Original Article JCMCTA 2023 ; 34 (2) : 107-110

# Associated Anomalies in Children with Cleft Lip and Palate in Bangladesh

Sharmin Fatema[1*] Asma Mostofa[2] Sarah Fatima Sumaiya[3] Md Ashrafuzzaman[4]

## Abstract

**Background:** Cleft lip and palate are the most frequent orofacial congenital defects that impact child health and survival. The incidence and types of cleft lip and palate vary across regions and countries. These children may potentially have associated anomalies that complicate their care and prognosis. The purpose of this study was to identify the associated anomalies linked to cleft lip and palate defects in Bangladeshi children.

**Materials and methods:** This cross-sectional observational study was conducted at Chittagong Medical College's Department of Anatomy between April 2021 and March 2022. A predesigned questionnaire was used to gather socio-demographic data, cleft type, and the existence of any associated anomalies from 100 children with cleft defects.

**Results:** There were 58 males and 42 females among the 100 children. A cardiac abnormality was the most common (31%), followed by a mixed anomaly (25%), in which more than one aberration coexisted in a child with a cleft lip and palate. Skeletal, ocular, ear and CNS anomalies were among the others. Regarding related malformations, there was no significant difference between male and female offspring. The presence of associated anomalies had no significant relationship with the types of cleft deformity in the Chi-square test. An equal number of male and female children had associated anomalies. Children with isolated cleft palate had the highest percentage of anomalies (27.3%), followed by those with combined cleft lip and palate (17.2%).

**Conclusion:** Before undergoing surgical repair, children with cleft lip and palate should have a thorough physical examination and necessary baseline investigations to rule out any associated anomalies. This would benefit their results of intervention and quality of life.

1. Lecturer of Anatomy
Chattagram Maa-O-Shishu Hospital Medical College, Chattogram.
2. Professor of Anatomy
Chattagram Maa-O-Shishu Hospital Medical College, Chattogram.
3. Lecturer of Oral Microbiology
Chittagong Medical College, Chattogram.
4. Professor of Anatomy
Chittagong Medical College, Chattogram.

***Correspondence: Dr. Sharmin Fatema**
*Cell : 01815 50 51 22*
*E-mail: sfatema10@gmail.com*

*Submitted on : 22.05.2023*
*Accepted on : 21.10.2023*

**Key words:** Associated anomaly; Cleft Lip (CL); Cleft Palate (CP); combined Cleft Lip and Palate (CLP).

## Introduction

Congenital defects are a prominent cause of infant mortality and disability, especially in developing nations. Every year, approximately 8 million infants are born with serious congenital anomalies.[1] One of the most common congenital malformations of the head and neck area is orofacial cleft.[2] Cleft Lip (CL), Cleft Palate (CP) and combined Cleft Lip and Palate (CLP) are the typical orofacial clefts.[3-5] It can appear as a single defect or in conjunction with other congenital anomalies.[6-8] Cleft defects are a characteristic of more than 300 syndromes.[3] Depending on the sample size, geographic region and ethnic background of different studies, the incidence of associated anomalies ranged from 1.5% to 64.2%.[8-9]

Cleft defects have a complicated etiology and pathophysiology that involves both genetic and environmental factors. In Bangladesh, there is a scarcity of data on the link of cleft defects with other congenital anomalies. Furthermore, no study has been conducted on the relationship between the kind of cleft and the associated anomalies in Bangladeshi children. Understanding this link is critical for understanding the embryonic processes behind the malformation process and providing preventative methods and genetic counseling to patients and their families.

The purpose of this study was to identify the associated anomalies linked to cleft lip and palate defects in Bangladeshi children.

## Materials and methods

A total of 100 children with CL, CP, or CLP were included in this cross-sectional observational research. Between April 2021 and March 2022, the research was conducted at Chittagong Medical College's Anatomy Department. After gaining clearance from Chittagong Medical College's ethical review committee, the study subjects were recruited based on the enrollment criteria.

