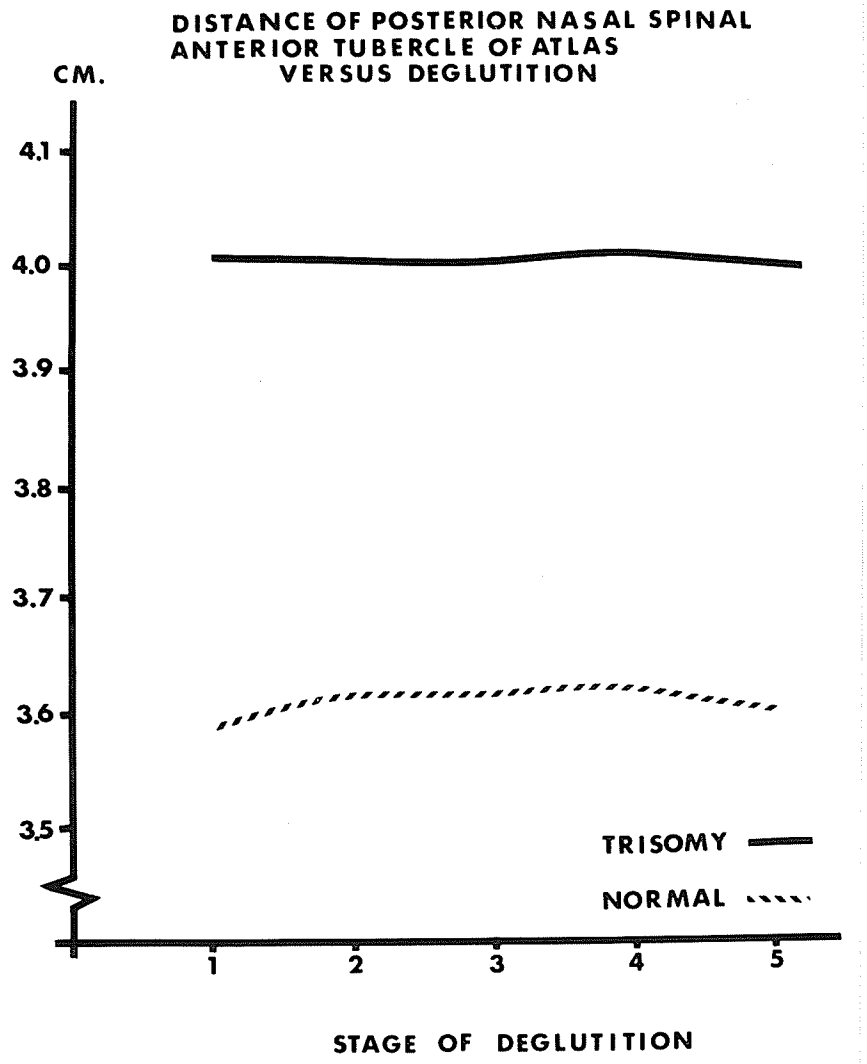

Figure 41. A graphic illustration of the distance in centimeters between the posterior nasal spine and the anterior tubercle of the atlas at the 5 stages of deglutition.

DISCUSSION

CHAPTER V

DISCUSSION

During this discussion an attempt will be made to integrate the results of the various analyses. This integration shall define some of the mechanisms of adaptation resulting from alterations in both bony and soft tissue morphology. The dichotomy of whether the structural morphology was due to the basic genetic defect or due secondarily to the tissues response to the basic defect was difficult to assess.

In this thesis an attempt has been made to apply several criteria in an attempt to solve this problem. However, there remains little doubt that the functional or morphological causes of adaptation are multi-factorial in nature.

Skeletal Morphology

The general morphology has been well documented, at least from a cephalometric view point. Some of the investigators have been Kisling (1966), Ghiz (1969), Frostad (1969), Nevile (1973), and Alimchandani (1973). The results of the present study generally concur with the findings of the above investigators.

Some of the significant structural differences found between the trisomy 21 and normal groups shall be presented in order to establish a framework upon which to make a differential assessment of morphology and function.

The angle of the cranial base, in the present study, was found to be more obtuse in the trisomy 21 subjects, with the anterior cranial base being considerably reduced in length as compared to the comparable normal group. The posterior cranial base appeared to be less affected than the other two parameters. These findings are consistent with those of Kisling (1966) and Ghiz (1969).

The antero-posterior position of the maxilla was found to be well related to the anterior cranial base, but was shorter in length in the trisomy 21 group relative to the normal group. These findings are consistent with those of Kisling (1966), Ghiz (1969), and are contrary to those of Sassouni et al. (1964). This antero-posterior position of the maxilla, as determined by SNA, was not consistent with the clinical picture of the trisomy 21 individual. However, the anterior cranial base was reduced in length, and since it was used as the base line, this essentially negates the hypoplasia in the maxilla.

These findings are consistent with Enlow's (1973) concept of craniofacial growth, in that one might anticipate a hypoplasic maxilla upon the finding of a hypoplasic

anterior cranial base. The link, suggested by Enlow, of underdevelopment of the temporal lobes of the brain being associated with anterior cranial base deficiency has not been substantiated in this study. However, Benda (1960) has reported decreased weight of the brain in the trisomy 21 individual at birth and throughout life. He reported flattening of the convolutions and compression of the frontal and temporal poles, resulting in the temporal lobes being twisted and distorted. These distortions of the temporal lobes include the upward displacement of the superior and middle temporal fissures and a twisting of the anterior temporal pole.

The mandible, in the present study, was found to be smaller but relatively less affected than other craniofacial structures in the trisomy 21 individuals. This finding was consistent with the findings of Nevile (1973) and suggested that this bone developed somewhat separately from the maxilla and the anterior cranial base structures.

