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Original Research Article

Ocular Anomalies in Patients of Cleft Lip and Palate in North Indian Population: An Observational Study

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Abstract

Introduction and objective: All types of cleft-associated anomalies do not occur with equal frequency. It has not been well-known if specific types of anomalies are commonly related with clefts, or which organ is most commonly affected. Cleft lip and palate (CL and P) could be associated with many other structural abnormalities of the adjacent vital structures of the face. This study aimed to identify the ocular anomalies in patients with CL and P in north Indian population.

Methods: Three hundred seventy consecutive syndromic and non-syndromic children with CL and P patients at medical colleges of Lucknow Uttar Pradesh India, from January 2019-December 2019 were studied. Data were analysed using Statistical Package for Social Sciences (SPSS) version 16.

Results: A total of 370 cleft lip and palate patients were managed. More than fifty-five percent were male and 44.3% were female. 9.7% patients had ocular anomalies. Out of 36 CL and P patients, 41.7% patients had bilateral cleft lip and palate, 22.2% patients had unilateral cleft lip and palate. Overall, 63 ocular defects were identified in 36 patients. Lid abnormalities were the common most followed by Orbital and squint.

Conclusion: Individual approach and long-term follow-up of multidisciplinary specialists for each syndromic cleft lip and palate. Patient is required to classify early forthcoming complications.

Keywords: Cleft lip and palate, Ocular anomalies, Congenital anomaly.

Introduction

Clefts of the lip and/or palate CL/P are the most common congenital malformation of the head and neck [1]. Prevalence rate for live births with cleft palate, cleft lip, or both was 1.39 per thousand live births [2]. Although the incidence varies among different ethnic groups, highest amounts have been reported among Asians [3, 4], and the least amounts have been found amount Afro-Caribbean populations [5]. Majority of CL/P Patients suffer from feeding difficulties in infancy and speech, hearing and dental problems as they grow older, and life-long social and psychological problems due to the facial deformity. The cause of cleft lip and palate is complex. Genetic and environmental risk factors have been identified as triggers for syndromic CL/P; however, the aetiology of the more common non-syndromic CL/P remains largely unknown [6]. geographical area, Gender, population, dietary habit, use of drugs, tobacco use, drinking alcohol, low contaminated water sources and birth weight have all been theorized as factors cumulative the incidence rate of CL/P in new-borns [7-11]. Cleft lip and palate (CL and P) could be associated with many other structural abnormalities of the adjacent vital structures of the face like the ears, eyes, nose, teeth and brain. CL and P are intrinsically known for functional difficulties disturbing the feeding, breathing hearing, speech, vision Moreover, their negative impact on cosmetic. From the neural crest cells, nearly all soft tissue components and skeletal of the craniofacial area are exceptionally derived [12]. Because eyes originate as an extension of the forebrain, malformations involving ocular structures invariably accompany those of the face and brain and vice versa. Instabilities in normal relocation of eye fields from the lateral to frontal areas of the embryo's face in the fifth through eighth weeks have been proposed as a probable cause of facial clefting and ocular hypotelorism and hypertelorism [13]. Ophthalmologists have a exclusive role in the initial recognition of many ocular anomalies associated with disorder. In this hospital-based study, an effort was made to identify the ocular anomalies seen in patients with CL and P in north Indian population, so that early treatment can be provided to these patients for the same.

Materials and Materials

This prospective study was carried out at ELMC hospital, Lucknow and Integral institute of medical sciences and research, Integral University, Lucknow Uttar Pradesh, India over one year period from January 2019 to December 2019 in accordance with the ethical standards set forth in the 1964 Declaration of Helsinki. Informed written as well as verbal consent was obtained from each patient and a parent or guardian. Using an alpha level of 0.05 and the survey sample size determination table created by Bartlett et al. [14], we determined that the minimum sample size required for this study was 370 participants. 370 consecutive syndromic and non-syndromic children with cleft lip and palate patients (206 boys and 164 girls). All patients with Clefts of the lip and/or palate CL/P were screened for ocular anomalies in the hospital and those with anomalies were further studied. Information about variables from the patients and their parents were taken for the study. These variables sub-grouped into demographic data such as age, gender, birth weight, age of mother, cleft type, family history of cleft.

Classification of the clefts

The patients were divided into cleft lip (CL), cleft palate (CP), and cleft lip and palate (CL and P) based on the location of their clefts. CL and (CL and P) were subdivided into unilateral and bilateral groups. All children had undergone full clinical and para- clinical examinations by a pediatrician, dentist, pediatric cardiologist, oral and maxillofacial surgeon and an otorhinolaryngologist.

Statistical analysis

Data were presented in number and percentage. The Statistical Package for Social Sciences (SPSS), Version 16.0 (SPSS Inc. Chicago, USA) was used to analyse the data. Chi-square tests were performed to determine the significance of the findings. