

The association of cleft lip and palate with cervical vertebral anomalies- a lateral cephalographic study

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Abstract

Background: Cleft lip and palate is the most common major congenital craniofacial abnormality noted, which is 1 in 700 births. Although inheritance may play a role, cleft lip and palate is considered to be of multifactorial etiology, including chemical exposures, radiation, maternal hypoxia, teratogenic drugs, nutritional deficiencies, especially folic acid and cervical vertebral anomalies.

Objective: To assess an association between cleft lip and palate and cervical vertebral anomalies.

Methodology: The lateral cephalographs of 74 cleft lip and palate patients aged 6- 20yr were traced for cervical spine from C1- C4 on an acetate paper. Cervical vertebral anomalies were recorded and categorized into posterior arch deficiencies and fusion or both.

Results: 12.16% of cleft lip and palate patients had cervical vertebral anomalies.

Conclusion: The association between cleft lip and palate and cervical vertebral anomalies indicates that cervical vertebral anomalies may be implicated in the etiology of cleft lip and palate. 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

1. Introduction

Cleft lip and palate are the most common major congenital craniofacial abnormality noted, which is 1 in 700 births in India.[1] Cleft lip anomalies result from a deficiency of mesenchyme in the maxillary prominence and the median palatal process. Cleft palate anomalies result from defective development of the secondary palate and growth distortions of the lateral palatal processes, which prevent their fusion.[2]

Although inheritance may play a role, cleft lip and palate are considered to be of multifactorial etiology, including chemical exposures, radiation, maternal hypoxia, teratogenic drugs, nutritional deficiencies, especially folic acid and cervical vertebral anomalies.[1]

A functional developmental relationship has been hypothesized between cervical vertebrae and facial structures by (Lindsay & Ross 1965, p 279).

The cervical vertebrae develop from sclerotomes, which surround the notochord and neural tube. A series of cartilaginous rings appears within the sclerotomal paraxial mesodermal sheath at about four-week intrauterine life. Each cartilaginous ring is formed from adjacent halves caudal and cranial of the original somite; hence, vertebrae occupy intersegmental planes of the body wall. The cartilaginous arch fuses with the body at about 8 weeks for most vertebrae; each ring ossifies from three centers to form the centrum and two halves of the neural arch of the vertebrae.[5] Ossification commences by the 8th week of fetal life and is completed at about three years. The development of the cleft palate and Posterior arch deficiency (PAD) anomaly of a cervical vertebral spine is from the same paraxial mesoderm. The mechanism involved in palatal shelf fusion during embryonic development also has an

effect upon the development and fusion of the posterior arch of first cervical vertebra.[3]

In the eighth or ninth week of embryonic life, the tongue lies between the vertically oriented palatal shelves. Shortly before the formation of the secondary palate, the head is lifted from the pericardial region, and the mandible and tongue drop down permitting the palatal shelves to meet and fuse in the midline. When the cervical vertebrae are grossly abnormal, the neck remains compressed against the chest and thus the tongue continues to lie between the palatal shelves during the time when palatal closure would normally occur, resulting in cleft palate.[4]

Vertebral anomalies are commonly divided into posterior arch deficiencies and fusions. Posterior arch deficiencies are subdivided into spina bifida and dehiscence. Fusion anomalies may be subdivided into fusion between two cervical vertebrae, block fusion and occipitalization.[5]

2. Objective

To identify cervical vertebral anomalies on lateral cephalograph of cleft lip and palate subjects.

To assess an association between cleft lip and palate and cervical vertebral anomalies.

3. Materials and method

The lateral cephalographs of 74 cleft lip and palate patients aged 6- 20yr were selected from a Cleft lip and Palate Centre, Bangalore and Dept of Oral medicine and Radiology, MRADC, Bangalore.

3.1. 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.

3.2. Exclusion criteria

- Radiographs of syndromic patients
- Deteriorated radiographs

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.



Fig. 1: Dehiscence.



Fig. 2: Fusion C2-C3.