From the results it became apparent that the clinical prognathic appearance of the typical trisomy 21 individual was due to several factors. The prime factors associated with this facial profile appeared to be the hypoplasia of the anterior cranial base and maxilla in conjunction with a relatively large mandible. The obtuse cranial base may effectively

diminish the prognathism but may exaggerate the anterior open bite tendency reported in trisomy 21 groups by Kisling (1966) and Jensen (1973). These investigators reported an open bite incidence of 54% and 48% respectively in their trisomy 21 samples.

Soft Tissue Morphology

In order to complete the morphological description, the soft tissue structures must be related to their corresponding hard tissue counterparts. The growth of the soft tissues often illustrated timing quite different from the bony tissues to which they were attached. This was exemplified by the nasopharynx, where the bony growth preceded the soft tissue lymphatic growth, Scammon (1953). The findings from the cephalometric area analyses generally agree with previous clinical "subjective" observations. The nasal area, because of its radiological definition, did not change appreciably in the skeletal-to-soft tissue measurements. This was not true for the nasopharynx. The total bony size of the nasopharynx was not found to be appreciably different between the two groups (table IV). This was explained by two independent findings. The cervical vertebrae, which supported the soft tissue for the posterior wall of the nasopharyngeal area, was more obtuse to the palatal plane in the trisomy 21 group, as illustrated in graph 37 and table XXVIII. This, along with a reduced palatal length (figure 30) appeared to adequately maintain

the size of the bony nasopharyngeal area. This was demonstrated by the measurement from the posterior nasal spine to the anterior tubercle of the atlas, which was found to be consistently larger in the trisomy 21 individuals (table XVI). The nasopharynx area analysis indicated that the soft tissue here was more abundant in the trisomy 21 individuals than in the normal individuals. This finding became increasingly significant from age groups 11-12 years through to adulthood as illustrated in figure 24 and table III. Since this measurement was one of airway space, it must be assumed that in the trisomy 21 group the airway did not increase appreciably after 11-12 years and in the normals group after 13-14 years of age.

The oropharynx and oral cavity encompass many soft tissue structures. The findings indicated that the soft palate was consistently larger in the trisomy 21 group at all age levels, compared to the normal sample (figure 27 and table VII). The reason for this increased size was not readily apparent, however, the increased distance to the posterior pharyngeal wall, as well as the invariable chronic inflammation present in this area, may be, responsible for the discrepancy.

The findings in regard to tongue size suggested that the tongue in the trisomy 21 individual was smaller

than that found in normal subjects. This was consistent with the findings of Thomson (1907), Gillis et al. (1968) and contrary to those of Gosman (1951). The tongue growth increased rapidly to age 11-12 years and then continued slowly to adulthood in the trisomy 21 group. In the normal subjects, however, the tongue size reached a maximum at 13-14 years and did not change appreciably thereafter. These findings are contrary to those of Vig (1974) who suggested that the tongue became relatively smaller from 10 years of age to adulthood. It became apparent that the clinical appearance of the "enlarged" trisomy 21 tongue was due to other factors rather than an absolute tongue size. It was felt that posture and position were vital factors in this regard.

The oropharynx and oral cavity may be considered together because of their continuity and general proximity. The measurements of these areas demonstrated a considerable amount of variability, however, both areas were significantly larger in the trisomy 21 individuals at the adult age groups.

The analysis of the soft tissue morphology indicated that these tissues display much variability as they follow the hard tissue contours. With the exception of the nasopharynx, these parameters have been little studied.

The trisomy 21 group illustrated certain distinct features not common to the normal group. For example, it can be noted in figure 42 that as the nasopharynx airway discontinued growth in the trisomy 21 group, the oropharynx airway starts to increase more rapidly. These findings became more dramatic when areas of the oropharynx and oral cavity were added together, as in figure 43.

There remains little doubt that these basic observations are over-simplifications of what is undoubtedly a multi-factorial, highly integrated mechanism. It was felt that this variation in structural morphology must have an affect on the functional aspects of the system.

