

Retrospective analysis of prevalence of cleft lip & cleft palate cases and their types at a tertiary care centre in Bihar

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Abstract

Introduction: Cleft lip and cleft palate (CL/P) are the most common congenital anomalies in the oral and maxillofacial region with a multi-factorial etiology, including environmental factors and genetic background.

Objectives: To assess the prevalence of cleft lip & cleft palate based on type, side, gender and associated syndromes.

Materials and methods: A total of 100 patients were included in the study. Patients were clinically evaluated and demographic factors related to diagnosed cases, including birth date, gender, birth weight, maternal age, number of pregnancies, type and side of cleft and presence of syndromes were recorded for analysis. The prevalence

was identified according to cleft type, side, gender and syndromes.

Results: Prevalence of cleft lip & palate was 55.0%. 27.0% had cleft palate and 18.0% had cleft lip.

Conclusions: Globally, extensive research on the exact etiological factors and epidemiological data is still required to explore the most applicable attempts to decrease the burden of the disease and to improve the quality of care provided for the affected individuals.

Keywords: Cleft lip, Cleft palate, Incomplete cleft lip/palate, Pierre Robin syndrome, Wardenburg Syndrome

Introduction

Cleft lip is defined as a congenital deformity that occurs in the primary palate which is located anteriorly to incisive foramen. Cleft palate is defined as a congenital

abnormality that occurs in the secondary palate (soft and hard palate).^{1,2} Both can be classified as unilateral, bilateral, complete or incomplete. Both genetics and environmental factors plays a role in etiology of clefts. Risk factors such as folic acid deficiency, maternal age, and maternal smoking have been linked to the development of clefts.³ CLP is accompanied by a wide variety of dental anomalies, which also have a long-term impact on the patient's facial anatomy and self-esteem.⁴ In addition to the aesthetic disfigurement, a child with cleft lip and/or palate suffers substantial functional morbidity such as restricted maxillofacial growth, speech abnormalities, engulfing and feeding difficulties, hearing impairment and/or recurrent ear infections.³ Most studies have also suggested that 70% of CL/P cases are non-syndromic, and that the remaining 30% are associated with structural abnormalities outside the cleft region (Schutte and Murray, 1999; Cobourne, 2004; Lidral et al., 2008).⁵

Clefts of the lip and palate (CL/P) are currently the most common craniofacial birth defects with overall incidence of approximately 1 in 700 live births.⁵ Even since long ago World Health Organization (WHO) has started its own surveys all over the world to analyze the data of such cases and how differently they occur in various countries. India is among one of the countries in the world where due to insufficiency of national registry of birth defects, the exact approximation of prevalence of such cases is not easy to obtain as major root of information are either hospital based surveys or studies.^{6,7}

Also, due to the inequality both in access to and qualities of cleft care with distinct differences among urban versus rural areas along with lack of awareness had lead to the accumulation of untreated clefts of the lip and palate leading to a significant health care problem in India.⁸ Very few studies have been conducted regarding prevalence of

cleft lip and palate in Bihar population Therefore, the purpose of the study is to analyze the prevalence of cleft lip & cleft palate cases and their types at a tertiary care centre in Bihar.

Material and methods

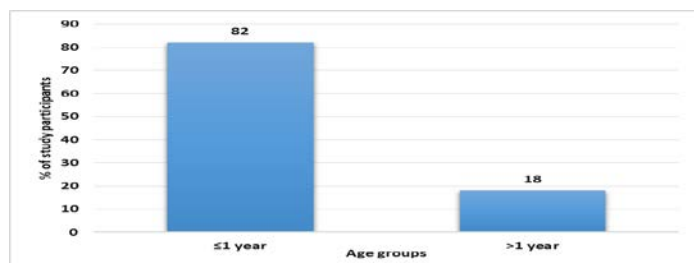
A retrospective study was conducted in outpatient department of Indira Gandhi Institute of Medical Sciences, Patna. A total of 100 patients were examined at duration of 3 months Out of which 58 were males & 42 were females patient. Before the commencement of survey, informed consent was taken from the parents. Clinical and demographic factors related to diagnosed cases, including birth date, birth type, gender, birth weight, maternal age, any drug usage, radiation exposure, number of children, presence of cleft in the family, number of pregnancies, type and side of cleft and presence of syndromes were recorded for analysis. All children with cleft lip and palate and associates syndromes were included in the study. Children with common dental problems were excluded from the study. Statistical analysis was carried out utilizing Chi-square test.