Children with atypical clefts were omitted from the research even though they had related defects. Study participants were selected from Smile Train, Chattagram Maa Shishu O General Hospital Cleft Project and LMRF Children's Hospital, Chittagong Lion's Foundation Complex. Only one child was from Chattagram Maa Shishu O General Hospital Paediatric Medicine department who was hospitalized for a respiratory tract infection. The parents of the children were given a full explanation of the study's goal and procedure. The children's respective parents or guardians provided written approval. A predesigned questionnaire was used to collect demographic information, cleft type, and related abnormalities. History taking, physical examinations, hospital record reviews, and accessible investigation reports were used to obtain data. The Chi-square test was performed to determine significance. SPSS Statistics for Windows, version 26.0, was used for all statistical analyses. The data was examined, and the results were presented in the form of tables and figures. Before commence the study necessary permission was obtained from the proper authorities.

## Results

The research included 100 children under the age of 18 who had CL, CP, or CLP. The ages of the participants ranged from one month to seventeen years. With 58 (58%) males and 42 (42%) females, the male-to-female ratio was 1.38:1. The participants' average age was 24.340.1 months.

The incidence and features of several types of clefts are shown in Table I. The frequent type was CLP (68%), followed by CP (22%), and CL (10%). CLP was more common in male children, whereas CL and CP were more common in female children. CL and CLP were both more common on the left side than on the right. 16 (16%) of the 100 children had related abnormalities. 6 of them had CP, while 10 had CLP.

**Table I** Characteristics of the Cleft lip and Palate in the study population (n=100)

| Pattern of clefts (n & %) | | CL 10 | CP 22 | CLP 68 |
|---|---|---|---|---|
| Gender (n) | Male | 4 | 9 | 45 |
| | Female | 6 | 13 | 23 |
| Laterality (%) | Right-sided | 20 | — | 24 |
| | Left-sided | 80 | — | 44 |
| | Bilateral | — | — | 32 |
| Occurrence (n) | Complete | 2 | 12 | 68 |
| | Incomplete | 8 | 10 | 0 |
| Associated anomaly | With | 0 | 6 | 10 |
| | Without | 0 | 16 | 58 |

Figure 1 depicts the distribution of associated anomalies across research participants. Cardiac anomaly was the most frequent form, affecting 5 (31%) of the infants, followed by a mixed anomaly, affecting 4 (25%) of the children. Skeletal and vascular anomalies impacted two (13%) of the children, whereas ocular, ear, and CNS defects affected one (6%) of the children.

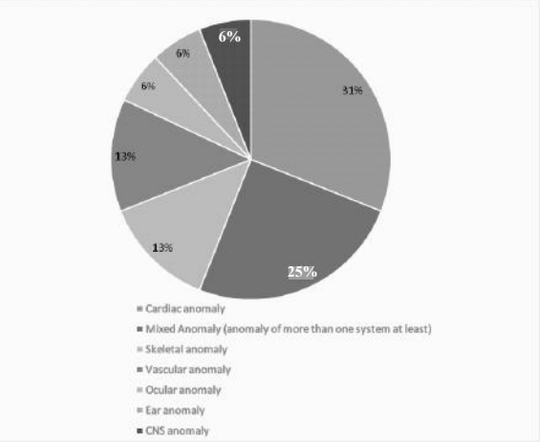


**Figure 1** Associated anomalies among the study subjects (n=16)

A brief detail about the associated anomaly with a description of the cleft subtype and gender is highlighted in Table II.

**Table II** Associated anomalies of different organ systems among the study subjects according to cleft pattern

| Associated anomaly | Description of the anomaly | Brief description of the pattern of cleft defect | Gender |
|---|---|---|---|
| Cardiac anomaly | Atrial septal defect | Complete cleft palate | Male |
| | | Bilateral complete cleft lip, palate, and alveolus | Female |
| | Patent ductus arteriosus | Bilateral complete cleft lip, palate and alveolus | Male |
| | Ventricular septal defect | Bilateral complete cleft lip, palate and alveolus | Female |
| Mixed anomaly | Atrial septal defect and club foot | Complete cleft palate | Male |
| | Umbilical hernia with syndactyly in both legs | Incomplete cleft palate | Male |
| | External ear deformity, preauricular skin tag, tongue tie | Complete cleft palate | Male |
| | Absence of right eyeball, preauricular skin tag, skin tags on cheek | Right-Sided complete cleft lip, palate, and alveolus | Female |
| Skeletal anomaly | Incomplete left lower limb below knee with only 3 toes which are abnormally long. | Complete cleft palate | Female |
| | Polydactyly,6 toes in both legs | Right-sided complete cleft lip, palate, and alveolus | Male |