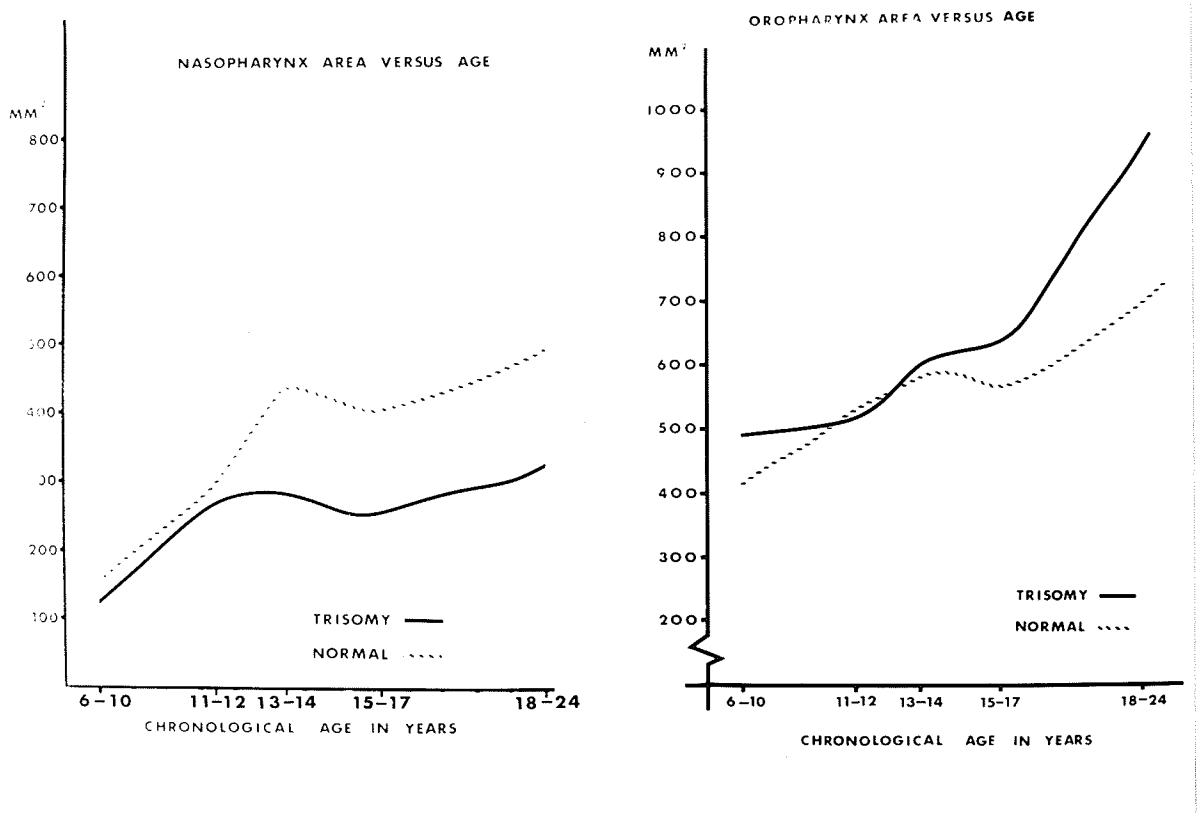

Figure 42. A graphic illustration of nasopharynx compared to age group and oropharynx compared to age group.

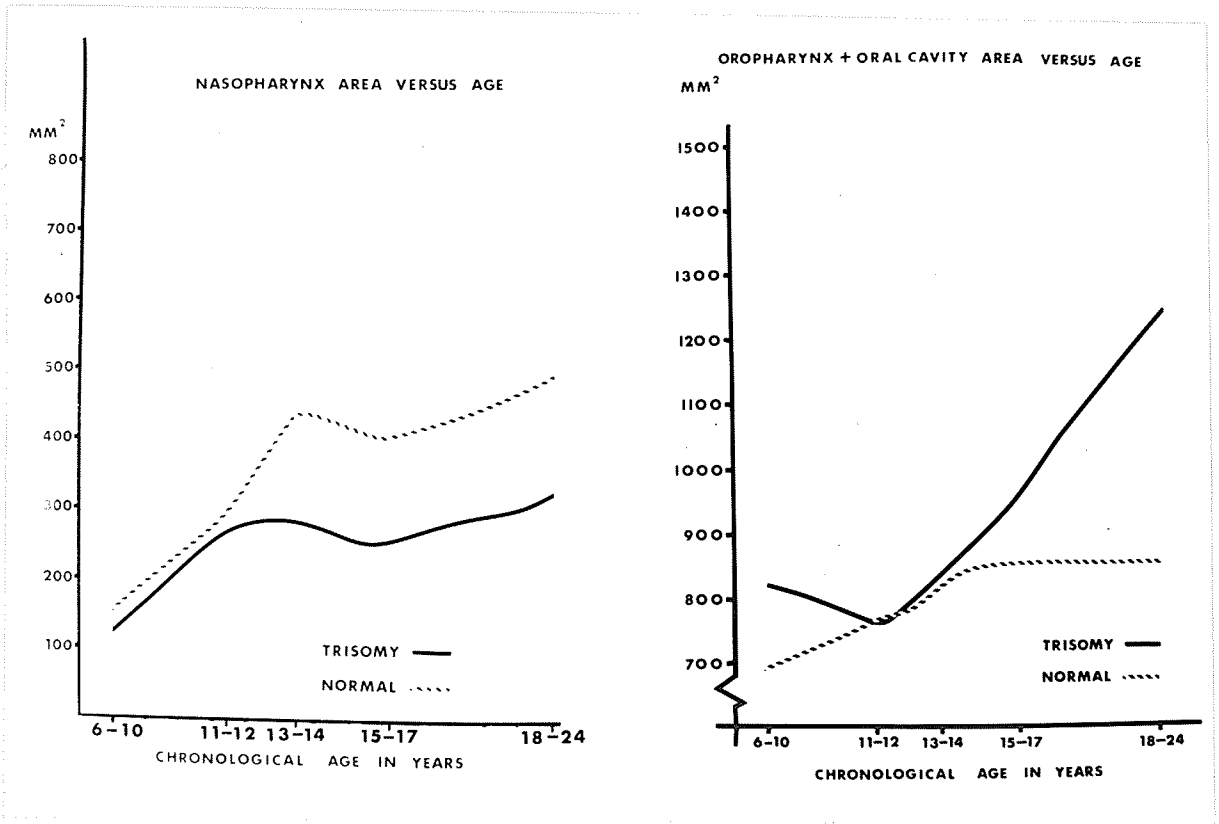

Figure 43. A graphic illustration of nasopharynx compared to age group and oropharynx plus oral cavity compared to age group.