Results

Majority of the study subjects were below the age of 1 year (Table 1). (59.0%) were males and 41.0% were females (Table 2). Majority of the study subjects had cleft lip/palate (55.0%); 27.0% had cleft palate and 18.0% had cleft lip (Table 3). In majority of the subjects, the cleft was on left side (57.0%) and in 43.0% the cleft was on right side (Table 4). (63.0%) had unilateral cleft, followed by bilateral (36.0%) and Bilateral/lip, Unilateral/palate (1%), Incomplete cleft (50.0%) followed by complete (36.0%), sub mucous (13.0%) and there was one case of Operated cleft alveolus (Table 5). (94%) of the subjects did not have any associated syndromes, among those with associated symptoms, there were 2 cases each of Ectodermal Dysplasia and Pierre Robin syndrome; and 1

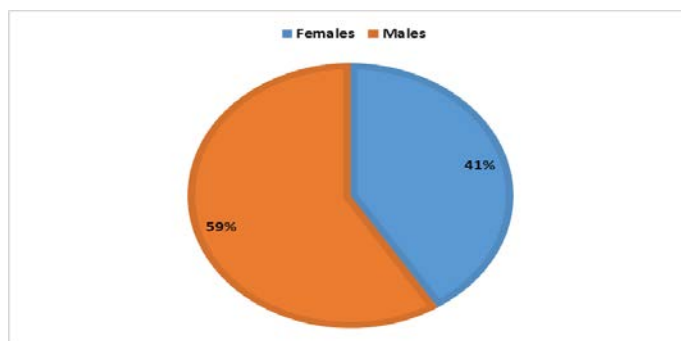
case each of Velo-cardio-facial syndrome and Waardenburg syndrome (Table 6). Among males & females, (61.0%), (46.3%) respectively had cleft lip/palate & the association was statistically significant (Table 7). (65.9%) females had cleft on left side, among males almost equal number of study subjects had cleft on left side (50.8%), no statistically significant association found (Table 8). Among males (61.0%) & females (65.9%), had bilateral cleft & there was no statistically significant association found (Table 9). (46.3%) females had complete cleft and among males (54.2%) had incomplete cleft & no statistically significant association found (Table 10). There was no statistically significant association found between gender & associated syndromes (Table 11).

| Table 1: Distribution of study population based on age groups | | | |
|---|---------|--------|----------------|
| | | Number | Percentage (%) |
| Age groups | <1 year | 82 | 82.0 |
| | >1 year | 18 | 18.0 |
| | Total | 100 | 100.0 |



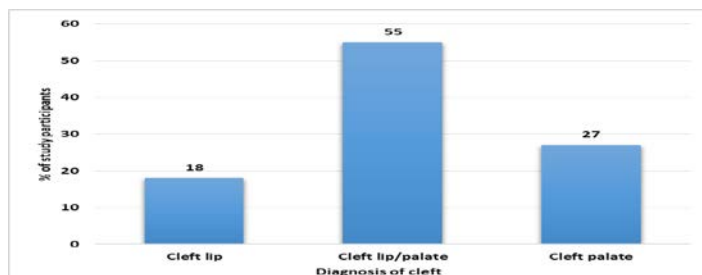
Graph 1: Distribution of study population based on age groups

| Table 2: Distribution of study population based on gender | | | |
|---|---------|--------|----------------|
| | | Number | Percentage (%) |
| Gender | Females | 41 | 41.0 |
| | Males | 59 | 59.0 |
| | Total | 100 | 100.0 |



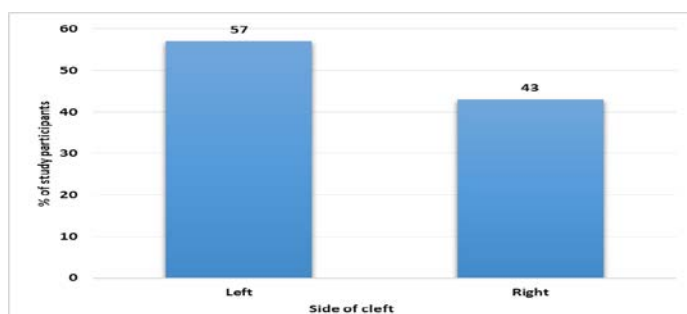
Graph 2: Distribution of study population based on gender

| Table 3: Distribution of study population based on diagnosis of cleft | | | |
|---|------------------|--------|----------------|
| | | Number | Percentage (%) |
| Diagnosis of cleft | Cleft lip | 18 | 18.0 |
| | Cleft lip/palate | 55 | 55.0 |
| | Cleft palate | 27 | 27.0 |
| | Total | 100 | 100.0 |



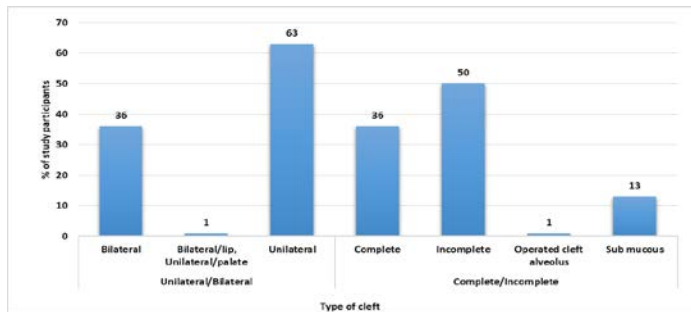
Graph 3: Distribution of study population based on diagnosis of cleft

| Table 4: Distribution of study population based on side of the cleft | | | |
|--|-------|--------|----------------|
| | | Number | Percentage (%) |
| Side | Left | 57 | 57.0 |
| | Right | 43 | 43.0 |
| | Total | 100 | 100.0 |



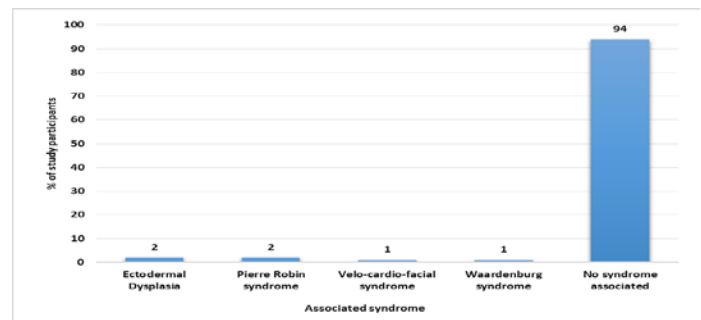
Graph 4: Distribution of study population based on side of the cleft

| Type of cleft | | Number | Percentage (%) |
|----------------------|----------------------------------|--------|----------------|
| Unilateral/Bilateral | Bilateral | 36 | 36.0 |
| | Bilateral/lip, Unilateral/palate | 1 | 1.0 |
| | Unilateral | 63 | 63.0 |
| Complete/Incomplete | Complete | 36 | 36.0 |
| | Incomplete | 50 | 50.0 |
| | Operated cleft alveolus | 1 | 1.0 |
| | Sub mucous | 13 | 13.0 |
| Total | | 100 | 100.0 |



Graph 5: Distribution of study population based on type of cleft

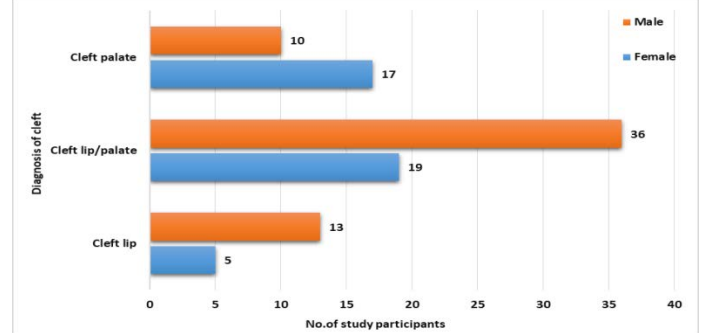
| Associated syndrome | | Number | Percentage (%) |
|---------------------|-----------------------------|--------|----------------|
| Associated syndrome | Ectodermal Dysplasia | 2 | 2.0 |
| | Pierre Robin syndrome | 2 | 2.0 |
| | Velo-cardio-facial syndrome | 1 | 1.0 |
| | Waardenburg syndrome | 1 | 1.0 |
| | No syndrome associated | 94 | 94.0 |
| Total | | 100 | 100.0 |



Graph 6: Distribution of study population based on associated syndrome

| Diagnosis of cleft | n | Gender | | Chi-square | P |
|--------------------|--------|--------|-------|------------------------------------|---|
| | | Female | Male | | |
| Cleft lip | 5 | 13 | 7.632 | 0.02* Statistically significant | |
| | 12.2% | 22.0% | | | |
| Cleft lip/palate | 19 | 36 | | | |
| | 46.3% | 61.0% | | | |
| Cleft palate | 17 | 10 | | | |
| | 41.5% | 16.9% | | | |
| Total | 41 | 59 | | | |
| | 100.0% | 100.0% | | | |