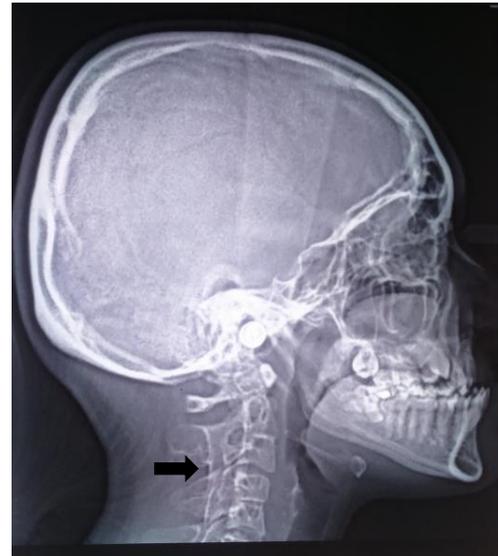


Fig. 3: Block Fusion.



Fig. 4: Occipitalisation.

4. Results

4.1. Age

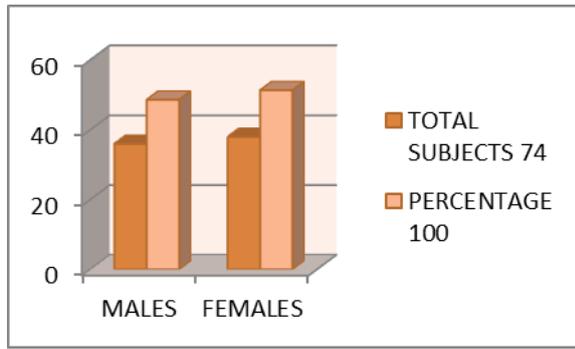
In the present study the mean age of the cleft lip and palate patients was found to be 13.15 yrs.

4.2. Gender

Out of 74 (100%) subjects 36(49%) were males and 38(51%) were females.

Table 1: Gender Distribution of CLCP

		Males	Females
Total subjects	74	36	38
Percentage	100	49	51



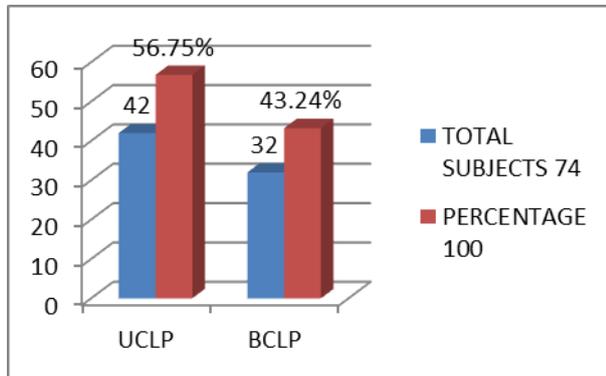
Graph. 1: Gender Distribution of CLCP.

4.3. Types of cleft lip and palate

Out of 74 (100%), 42(56.8%) had Unilateral CLP and 32(43.2%) had Bilateral CLP.

Table 2: Types of CLCP

Types	No. of subjects	Percentage
UCLP	42	56.8
BCLP	32	43.2
Total	74	100



Graph. 2: Types of CLCP.

Gender Distribution of Types of CLCP

19 (45%) males and 23(55%) females had UCLP. 17(53%) males and 15(47%) females had BCLP.

Table 3: A - Gender Distribution of UCLP

Types	Gender	No. of subjects	Percentage
UCLP	M	19	45
	F	23	55
Total		42	100

Table 3: b - Gender Distribution of BCLP

Types	Gender	No. of subjects	Percentage
BCLP	M	17	53
	F	15	47
Total		32	100



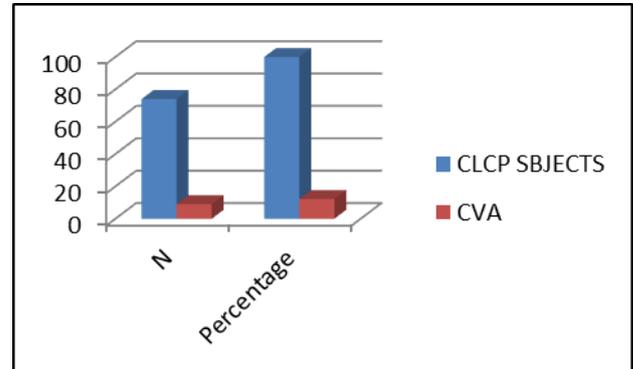
Graph. 3: Gender Distributions of Types of CLCP.