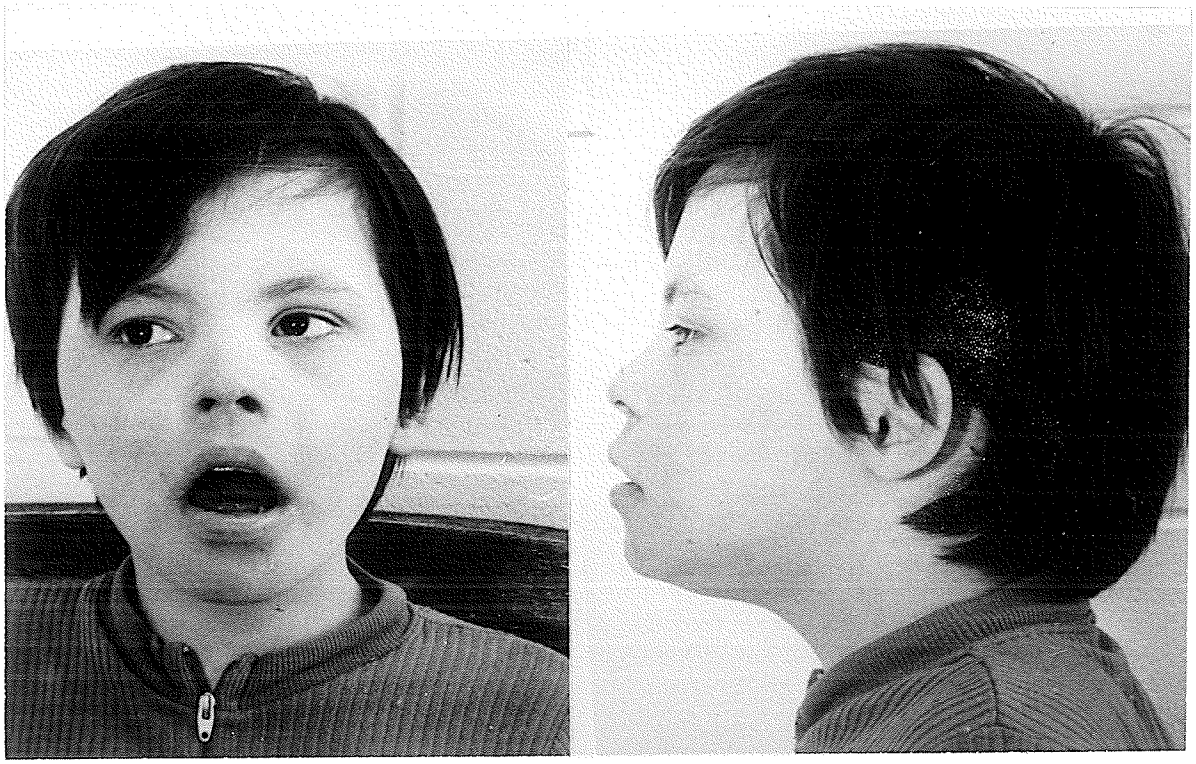

Figure 44. A photograph of a trisomy 21 individual suffering from chronic respiratory distress.

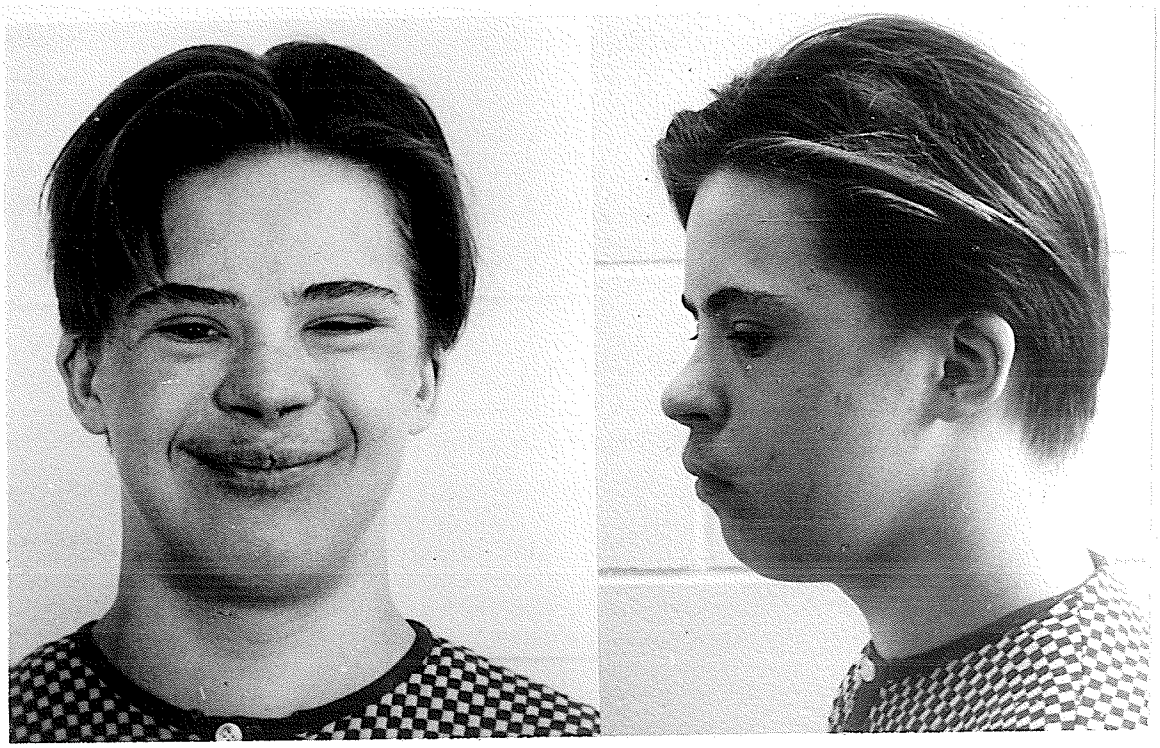

Figure 45. An adult trisomy 21 individual.

Functional Analysis

The analysis of deglutition consisted of two stages at rest position and three stages during function which were similar to those used by Cleall (1965). The differentiation of these functions were necessary since the findings suggested that the mechanism of the deglutition cycle (and hence three 'moving' stages) were essentially the same in the trisomy 21 and normal groups. There were, however, distinct differences present at the rest position. The position of the tongue at rest position was found to be lower (figure 35, table XXIV) as well as in a more protruded position (figure 34, table XXIII). These findings were significant at the 0.01 level of confidence. The position of the tongue was undoubtedly related to several factors; those related to morphology and those related to maintenance of the vital function of respiration. From the morphological viewpoint, the hypoplasia of the maxilla, documented by Kisling (1966) and Frostad (1969), dictates that the tongue occupy a lower position. The tongue, however, need not protrude to adapt to the hypoplasic maxilla; the tongue protrusion effectively increased the area of the oropharynx and established a more substantial route for air flow for both nasal and oral breathing.