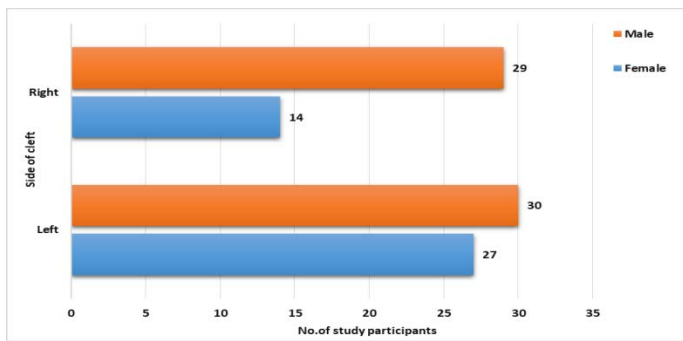
Chi-square test; * Statistically significant; p<0.05, NS- Not significant



Graph 7: Association of gender and diagnosis of cleft.

| Side of cleft | n | Gender | | Chi-square | P |
|---------------|--------|--------|-------|------------|---|
| | | Female | Male | | |
| Left | 27 | 30 | 2.222 | 0.09 NS | |
| | 65.9% | 50.8% | | | |
| Right | 14 | 29 | | | |
| | 34.1% | 49.2% | | | |
| Total | 41 | 59 | | | |
| | 100.0% | 100.0% | | | |

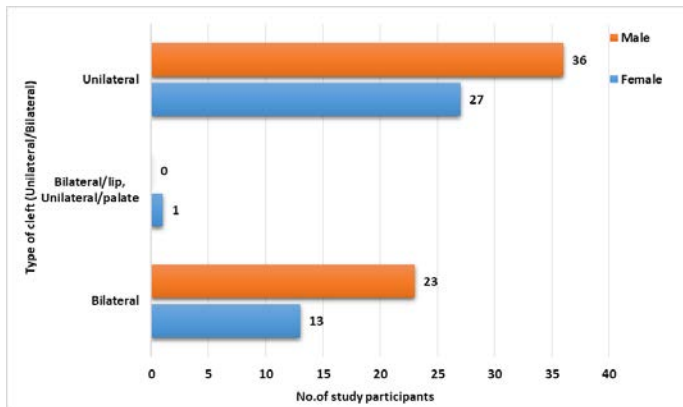
Chi-square test; * Statistically significant; p<0.05, NS- Not significant



Graph 8: Association of gender and side of cleft

| Type of cleft (Unilateral/Bilateral) | | Gender | | Chi-square | P |
|--------------------------------------|---|--------|--------|------------|------------|
| | | Female | Male | | |
| Bilateral | n | 13 | 23 | 1.885 | 0.39 NS |
| | % | 31.7% | 39.0% | | |
| Bilateral/lip, Unilateral/palate | n | 1 | 0 | | |
| | % | 2.4% | 0.0% | | |
| Unilateral | n | 27 | 36 | | |
| | % | 65.9% | 61.0% | | |
| Total | n | 41 | 59 | | |
| | % | 100.0% | 100.0% | | |

Chi-square test; * Statistically significant; p<0.05, NS- Not significant

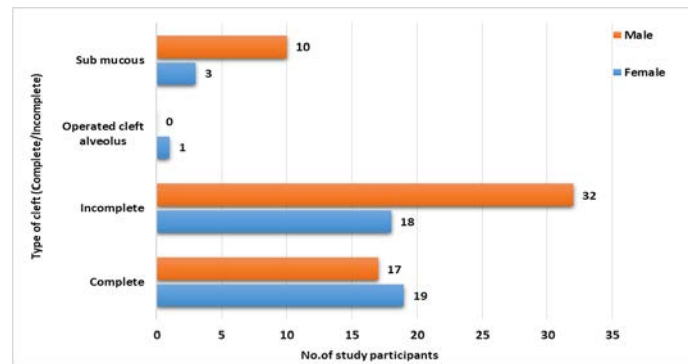


Graph 9: Association of gender and type of cleft (Unilateral/Bilateral)

| Type of cleft (Complete/Incomplete) | n | Gender | | Chi-square | P |
|-------------------------------------|----|--------|-------|------------|---|
| | | Female | Male | | |
| Complete | 19 | 17 | 5.747 | 0.12 | |

| | % | 46.3% | 28.8% | NS |
|-------------------------|---|--------|--------|----|
| Incomplete | n | 18 | 32 | |
| | % | 43.9% | 54.2% | |
| Operated cleft alveolus | n | 1 | 0 | |
| | % | 2.4% | 0.0% | |
| Sub mucous | n | 3 | 10 | |
| | % | 7.3% | 16.9% | |
| Total | n | 41 | 59 | |
| | % | 100.0% | 100.0% | |