| Associated anomaly | Description of the anomaly | Brief description of the pattern of cleft defect | Gender |
|---|---|---|---|
| Vascular anomaly | Haemangioma on forehead | Incomplete cleft palate | Female |
| | Haemangioma on the left hand (Both palmar and dorsal aspect) | Left-sided complete cleft lip, palate and alveolus. | Male |
| Ocular anomaly | Bilateral absence of eyeball | Right sided complete cleft lip, palate, and alveolus | Female |
| Ear anomaly | Preauricular skin tag | Bilateral complete cleft lip, palate and alveolus | Female |
| CNS anomaly | Does not follow verbal commands, no eye contact | Complete cleft palate | Female |

Table II includes a brief explanation of the related abnormality, as well as a description of the cleft subtype and gender.

**Table III** Relationship between the cleft pattern and associated anomalies (n=100)

| Type of deformity | Associated anomalies | | p value |
|---|---|---|---|
| | Yes | No | |
| Cleft lip | 0 (0%) | 10 (100%) | 0.15 Ns |
| Cleft Palate | 6 (27.3%) | 16 (72.7%) | 0.10 Ns |
| Cleft lip and palate | 10 (14.7%) | 58 (85.3%) | 0.61 Ns |

Table III depicts the link between the cleft pattern and the associated anomalies. It was discovered that associated anomalies are more common in children with CP defects (27.3%) than in children with CLP (17.2%) and none with CL. A chi-square test, however, revealed no statistically significant change.

## Discussion

The current study included 100 children under the age of 18, with 58 males and 42 females, for a male-to-female ratio of 1.38:1. The ages of the respondents varied from one month to seventeen years, with a mean age of 24.340.1 months.

CLP was used by the majority of responders (68%) followed by CP (22%), and CL (10%). 16 (16%) of the children had associated anomalies. The current study's findings were almost identical to those of previous studies in India (14.8%), Ethiopia (11%), and Pakistan (10.7%).[10-12] However, some investigations found greater percentages, such as 21% by Milerad et al. 29% by Shafi et al. 29.5% by Aljohar et al. 31.2% by Pereira et al. 36.7% by Stoll et al. discovered cardiac abnormality to be the most frequent associated anomalies, with l., and 63.4% by Sphrintzen et al.[13-18]

In the current study, the most common abnormality was cardiac anomaly (31%), followed by mixed anomaly (25%), skeletal anomaly (13%), vascular anomaly (13%), and ear, ocular, and CNS abnormalities (6% each). Research conducted in Pakistan in 2003 and Saudi Arabia in 251% and 38.2%, respectively. Bekele et al., Pereira et al. and Milerad et al. found that the cardiac abnormality was the second most common in their studies.[14,15,11,16,13]

In this study, associated anomalies were observed to be more common in cases of CLP problems. 6 of the 16 children with associated anomalies had CP, whereas 10 had CLP. There were no associated anomalies in any of the CL instances. When the Chi-square test was used, there was no significant difference between the types of cleft defect and the associated anomalies. Similarly, 17 incidences of associated anomalies were discovered among 159 children with cleft deformities in Pakistani research.[12] They discovered 8 instances of CP out of 50 cases, 9 cases of CLP out of 82 cases, and no cases of CL out of 27. An Italian research published in 2019 discovered that related abnormalities were more common with CLP (50%) followed by CP (35%) and CL (15%).

In contrast to the current study, associated anomalies with CP were more common in a Portuguese study published in 2018 and a Saudi Arabian study published in 2008.[15,16]

Differing research results may change due to differences in sample size, sampling procedures, racial and ethnic variance, how long after birth the cases were evaluated, or differing inclusion and exclusion criteria.

## Limitation

The sample size of the study was relatively small and obtained only from Chattogram.

## Conclusion

This study revealed that some associated anomalies were found among the children with CP and CLP defects. So careful history taking, routine physical examination and relevant investigations should be carried out prior to surgical correction in children with cleft defects.

**Recommendation**

To increase the sample size, countrywide research involving the country's indigenous people should be explored. Clinical geneticists' participation, as well as a complete baseline and specialized investigation setup, including karyotyping facilities, will result in more reliable results.