The angulation of the soft palate was found to be more obtuse in the trisomy 21 group and the author attributed

this to the tendency for these individuals to be mouth breathers, to the larger size of the soft palate (tables XXVI and VII and figure 27) and also to the tendency for these individuals to have chronic respiratory infections.

The cervical vertebrae have demonstrated considerable functional adaptation, in respect to positional changes relative to the palatal plane, this probably occurred in response to constriction of the nasopharyngeal air flow. This adaptation could result in an increase in the oropharynx and oral cavity areas and would seem to correspond to the amount of soft tissue constriction noted in the nasopharynx. This was especially true in the trisomy 21 group, where both growth and absolute size of the airway were considerably reduced. The position of the head, as determined by the plane of the cervical vertebrae to the palatal plane, suggested that the head was significantly more extended in the trisomy 21 group, as compared to the normal group. This extension effectively increased the distance between the posterior nasal spine and the anterior tubercle of the atlas. The measurement was significantly larger in the trisomy 21 group, hence supporting the hypothesis that this was an adaptive respiratory mechanism.

The possibility that palatal tipping influenced the angular measurements of the palatal plane to the plane of the cervical vertebrae can be eliminated, since the

cephalometric findings suggest that the palatal plane in the trisomy 21 group tips downward anteriorly more than that of the normal sample. (Appendix tables XIII and XIV). This was consistent with the findings of Frostad (1969).

These findings suggested that a series of structures adapted in an integrated fashion to compensate for the decreased nasopharyngeal air flow. These structures included the cervical vertebrae, which become more removed from the palatal plane as the nasopharynx airway decreases in growth (tables XXXIX and XL); the oropharynx and oral cavity airways simultaneously enlarged (tables XLII and XLIII), and the tongue tended to occupy a lower and more protruded position.

It became apparent that both hard and soft tissue adaptation were involved in what have long been considered the mongoloid 'phenotype'. The effect of the compensatory mechanisms of the soft tissues of the tongue and soft palate, as well as the hard tissues of the cervical vertebrae, co-ordinate in maintaining the vital function of respiration. The affect of these compensatory changes on adjacent structures was indeed difficult to assess. Although it has been well-documented that the maxilla is hypoplastic, there was evidence of a low tongue posture which may have contributed to both the lateral constriction of the palatal shelves as well as the high incidence of posterior crossbites. The finding of Jensen (1973) indicated that there was a high incidence of bilateral posterior crossbites. These occurred

in 68.5% of the females and 42.5% of the males in his trisomy 21 sample. This conclusion is supported by Friel (1926), who suggested that the static function of the soft tissue structures were responsible for moulding the dental arches. It was contrary to the statements of Brash (1929) who related that it was improbable that the tongue exercised any important mechanical influence on the general form and size of the mandible or maxilla.

The proclined maxillary incisors found in the trisomy 21 sample were undoubtedly aggravated by, if not due to, the protruded tongue upon which these incisors very often rest. The high incidence of open bites may, in part, be attributable to the obtuse cranial base flexure and to the size and shape of the mandible.

This thesis has presented evidence to support treatment therapy directed at increasing and maintaining a patent airway in the trisomy 21 individual. It was felt that the postural positions, due to respiratory impairment, have minimal effect on the cranial facial structures, relative to the other major implications of the syndrome. Nonetheless, far reaching results may be obtained by increasing the nasal air flow. This includes improved lower respiratory and heart efficiency, as suggested by Unno et al. (1969) and Menasche (1965). Further research is required in the treatment of the nasopharyngeal obstruction, however, one may postulate

that partial removal of the pharyngeal tonsil and rapid maxillary expansion in selective cases may be helpful.

There is little doubt that the effects of compensation due to decreased nasal air flow as presented for the trisomy 21 group are essentially the same as those acting in normal individuals. Thus, the study of these compensatory changes offers an insight into the etiology and treatment not only for trisomy 21 children, but also for normal individuals affected with a similar problem.

SUMMARY AND CONCLUSIONS

CHAPTER VI

SUMMARY AND CONCLUSIONS

This study attempted to integrate information obtained from several radiographic analyses of the craniofacial area, in order to contribute to the understanding of the phenotype presented by trisomy 21 individuals. Emphasis was placed on the relationship of respiration and deglutition to the morphological size of structures within the oral nasal region. The analyses consisted of an area analysis obtained from a lateral cephalogram, a co-ordinate analysis also using a lateral cephalogram and a functional analysis utilizing a cinefluorographic sequence of deglutition.

The sample consisted of 40 trisomy 21 individuals divided into 25 males and 15 females with an age range of 6 years to 24 years. Each individual was placed in one of 10 groups according to sex and age. A "normal" sample was used for comparison having a similar distribution.

The results consisted of area, linear and angular measurements which were evaluated statistically by a multivariant analysis. The statistical and subjective evaluation of these results suggest the following conclusions.

1. The areas of the nasal cavity and nasopharynx were found to be significantly smaller in the trisomy 21 group.
2. The oropharynx and oral cavity tended to be larger

in the trisomy 21 individuals especially at the adult age level.

3. The soft palate was consistently larger in the trisomy 21 sample.
4. The tongue tended to be smaller in the trisomy 21 group as compared to the normal sample.
5. The pharyngeal tonsil in the trisomy 21 group discontinued involution earlier than the normal group, resulting in a smaller nasopharyngeal airway. As the growth in the nasopharyngeal airway decreased, the plane of the cervical vertebrae to the palatal plane was found to become more obtuse. The resulting progressive extension of the head increased the distance from the posterior nasal spine to the anterior tubercle of the atlas, thus effectively increasing the size of the oropharyngeal airway in the trisomy 21 group.
6. The tongue in the trisomy 21 group was found to occupy a lower and more protruded position than the comparable normal sample.
7. The tongue posture may be a contributing factor to the high incidence of palatal constriction, posterior crossbites and proclined maxillary incisors found in the trisomy 21 group.

8. The high incidence of anterior open bites may be in part attributable to the obtuse cranial base found in the trisomy 21 group.
9. The apparent mandibular prognathism observed in the trisomy 21 group appears to be related to the differential growth of the maxilla and mandible, as well as, hypoplasia of the anterior cranial base.
10. The typical mongoloid appearance was felt to be due not only to the basic genetic defect but also to adaptive mechanisms involving the tongue and the cervical vertebrae which function in response to a constricted airway. The most important site of this constriction appeared to be the nasopharynx.
11. This thesis presented evidence to support treatment therapy directed at increasing and maintaining a patent airway in the trisomy 21 individual.

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APPENDIX

TABLE XII

ANTERIOR CRANIAL BASE LENGTH (SELLA-NASION), MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

Trisomy			Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	5.79	0.173	6.40	0.126	3.64
11-12 yrs	5.98	0.054	6.64	0.246	4.41 *
13-14 yrs	6.00	0.112	6.86	0.094	6.04 **
15-17 yrs	6.25	0.087	6.81	0.165	4.73 **
18-24 yrs	6.05	0.090	6.81	0.109	8.18 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

TABLE XIII

ANTERIOR NASAL HEIGHT, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	3.58	0.294	4.48	0.114	4.05 *
11-12 yrs	4.14	0.139	4.59	0.085	2.26
13-14 yrs	4.24	0.093	4.91	0.092	4.77 **
15-17 yrs	4.46	0.117	4.87	0.127	2.61
18-24 yrs	4.32	0.145	4.96	0.221	5.61 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

TABLE XIV

NASOPHARYNX HEIGHT (52 24), MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	2.16	0.218	2.57	0.174	2.450
11-12 yrs	2.46	0.141	2.60	0.146	0.935
13-14 yrs	2.60	0.142	2.76	0.111	1.512
15-17 yrs	2.58	0.110	2.83	0.103	2.113
18-24 yrs	2.55	0.092	3.09	0.124	5.818 **

** Significant at the 0.01 level of confidence.

TABLE XV

NASAL LENGTH (52 4), MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

Trisomy			Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	4.30	0.258	4.81	0.249	1.277
11-12 yrs	4.15	0.076	4.97	0.181	2.296
13-14 yrs	4.33	0.130	5.08	0.121	2.969
15-17 yrs	4.91	0.110	5.47	0.147	1.983
18-24 yrs	4.57	0.120	4.69	0.486	0.475

TABLE XVI

NASOPHARYNX WIDTH (45 47), MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN THE TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	3.10	0.104	2.97	0.202	0.732
11-12 yrs	3.36	0.093	3.02	0.208	2.142
13-14 yrs	3.35	0.116	3.16	0.083	1.693
15-17 yrs	3.24	0.085	2.97	0.092	2.151
18-24 yrs	3.22	0.113	3.94	0.118	2.844

TABLE XVII

PALATAL LENGTH, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN THE TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	3.93	0.086	4.41	0.059	2.80
11-12 yrs	3.92	0.055	4.51	0.076	3.85 *
13-14 yrs	4.07	0.073	4.57	0.075	4.62 **
15-17 yrs	4.25	0.116	4.82	0.106	4.71 **
18-24 yrs	4.09	0.098	4.50	0.179	4.32 **

* Significant at the 0.05 level of confidence.

** Significant at the 0.01 level of confidence.

TABLE XVIII

UPPER INCISOR TO SN PLANE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	109.99	2.526	102.22	1.485	2.38
11-12 yrs	110.48	2.194	104.98	3.284	0.51
13-14 yrs	110.34	1.923	104.67	2.056	2.77
15-17 yrs	108.18	2.386	105.20	3.406	1.30
18-24 yrs	110.95	1.591	105.69	1.802	2.95

TABLE XIX

SELLA-NASION-B POINT, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	82.58	1.309	75.50	1.614	3.754
11-12 yrs	79.25	1.039	78.64	1.285	0.339
13-14 yrs	82.89	1.283	77.77	0.961	4.070 *
15-17 yrs	78.72	1.574	79.70	1.248	0.700
18-24 yrs	82.43	1.148	81.48	1.251	0.864

* Significant at the 0.05 level of confidence.

TABLE XX

THE ANGLE OF CONVEXITY, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	182.77	3.042	165.33	0.750	5.529 *
11-12 yrs	178.21	1.373	166.85	3.158	4.050 *
13-14 yrs	179.66	2.095	171.93	1.353	3.869 *
15-17 yrs	177.99	2.300	172.06	1.954	2.652
18-24 yrs	182.22	2.281	178.73	1.693	2.005

* Significant at the 0.05 level of confidence.

TABLE XXI

THE SELLA-NASION-POGONION ANGLE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN THE TRIOSMY AND NORMAL GROUPS

	Trisomy		Normal		
Group Age	Mean	Standard Error	Mean	Standard Error	Duncan Test Difference
6-10 yrs	82.65	1.475	75.70	1.598	3.633
11-12 yrs	79.31	1.048	79.76	1.648	0.263
13-14 yrs	83.51	1.311	78.67	0.902	4.000 *
15-17 yrs	79.59	1.320	80.62	1.200	0.761
18-24 yrs	83.65	1.194	83.65	1.233	0.010

* Significant at the 0.05 level of confidence.

TABLE XXII

ANGLE OF THE DORSUM OF TONGUE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference
Stage 1	119.51	1.889	115.95	1.952	0.52
Stage 2	134.81	1.993	134.09	2.426	0.01
Stage 3	107.74	1.335	107.28	1.200	0.04
Stage 4	109.39	1.201	107.47	1.173	0.17
Stage 5	120.96	1.982	115.87	1.482	4.74 *

* Significance at the 0.05 level of confidence.

TABLE XXV

HYOID TO PALATAL PLANE ANGLE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference
Stage 1	47.98	0.944	46.93	0.905	1.56
Stage 2	46.60	0.817	46.60	0.858	0.10
Stage 3	45.60	1.058	44.06	0.887	2.14
Stage 4	46.91	0.958	46.97	0.880	0.03
Stage 5	48.67	1.016	47.50	0.863	0.85

TABLE XXVI

SOFT PALATE TO PALATAL PLANE ANGLE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference
Stage 1	133.65	1.056	131.56	1.236	1.48
Stage 2	134.78	1.076	131.83	1.312	5.98 *
Stage 3	135.10	1.306	133.62	1.293	3.61
Stage 4	143.75	1.138	141.33	1.206	3.39
Stage 5	132.57	1.274	129.88	1.182	4.10

* Significant at the 0.05 level of confidence.

TABLE XXVIII

DISTANCE BETWEEN THE FIRST AND SECOND VERTEBRAE, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	F Value Difference
Stage 1	2.30	0.061	2.81	0.078	38.88 **
Stage 2	2.32	0.065	2.64	0.057	17.67 **
Stage 3	2.30	0.064	2.60	0.060	17.05 **
Stage 4	2.30	0.067	2.59	0.062	14.47 **
Stage 5	2.29	0.067	2.61	0.057	19.82 **

** Significant at the 0.01 level of confidence.

TABLE XXXI

FIRST AND SECOND CERVICAL VERTEBRAE TO PALATAL PLANE ANGLE FOR AGE GROUP 6-10 YEARS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal			
Stage of Deglutition	Means	Standard Error	Means	Standard Error	Duncan Test Difference	
Stage 1	108.45	4.55	93.26	5.64	3.50	
Stage 2	107.66	4.46	91.19	4.36	4.16	*
Stage 3	107.87	5.56	92.83	4.21	3.66	
Stage 4	104.66	5.65	92.08	3.39	3.02	
Stage 5	108.79	3.85	92.65	4.16	3.60	

* Significant at the 0.05 level of confidence.

TABLE XXXII

FIRST AND SECOND CERVICAL VERTEBRAE TO PALATAL PLANE ANGLE FOR AGE GROUP 11-12 YEARS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	Duncan Test Difference
Stage 1	92.73	5.72	94.51	3.45	0.45
Stage 2	92.29	5.00	95.78	4.19	0.98
Stage 3	93.04	4.41	93.57	3.93	0.14
Stage 4	93.56	3.90	93.51	4.72	0.90
Stage 5	93.71	5.24	93.47	4.21	0.06

TABLE XXXIII

FIRST AND SECOND CERVICAL VERTEBRAE TO PALATAL PLANE ANGLE FOR AGE GROUP 12-13 YEARS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	Duncan Test Difference
Stage 1	94.93	2.20	88.42	2.74	2.37
Stage 2	95.87	2.65	87.34	2.12	3.40 *
Stage 3	95.67	2.49	86.89	2.72	3.38 *
Stage 4	97.85	3.02	89.95	2.83	3.14 *
Stage 5	95.22	2.91	88.05	2.98	2.51

* Significant at the 0.05 level of confidence.

TABLE XXXIV

FIRST AND SECOND CERVICAL VERTEBRAE TO PALATAL PLANE ANGLE FOR AGE GROUP 15-17 YEARS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal		
Stage of Deglutition	Means	Standard Error	Means	Standard Error	Duncan Test Difference
Stage 1	96.60	3.09	89.20	2.67	2.41
Stage 2	97.14	3.07	89.36	2.00	2.77
Stage 3	94.79	3.10	90.49	2.61	1.46
Stage 4	97.44	2.86	91.28	2.73	2.09
Stage 5	94.58	3.58	88.05	2.98	2.17

TABLE XXXV

FIRST AND SECOND CERVICAL VERTEBRAE TO PALATAL PLANE ANGLE FOR AGE GROUP 18-24 YEARS, MEANS, STANDARD ERRORS AND THE SIGNIFICANCE OF THE DIFFERENCES BETWEEN TRISOMY AND NORMAL GROUPS AT THE 5 STAGES OF DEGLUTITION

	Trisomy		Normal			
Stage of Deglutition	Means	Standard Error	Means	Standard Error	Duncan Test Difference	
Stage 1	98.22	2.72	90.85	1.98	3.06	*
Stage 2	98.92	2.21	91.01	2.26	3.56	*
Stage 3	98.88	2.33	91.20	1.84	3.34	*
Stage 4	100.34	2.06	94.14	2.35	2.68	
Stage 5	98.18	2.32	90.05	1.71	3.10	*

* Significant at the 0.05 level of confidence.

GLOSSARY

GLOSSARY

Cephalometric Landmarks

Landmark:

1. Occipital point.

   The most posterior point on the occipital bone.

2. Porion.

   The mid-point on the upper edge of the external auditory meatus. As a cephalometric radiograph landmark it is located 4 millimeters directly superior to the mid-point of the metal ear rods.

3. Frontale.

   The most anterior point on the frontal bone determined by a perpendicular line from the SN line.

4. Nasion.

   The mid-point of the frontonasal suture at its most anterior margin.

5. Nasal tip.

   The most anterior inferior point on the nasal bones.

6. Orbitale.

   The deepest point on the infraorbital margin of the bony orbit.

7. Soft tissue nasion.

   The most anterior point on the soft tissue nose parallel to nasion.

8. Pronasale.

   The most anterior point on the contour of the soft tissue nose as measured from the N-Pog line.

9. Soft tissue A point.

   The most posterior point of the philtrum of the upper lip.