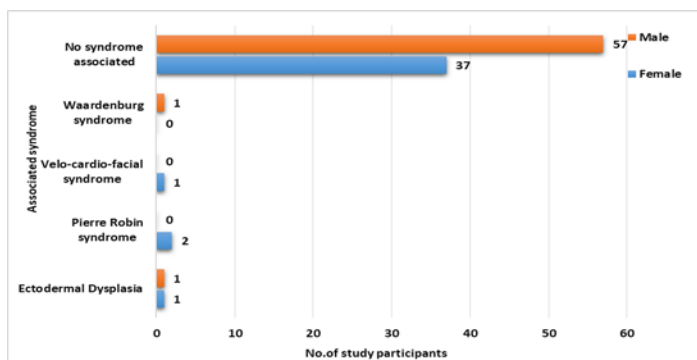
Chi-square test; * Statistically significant; p<0.05, NS- Not significant



Graph 10: Association of gender and type of cleft (Complete/Incomplete)

| Associated syndrome | | Gender | | Chi-square | P |
|-----------------------------|---|--------|--------|------------|------------|
| | | Female | Male | | |
| Ectodermal Dysplasia | N | 1 | 1 | 5.183 | 0.27 NS |
| | % | 2.4% | 1.7% | | |
| Pierre Robin syndrome | N | 2 | 0 | | |
| | % | 4.9% | 0.0% | | |
| Velo-cardio-facial syndrome | N | 1 | 0 | | |
| | % | 2.4% | 0.0% | | |
| Waardenburg syndrome | N | 0 | 1 | | |
| | % | 0.0% | 1.7% | | |
| No syndrome associated | N | 37 | 57 | | |
| | % | 90.2% | 96.6% | | |
| Total | N | 41 | 59 | | |
| | % | 100.0% | 100.0% | | |

Chi-square test; * Statistically significant; p<0.05, NS- Not significant



Graph 11: Association of gender and associated syndrome

Discussion

Cleft lip and palate are the most common facial deformity and clefting may involve lip only, lip and palate and palate only. Environmental & genetic factors (such as smoking, alcohol, poor nutrition, familial factors and chromosomes) are the main reasons of clefting in infants. OFCs impact a considerable proportion of the global society. It affects around 1.5 per 1,000 live births (about 220,000 new cases per year), with wide variation across geographic areas and ethnic groups, with substantial evidence of both health inequality and inequity.³ For the present study, we have considered the Bihar population. Total 100 cases of cleft lip and palate were included in the study. In the present study frequency of combined cleft lip and palate is highest followed by cleft palate than cleft lip. Similar results were also shown by (Parihar et al).⁹ whereas, (Khajanchi et al) found highest cases of cleft lip followed by combined clefts in Gujrat population.¹⁰ This variation in incidences could be attributed to diversities of the region. Clefts are polygenic in nature and considered as outcome of multiple factors involving environmental and hereditary.

In the present study majority of the subjects were males (59.0%) and 41.0% were females. In comparison with females, males are affected more often and show more severe clefting. This was in agreement with the findings of (Fogh-Andersen, 1967; Natsume et al., 1988; Conway and Wagner, 1966; Christensen, 1999; Cooper et al., 2000; Al-Balkhi, 2008).^{5,11,12,13,14,15,16}

In majority of the subjects, the prevalence of cleft was more common on left side 57.0% than on right side 43.0%. The study done by (M. Okan Akcam, Desai BB, Dewinter et al.) reported the left side is more affected than the right side with (ratio 2:1).^{17,18,19}

Majority of the subjects had unilateral cleft 63.0%, followed by bilateral 36.0% and Bilateral/lip, Unilateral/palate 1%. 50.0% had incomplete cleft followed by complete 36.0%, sub mucous 13.0% and there was one case of Operated cleft alveolus. Study done by (Haque S) also reported highest prevalence of unilateral cleft than bilateral cleft.⁴

In the present study 94% of the subjects did not have any associated syndromes. Among those with associated syndromes, there were 2 cases each of Ectodermal Dysplasia and Pierre Robin syndrome; and 1 case each of Velo-cardio-facial syndrome and Waardenburg syndrome. The study done by (Hadadi AL) showed 19% patients with associated syndromes which was quite higher.²⁰

Among females & males, majority of the study subjects had bilateral cleft 65.9%, 61.0% respectively. There was no statistically significant association found.

Conclusion

In India, more surveys and research by the various organization that provides medical services for cleft lip and palate patients, availability of care, effects of previous surveys and importance of specific management techniques in bringing about improvement in the quality health care for OFC is required.

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