



A LATERAL CEPHALOGRAPHIC EVALUATION OF ASSOCIATION OF CLEFT LIP AND PALATE WITH CERVICAL VERTEBRAL ANOMALIES

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Article Info: Received 02 January 2020; Accepted 28 January 2020

DOI: <https://doi.org/10.32553/ijmbs.v4i1.1026>

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Conflict of interest: No conflict of interest.

Abstract

Generally, orthodontic treatment and intervention are timed to take place before or during the peak growth velocity or pubertal growth spurt to achieve favorable effects in correcting sagittal, transverse, and vertical plane disharmonies. Skeletal maturity and growth spurts have been assessed by several methods in the literature, including chronological age, dental development, and sexual maturation characteristics. All of these methods have limitations, such as poor correlation with growth spurt. Hence based on above findings the present study was planned for A Lateral Cephalographic Evaluation of Association of Cleft Lip and Palate with Cervical Vertebral Anomalies.

The present study was planned in Department of Dentistry, Anugrah Narayan Magadh Medical College and Hospital, Gaya, Bihar, India. In the present study 20 cases of the age from 5 to 20 years suffered from the cleft lip and Palate were enrolled and evaluated with the different parameters. The radiographs were traced for cervical spine from C1- C4 on an acetate paper with 3H pencil under optimal illumination. Cervical vertebral anomalies were recorded and categorized into posterior arch deficiencies and fusion or both.

The data generated from the present study concludes that the association between cleft lip and palate and cervical vertebral anomalies indicates that cervical vertebral anomalies may be implicated as the etiology of cleft lip and palate. As oral and maxillofacial radiologists, lateral cephalographs are easily available in records of dental patients and can be used to identify cervical vertebral anomalies. Cervical vertebral anomalies can predispose to further disorders and identification of such anomalies mandates referral to the concerned specialist for appropriate management.

Keywords: Cleft Lip and Palate; Cervical Vertebral Anomalies; Dehiscence; Fusion; Lateral Cephalographs, etc.

Introduction

Orofacial clefts—including cleft lip (CL), cleft lip and palate (CLP), and cleft palate (CP) alone, as well as median, lateral (transversal), and oblique facial clefts—are among the most common congenital anomalies. [1] The incidence of orofacial cleft is approximately 1 in every 500-550 births. The prevalence varies by ethnicity, country, and socioeconomic status. Nonsyndromic CLP, the largest subgroup of craniofacial anomalies, occurs in 1.5-2.5 cases per 1000 live births. In the United States, 20 infants are born with an orofacial cleft on an average day (~7500 annually).

Children who have an orofacial cleft require several surgical procedures and multidisciplinary treatment and care; the conservative estimated lifetime medical cost for each child with an orofacial cleft is \$100,000, amounting to \$750 million for all children with

orofacial cleft born each year in the United States. [2] In addition, these children and their families often experience serious psychological problems. [3, 4]

With rapidly advancing knowledge in medical genetics and with new DNA diagnostic technologies, more cleft lip and palate anomalies are diagnosed antenatally and more orofacial clefts identified as syndromic. Although the basic rate of clefting (1:500 to 1:550) has not changed since Fogh-Andersen performed his pioneering 1942 genetic study distinguishing two basic categories of orofacial clefts—namely, CL with or without CP (CL/P) and CP alone [5]—these clefts can now be more accurately classified.

The correct diagnosis of a cleft anomaly is fundamental for treatment, for further genetic and etiopathologic studies, and for preventive measures

correctly targeting the category of preventable orofacial clefts.

Most individuals with CL, CP, or CLP, as well as many individuals with other craniofacial anomalies, require the coordinated care of providers in many fields of medicine (including otolaryngology) and dentistry, along with that of providers in speech pathology, audiology, genetics, nursing, mental health, and social medicine.

No single treatment concept has been identified, especially for a CLP. The timing of the individual procedures varies in different centers and with different specialists. In facial morphogenesis, neural crest cells migrate into the facial region, where they form the skeletal and connective tissue and all dental tissues except the enamel. Vascular endothelium and muscle are of mesodermal origin. [6]

The upper lip is derived from medial nasal and maxillary processes. Failure of merging between the medial nasal and maxillary processes at 5 weeks' gestation, on one or both sides, results in CL. CL usually occurs at the junction between the central and lateral parts of the upper lip on either side. The cleft may affect only the upper lip, or it may extend more deeply into the maxilla and the primary palate. (Cleft of the primary palate includes CL and cleft of the alveolus.) If the fusion of palatal shelves is impaired also, the CL is accompanied by CP, forming the CLP abnormality.

The secondary palate develops from the right and left palatal processes. Fusion of palatal shelves begins at 8 weeks' gestation and continues usually until 12 weeks' gestation. One hypothesis is that a threshold is noted beyond which delayed movement of palatal shelves does not allow closure to take place, and this results in a CP.

The group of orofacial cleft anomalies is heterogeneous. It comprises typical orofacial clefts (eg, CL, CLP, and CP) and atypical clefts (eg, median, transversal, oblique, and other Tessier types of facial clefts). [7, 8] Typical and atypical clefts can both occur as an isolated anomaly, as part of a sequence of a primary defect, or as a multiple congenital anomaly (MCA). In an MCA, the cleft anomaly could be part of a known monogenic syndrome, part of a chromosomal aberration, part of an association, or part of a complex of MCA of unknown etiology.

In those instances, genetic factors create a susceptibility for clefts. When environmental factors (ie, triggers) interact with a genetically susceptible genotype, a cleft develops during an early stage of development. The proportion of environmental and genetic factors varies with the sex of the individual affected with cleft. In CL and CP, it also varies with the severity and the unilaterality or bilaterality of the cleft anomaly; the highest proportion of genetic factors are in the subgroup of females with a bilateral cleft, and the smallest proportion is in the subgroup of males with a unilateral cleft. Thus, the classic multifactorial threshold (MFT) model of liability (see the first image below) can be applied to CL/P as the multifactorial model of liability with four different thresholds.

This model can facilitate understanding of differences in values of risk of recurrence as well as differences in prevention approaches between different subgroups of clefts. Theoretically, the subgroup of clefts closest to the population average should have the highest population prevalence, the lowest value of heritability, and thus the lowest risk of recurrence. This was confirmed in a large, population-based study of whites with clefts. [9]

The value of heritability expresses a ratio of genetic and nongenetic factors. Heritability is equal to 1 for conditions completely controlled by genetic factors and equal to 0 for conditions completely controlled by environmental factors.

A higher proportion of environmental factors indicates a lower risk of recurrence and also gives a better chance to act in prevention, because the only etiologic factors that can be changed are environmental factors. Thus, the subgroup whose average prevalence is closest to the population average represents males affected with a unilateral CL/P. This subgroup is most common among orofacial clefts; the risk of recurrence for siblings and for offspring of an individual with cleft is the lowest, the value of heritability is the lowest, and efficacy of primary prevention is the highest.

A cleft develops when embryonic parts called processes (which are programmed to grow, move, and join with each other to form an individual part of the embryo) do not reach each other in time and an open space (cleft) between them persists. In the normal situation, the processes grow into an open

space by means of cellular migration and multiplication, touch each other, and fuse together.

In general, any factor that could prevent the processes from reaching each other—for instance, by slowing down migration or multiplication of neural crest cells, by stopping tissue growth and development for a time, or by killing some cells that are already in that location—would cause a persistence of a cleft. Also, the epithelium that covers the mesenchyme may not undergo programmed cell death, so that fusion of processes cannot take place. [6]

Most children who have their clefts repaired early enough are able to have a happy youth and social life. Having a cleft palate/lip does not inevitably lead to a psychosocial problem. However, adolescents with cleft palate/lip are at an elevated risk for developing psychosocial problems especially those relating to self-concept, peer relationships and appearance. Adolescents may face psychosocial challenges but can find professional help if problems arise.[citation needed] A cleft palate/lip may impact an individual's self-esteem, social skills and behavior. There is research dedicated to the psychosocial development of individuals with cleft palate. Self-concept may be adversely affected by the presence of a cleft lip or cleft palate, particularly among girls. Negative outcomes can also be associated with the long durations of hospitalization. Psychological issues extend not just to the individual with CLP but also to their families, particularly their mothers, that experience varying levels of depression and anxiety. [10]

Research has shown that during the early preschool years (ages 3–5), children with cleft lip or cleft palate tend to have a self-concept that is similar to their peers without a cleft. However, as they grow older and their social interactions increase, children with clefts tend to report more dissatisfaction with peer relationships and higher levels of social anxiety. Experts conclude that this is probably due to the associated stigma of visible deformities and possible speech impediments. Children who are judged as attractive tend to be perceived as more intelligent, exhibit more positive social behaviors, and are treated more positively than children with cleft lip or cleft palate. [18] Children with clefts tend to report feelings of anger, sadness, fear, and alienation from

their peers, but these children were similar to their peers in regard to "how well they liked themselves."