10. Labrale superius.

    The most prominent point on the upper lip measured perpendicular to the N-Pog line.

11. Stomion.

    The lowest point on the upper lip or the highest point on the lower lip (Burstone, 1952).

12. Labrale inferius.

    The most prominent point on the lower lip measured perpendicular to the N-Pog line.

13. Soft tissue B point.

    The most posterior point on the contour between the labrale inferius and the soft tissue pogonion.

14. Soft tissue pogonion.

    The most prominent point on the contour of the soft tissue covering of the chin.

15. Menton (Me).

    The most inferior point on the symphysis menti of the mandible.

16. Pogonion (Pog).

    The most anterior point on the contour of the chin.

17. B point (B).

    The deepest point on the midline contour of the mandible between infradentale and pogonion.

18. The apex of the mandibular central incisor.

19. The incisal edge of the mandibular central incisor.

20. The incisal edge of the maxillary central incisor.

21. The apex of the maxillary central incisor.

22. A point (A).

    The deepest point on the midline contour at the alveolar process between the anterior nasal spine and alveolar crest of the maxillary central incisor.

23. Anterior nasal spine (ANS).

    The median, sharp bony process of the maxilla at the lower margin of the anterior nasal opening.

24. Posterior nasal spine (PNS).

    The process formed by the united projecting ends of the posterior borders of the palatal processes of the palatal bones.

25. Pterygomaxillary fissure (PTM).

    The projected contour of the fissure formed by the anterior curvature of the pterygoid process and the posterior wall of the tuberosity of the maxilla. The cephalometric radiographic point is the most posterior point on the posterior wall of the maxillary tuberosity.

26. Sella (S).

    The centre of the sella turcica (pituitary fossa).

27. Basion (Ba).

    The most forward and lowest point on the anterior margin of the foramen magnum.

28. Articulare (Ar).

    The point of intersection of the external dorsal contour of the mandibular condyle and the temporal bone. The midpoint is used when the profile radiograph shows double projections of the rami.

29. The point of intersection of a tangent to the superior border of the odontoid process of Cervical 2.

30. The most inferior point on the posterior one third of the lower border of the mandible.

31. A point tangent to the anterior edge of the hyoid bone.

32. Distobuccal cusp tip of the maxillary left first molar.

33. Distobuccal cusp tip of the mandibular left first molar.

34. Gonion (Go).

    The lowest most posterior, and most outward point on the angle of the mandibular base line and the line tangent to the posterior border of the ramus.

35. The most inferior anterior point that can be identified on the tongue.

36. The tongue tip.

37. The highest point on the dorsum of the tongue.
38. That point on the tongue formed by drawing a line parallel to the palatal plane from the most anterior inferior point on the first cervical vertebra .
39. That point on the tongue formed by drawing a line parallel to the palatal plane from the most anterior inferior point on the second cervical vertebra .
40. That point on the tongue formed by drawing a line parallel from the most anterior inferior point on the third cervical vertebra .
41. That point on the posterior pharyngeal wall formed by drawing a line parallel to the palatal plane from the most anterior inferior point on the third cervical vertebra .
42. The most anterior inferior point on the third cervical vertebra .
43. The most anterior inferior point on the second cervical vertebra .
44. That point on the posterior pharyngeal wall formed by drawing a line parallel to the palatal plane from the most anterior inferior point on the second cervical vertebra .
45. The most anterior point on the anterior tubercle of the atlas.

46. That point on the posterior pharyngeal wall formed by drawing a line parallel to the palatal plane from the most anterior point on the anterior tubercle of the atlas.
47. That point on the first cervical vertebra formed by the posterior extension of the palatal plane.
48. That point on the posterior pharyngeal wall formed by the distal extension of the palatal plane.
49. The most posterior superior position on the condyle of the mandible.
50. That point on the cranial base formed by the intersection of a line from sella perpendicular to the palatal plane to the interior border of the cranial base.
51. That point on the posterior soft tissue wall of the nasopharynx by the intersection of a line from sella perpendicular to the palatal plane with the posterior soft tissue wall of the nasopharynx.
52. That point on the inferior surface of the cranial base formed by the extension of a vertical perpendicular from the posterior nasal spine to the inferior surface of the cranial base.
53. That soft tissue point adjacent to the cranial base formed by the extension of a vertical perpendicular from the posterior nasal spine to that soft tissue point.
54. The tip of the soft palate.

55. The point on the posterior surface of the soft palate at its greatest width.
56. That point on the anterior surface of the soft palate at its greatest width.
57. The highest point on the inferior surface of the hard palate.
58. The point of greatest curvature on the posterior surface of the premaxilla.
59. That point on the palatal plane formed by the vertical extension from "A" point (point 22) perpendicular to the palatal plane.
60. Origin.
    The reference point taken at sella (point 26).
61. Direction.
    The reference point nasion (point 4).

Cinefluorographic Landmarks

Fixed Landmarks:

A. Posterior nasal spine.
B. That point on the palatal plane formed by the vertical extension from "A" point perpendicular to the palatal plane.
C. The tip of the upper incisor.

Moveable Landmarks:

1. The most inferior anterior position on the anterior tubercle of the atlas.
2. The most inferior anterior position on the second cervical vertebra .
3. The most inferior anterior position on the third cervical vertebra .
4. That point on the posterior of the tongue formed by an extension of point "2" parallel to the palatal plane.
5. The tip of the soft palate.
6. The highest point on the dorsum of the tongue.
7. The anterior tip of the tongue.
8. The tip of the lower incisor.
9. The most inferior position on the upper lip.
10. The most superior position on the lower lip.
11. Pogonion Point.
    The most anterior point on the bony contour of the chin.
12. Menton.
    The most inferior point on the symphysis menti of the mandible.
13. The most anterior superior point on the hyoid bone.