4.4. Cervical vertebral anomaly

Out of the 74(100%) CLCP subjects, 9(12.16%) of the subjects were found to have Cervical vertebral anomalies.

Table 4: Prevalence of CVA in CLCP

	No. of subjects	Percentage
CVA	9	12.16
CLCP	74	100



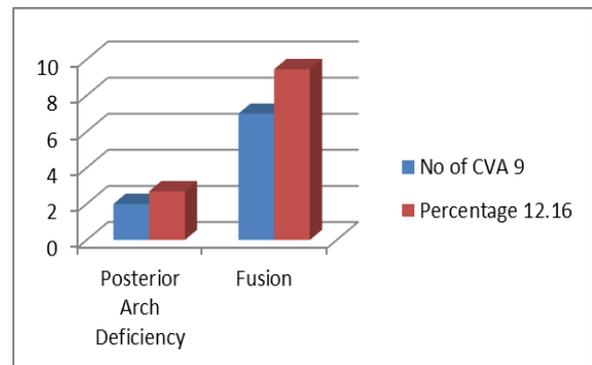
Graph. 4: Cervical Vertebral Anomaly.

Types of Cervical Vertebral Anomaly

Among the 9(12.16%) CVA, 2(2.70%) were posterior arch deficiencies which included 2 with dehiscence, and 7(9.45%) were fusion anomalies which included 5(6.75%) with fusion of c2 and c3, 1(1.35%) with block fusion and 1(1.35%) with occipitalisation.

Table 5: Types of CVA

		PAD	Fusion
CVA		2	7
Percentage		2.70	9.45



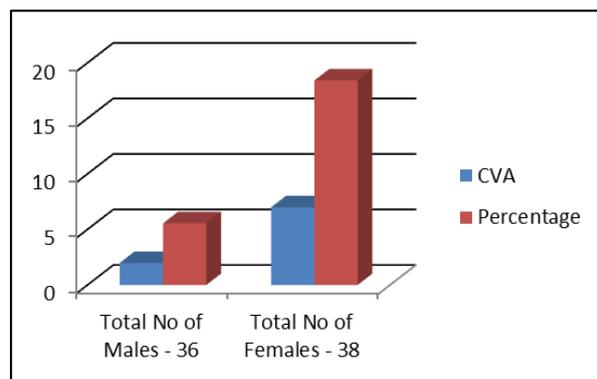
Graph. 5: Types of CVA.

Gender Distribution of Cervical Vertebral Anomaly

2 (2.7%) males and 7(9.46%) females were found to have CVA.

Table 6: Gender Distribution of CVA

	Gender	No. of Subjects	Percentage
CVA	M	2	2.7
	F	7	9.46
Total		9	12.16



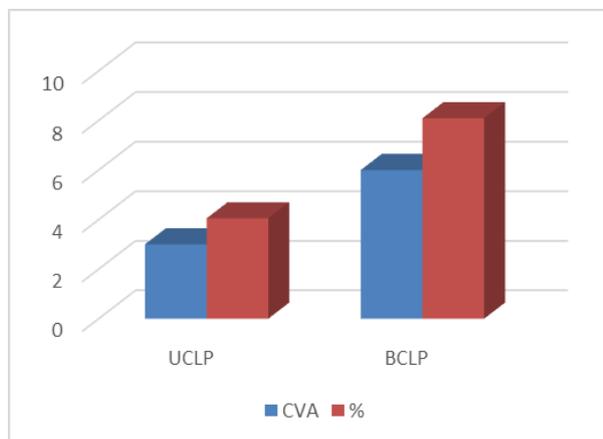
Graph. 6: Gender Distribution of CVA.

Cervical Vertebral Anomaly in UCLP and BCLP

Out of 9 (12.16%) CVA, 3(4.05%) anomalies were found in UCLP and 6(8.09%) anomalies were found in BCLP.