The relationship between parental attitudes and a child's self-concept is crucial during the preschool years. It has been reported that elevated stress levels in mothers correlated with reduced social skills in their children. Strong parent support networks may help to prevent the development of negative self-concept in children with cleft palate. In the later preschool and early elementary years, the development of social skills is no longer only impacted by parental attitudes but is beginning to be shaped by their peers. A cleft lip or cleft palate may affect the behavior of preschoolers. Experts suggest that parents discuss with their children ways to handle negative social situations related to their cleft lip or cleft palate. A child who is entering school should learn the proper (and age-appropriate) terms related to the cleft. The ability to confidently explain the condition to others may limit feelings of awkwardness and embarrassment and reduce negative social experiences. [11]

As children reach adolescence, the period of time between age 13 and 19, the dynamics of the parent-child relationship change as peer groups are now the focus of attention. An adolescent with cleft lip or cleft palate will deal with the typical challenges faced by most of their peers including issues related to self-esteem, dating and social acceptance. Adolescents, however, view appearance as the most important characteristic, above intelligence and humor. This being the case, adolescents are susceptible to additional problems because they cannot hide their facial differences from their peers. Adolescent boys typically deal with issues relating to withdrawal, attention, thought, and internalizing problems, and may possibly develop anxiousness-depression and aggressive behaviors. [12] Adolescent girls are more likely to develop problems relating to self-concept and appearance. Individuals with cleft lip or cleft palate often deal with threats to their quality of life for multiple reasons including: unsuccessful social relationships, deviance in social appearance and multiple surgeries.

Due to lack of suction, an infant with a cleft may have trouble feeding. An infant with a cleft palate will have greater success feeding in a more upright position. Gravity will help prevent milk from coming through

the baby's nose if he/she has cleft palate. Gravity feeding can be accomplished by using specialized equipment, such as the Haberman Feeder. Another equipment commonly used for gravity feeding is a customized bottle with a combination of nipples and bottle inserts. A large hole, crosscut, or slit in the nipple, a protruding nipple and rhythmically squeezing the bottle insert can result in controllable flow to the infant without the stigma caused by specialized equipment.

Individuals with cleft also face many middle ear infections which may eventually lead to hearing loss. The Eustachian tubes and external ear canals may be angled or tortuous, leading to food or other contamination of a part of the body that is normally self-cleaning. Hearing is related to learning to speak. Babies with palatal clefts may have compromised hearing and therefore, if the baby cannot hear, it cannot try to mimic the sounds of speech. Thus, even before expressive language acquisition, the baby with the cleft palate is at risk for receptive language acquisition. Because the lips and palate are both used in pronunciation, individuals with cleft usually need the aid of a speech therapist. Tentative evidence has found that those with clefts perform less well at language. [13]

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Methodology:

The present study was planned in Department of Dentistry, Anugrah Narayan Magadh Medical College and Hospital, Gaya, Bihar, India. In the present study 20 cases of the age from 5 to 20 years suffered from the cleft lip and Palate were enrolled and evaluated with the different parameters. The radiographs were traced for cervical spine from C1- C4 on an acetate

paper with 3H pencil under optimal illumination. Cervical vertebral anomalies were recorded and categorized into posterior arch deficiencies and fusion or both.

All the patients were informed consents. The aim and the objective of the present study were conveyed to them. Approval of the institutional ethical committee was taken prior to conduct of this study.

Following was the inclusion and exclusion criteria for the present study.

Inclusion criteria: Lateral cephalographs of cleft lip and cleft palate patients. Lateral cephalometric radiographs extending upto the 4th cervical vertebrae and covering the entire anatomy of a cervical spine.