**Acknowledgment**

All the authors express their gratitude to all, who assisted them on this path.

**Contribution of authors**

SF-Acquisition of data, data analysis, drafting & final approval.
AM-Conception, critical revision & final approval.
SFS-Data analysis, drafting & final approval.
MA-Design, interpretation of data, critical revision & final approval.

**Disclosure**

The authors declared no conflicts of interest.

**References**

**1.** Walani S, Pachon H, Hiebert L, Mwaisaka R. Prevention of birth defects in East Africa: A review of national politics. African Journal of Food, Agriculture, Nutrition and Development. 2020;20(3):15740-15763.

**2.** https://www.cdc.gov/ncbddd/birthdefects/ surveillanc-manual/chapters/chapter-4/chapter4-6.html.

**3.** Williams NS, Bulstrode CJK, O'Connell PR editors. Bailey & Love's Short practice of Impellizzeri A, Giannantoni I, Polimeni A, Barbato E, Galluccio G. EpidemiologSurgery. 27th ed. Florida: CRS press, Taylor and Francis group. 2018.

**4.** ical characteristic of Orofacial clefts and its associated congenital anomalies: Retrospective study. BMC Oral Health. 2019;19(1):1-4.

**5.** Calzolari E, Bianchi F, Rubini M, et al. EUROCAT Working Group. Epidemiology of cleft palate in Europe: implication for genetic research. Cleft Palate Craniofac J. 2004; 41:244-249.

**6.** Tolarova MM & Cervenka J. Classification and birth prevalence of Orofacial clefts. Am J Med Genet. 1998; 75:126-137.

**7.** Tessier P, Anatomical Classification of facial, cranio-facial and latero-facial clefts. J maxillofac surg. 1976; 4: 69-92.

**8.** Calzolari E, Pierini A, Astolfi G, et al. Associated anomalies in multi malformed infants with cleft lip and palate: An epidemiologic study of nearly 6 million births in 23 EUROCAT registries. Am J Med Genet A. 2007;143A:528-537.

**9.** Hagberg C, Larson O, Milerad J. Incidence of cleft lip and palate and risks of additional malformations. Cleft Palate Craniofac J. 1989; 35:40-45.

**10.** Sekhon PS, Ethunandan M, Markus AF, Krishnan G, Rao CB. Congenital Anomalies Associated with Cleft Lip and Palate- An Analysis of 1623 Consecutive Patients. Cleft Palate Craniofac J. 2011;48(4):371-378.

**11.** Bekele KK, Ekanem PE, Meberate B. Anatomical patterns of cleft lip and palate deformities among neonates in Mekelle, Tigary, Ethiopia: Implication of environmental impact. BMC Pediatrics. 2019; 19(1): 1-7.

**12.** Khan M, Hidayet U, Shazia N, Tahmeed U, Hafeezullah K, Muhammad T, et al. Patterns of Cleft Lip and Cleft Palate in Northern Pakistan. Archives of Clinical & Surgery. 2012; 1(2):63-71.

**13.** Milerad J, Larson O, Hagberg C, et al. Associated malformations in infants with cleft lip and palate; a prospective, population-based study. Pediatrics. 1997;100;180-186.

**14.** Shafi T, Khan MR, Atiq M. Congenital heart disease and associated malformations in Children with cleft lip and palate in Pakistan. Br J Plast Surg. 2003; 56:106-109.

**15.** Aljohar A, Ravichandran K, Subhani S. Patterns of Cleft Lip and Palate in Hospital in Hospital-Based Population in Saudi Arabia : Retrospective Study. Cleft Palate Craniofac J. 2008; 45(6): 592-596.

**16.** Pereira AV, Fradinho N, Carmo S, de Sousa JM, Rasterio D, Duarte R et al. Associated Malformations in Children with Orofacial Clefts in Portugal: A 31-year study. International Open Access Journal of the American Society of Plastic Surgeons. 2018; 6(2):1-7.

**17.** Stoll C, Alembik Y, Dott B, et al. Associated malformations in cases with oral clefts. Cleft Palate Craniofac J. 2000; 37:41-47.

**18.** Shprintzen RJ, Siegel-Sadewitz VL, Amato J, et al. Anomalies associated with cleft lip, cleft palate, or both. Am J of Med Genet. 1985; 20:585-595.