Table 7: Prevalence of CVA in UCLP and BCLP

Types	CVA	Percentage
UCLP	3	4.05
BCLP	6	8.09
Total	9	12.16



Graph. 7: Prevalence of CVA in UCLP and BCLP.

5. Discussion

Cleft lip and palate are common congenital malformations of the lip, palate, or both caused by complex genetic and environmental factors.[6] Genetic susceptibility has long been identified as a major component of CLP.[7] The most common environmental risk factors are maternal exposure to tobacco products, alcohols, nutritional deficiencies, some viral infections (rubella), medications, and teratogens in early pregnancy. Recognized teratogens included rare exposures such as phenytoin, valproic acid, thalidomide, and herbicides such as dioxin.[8] Other proposed risk factors include various occupational and chemical exposures, hyperthermia, stress, maternal obesity, oral hormone supplementation, ionizing radiation. Folic acid and zinc are also important in fetal development, and deficiency of this nutrients causes isolated cleft palate and other malformations. [7]

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 (Fig. 1), 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 (Fig.2) 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 (Fig.3) in which the bony union includes the vertebral bodies;

and occipitalization (Fig.4), 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. [9]

Age

In the present study the mean age of the cleft lip and palate patients was found to be 13.15 yrs.

Gender

Out of 74 (100%) subjects 36 (49%) were males and 38(51%) were females. In the present study, females were slightly more than males. Cleft lip occurs more common in male than female whereas cleft palate occurs more commonly in female than in male reason for this is that fusion of the palatine shelves 1 week later in girls than in boys is thought thus it could be one of the factors contributing in higher frequency of cleft palate in girls.[1]

Types of cleft lip and palate

Out of 74 (100%), 42(56.8%) had Unilateral CLP and 32(43.2%) had Bilateral CLP.

Gender Distribution of Types of CLCP

19(45%) males and 23(55%) females had UCLP. 17(53%) males and 15(47%) females had BCLP. In the present study, UCLP was more than BCLP. More females were found to have UCLP. More males were found to have BCLP.

Cervical vertebral anomaly

Out of the 74(100%) CLCP subjects, 9(12.16%) of the subjects were found to have Cervical vertebral anomalies. This finding was in concordance with (Osborne et al 1968, Sandham 1986, p.211)14.1% and 13.3% respectively. [10]

Types of a cervical vertebral anomalies

Among the 9(12.16%) CVA, 2(2.70%) were posterior arch deficiencies, which included 2 with dehiscence, and 7(9.45%) were fusion anomalies, which included 5(6.75%) with fusion of c2 and c3, 1(1.35%) with block fusion and 1(1.35%) with occipitalization.

In the present study fusion, anomalies were more than PAD. This finding was in agreement with (Ugar and Semb2001, p.501) and contrast with (Sandham 1986, Datana 2014). [11] We did not find any spina bifida. Vertebral fusion almost always occurs between C2 and C3. This finding might be of diagnostic importance since in children, the support of motion in the cervical spine is at the C2-C3 level, while in the adult cervical spine is at the C5-C6 level (Lustrin et al. 2003).[12]

Gender distribution of the cervical vertebral anomalies

2 (2.7%) males and 7(9.46%) females were found to have CVA. In the present study, more females were found to have cervical vertebral anomalies. This was consistent with (Ugar and Semb 2001, p.501) but no gender difference was found by (Giannakari 2004, Rajion et al 2006, Lima et al 2009). [13] [14] [15]

Cervical vertebral anomaly in UCLP and BCLP

Out of 9 (12.16%) CVA, 3(4.05%) anomalies were found in UCLP and 6(8.09%) anomalies were found in BCLP. In the present study, CVA was found to be more in BCLP, which was in correlation with study by (Srivastava et al 2014 p.45). [3]

The importance of diagnosis of CVA can be beneficial in understanding the embryonic phenomena underlying cleft development process. Early diagnosis of these pathologies on lateral cephalographs can offer valuable documentation of change due to injury and progression of the degenerative process. Cervical vertebral anomalies can predispose to further disorders such as cervical spondylosis, cervical spondylotic myelopathy and rheumatoid arthritis. It can also help in genetic counseling and recommendation for immediate and future treatment. [16]

6. Conclusion

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 our 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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