Exclusion criteria: Radiographs of syndromic patients, Deteriorated radiographs, etc

Results & Discussion:

Cleft lip or palate (or both) is considered a common congenital facial malformation, and its prevalence in Saudi Arabia ranges from 0.3 to 2.19 per 1000 live births. [14-15] In addition to their social difficulties [16], children with a cleft lip and/or palate (CLP) inherit multiple complications related to inadequate nutrition, feeding problems [17], and speech impairment. [18] The literature has shown that growth in general and craniofacial complex growth in particular could be affected in children with CLP, leading to marked skeletal discrepancies in all three planes of space. [19] Other complications include several occlusal and dental discrepancies. [19] Understanding craniofacial growth and development is essential for the comprehensive and successful management of these orthodontic patients. Such knowledge plays a crucial role in the diagnosis, treatment planning, outcomes, and overall stability of patient's orthodontic treatment. [20] Cleft patient treatment aims to address skeletal and dental disharmony through multidisciplinary care, where skeletal discrepancies in children with CLP may require orthopedic and/or surgical correction. [19]

The cervical vertebral column supporting the head comprises seven vertebrae. The first vertebra (C1) or atlas and the second vertebra or axis together form the superior or suboccipital segment connecting the spine to the occiput and involving a complex chain of joints. Fusion anomalies and posterior arch

deficiencies are the two important morphological deviations of cervical vertebrae according to Sandham. [23] Fusion anomalies can be fusion, block fusion, and occipitalization. Fusion is defined as the fusion of 1 unit with another at the articulation facets, neural arch, or transverse processes. Occipitalization is defined as the assimilation of the atlas (C1) either partially or completely with the occipital bone. Block fusion is a fusion of more than two vertebrae at the bodies, articulation facets, neural arch, or transverse processes. Posterior arch deficiency is partial cleft and dehiscence of the vertebrae according to Sandham.

Previous literature has reported anomalies of cervical vertebrae in many malocclusions. According to a study done by Sonnesen and Kjaer,[24] in 2007, deviations in the morphology of cervical vertebrae occurred significantly more often in the deep bite group compared with the control group (patients with neutral occlusion not requiring orthodontic treatment), fusion always occurred between C2 and C3. The incidence of posterior arch deficiency was also greater in skeletal deep bite patients. Sonnesen and Kjaer, in 2008,[25] concluded that associations were found between fusions of the cervical column and mandibular retrognathia, large cranial base angle, and large horizontal overjet, and between posterior arch deficiency and large maxillary inclination and cranial base angle. Sonnesen and Kjaer [26] evaluated the morphology of cervical column in Class III skeletal malocclusion and

concluded that the morphologic deviations of the cervical column occurred significantly more often in Class III (maxillary retrognathism) group (61.4%) not only compared with the control group but also compared with a group of patients with skeletal deep bite (41.5%). D'Attilio et al.[26] found a statistically significant correlation of over-jet, mandibular position and length, and mandibular plane angle to the cervical curvature.

Table 1: Demographic Details

Parameters	No. of Cases
Sex:	
Males	11
Females	9
Age:	
5 – 10 years	10
11 – 15 years	6
15 – 20 years	4

Table 2:

Parameters	No. of Cases
Unilateral cleft lip and palate	12
Bilateral cleft lip and palate	8
Total	20

Table 3: Prevalence of CVA in CLCP

Parameters	No. of Cases
Cervical Vertebral Anomaly	2
Cleft lip and Cleft palate	20



Fusion



Dehiscence

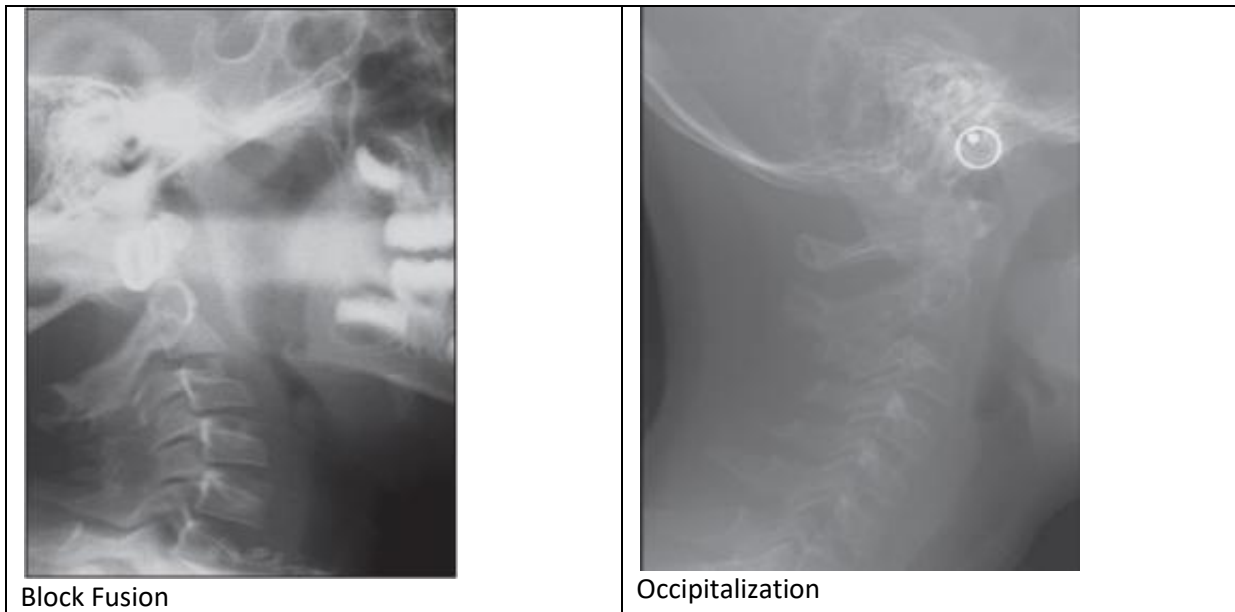


Figure: 1

Facial growth and development can be evaluated efficiently with the use of cephalometric radiographs. Cephalometric radiographs have been used from centuries to analyze dental and skeletal anomalies as well as soft tissue structures and form. [27] It is a relatively inexpensive method and permits a good assessment of the soft tissue elements of oral cavity, including the soft palate. Morphometric assessment of the soft palate can be carried out by measuring its length and height in the median sagittal plane on lateral cephalogram. [28]

Treatment is directed toward specific symptoms that present in each individual. Such treatment requires a coordinated multidisciplinary care which includes pediatricians, endocrinologists, surgeons, orthopedists, neurologists, cardiologists, otorhinolaryngologists, ophthalmologists, physiotherapists, occupational therapists and / or other doctors various specialties. Most affected individuals have a good prognosis if the syndrome is diagnosed early and the symptoms are treated in time. Activities that can cause neck injuries should be avoided.

The cervical vertebral anomalies are commonly divided into posterior arch deficiencies (PAD) and fusions (FUS). Posterior arch deficiencies are subdivided into spina bifida, which implies incomplete ossification in the spinous process and generally occurs in the posterior arch of the vertebral

unit, and dehiscence, which implies incomplete development of the structures. Dehiscence in the atlas affects either the anterior arch or the posterior arch, posterior arch dehiscence being most common in the midline. Fusion is a bony union of one unit with another at the articulation facets, neural arch, or transverse processes and may be subdivided into fusion between two cervical vertebrae; block fusion in which the bony union includes the vertebral bodies; and occipitalization, the assimilation of the atlas to the base of the skull or atlanto-occipital fusion or some degree of bony union between the skull and the atlas. [29]

Sonnesen et al. [30] had reported that the deviations in head posture and cranial base angle were sexually dimorphic, showing larger cervicohorizontal and cranial base angles in females than males. They also observed a positive correlation of cervical lordosis, inclination of upper cervical spine and cranial base angle in females with fusions of the cervical column, whereas this correlation was not found in males. Hence, it could be hypothesized that fusion anomalies show dimorphic pattern in their occurrence. However, Sonnesen and Kjaer [31] and Arntsen and Sonnesen [32] proposed that there was no significant gender difference in the occurrence of cervical vertebral anomalies.

Current literature suggests that variations of the cervical column morphology occur in healthy subjects

with neutral occlusion and normal craniofacial morphology as well as in orthodontic patients with deviating craniofacial morphology and severe malocclusion. A recent study found that the fusion between the upper cervical vertebrae (C2 and C3) occurred in 14.3% of healthy subjects. Fusions of the upper cervical column within that range are thus considered normal. A great deal of effort has been made to identify the association of cervical vertebral anomalies with abnormal craniofacial morphology, and it was found that the deviation of cervical vertebral morphology is associated with skeletal deep bite, skeletal open bite, skeletal maxillary overjet, skeletal mandibular overjet, as well as condylar hypoplasia. [33] Deviations occurred significantly more often in all five patient groups compared with the control group.

Conclusion:

The data generated from the present study concludes that the association between cleft lip and palate and cervical vertebral anomalies indicates that cervical vertebral anomalies may be implicated as the etiology of cleft lip and palate. As oral and maxillofacial radiologists, lateral cephalographs are easily available in records of dental patients and can be used to identify cervical vertebral anomalies. Cervical vertebral anomalies can predispose to further disorders and identification of such anomalies mandates referral to the concerned specialist for appropriate management.

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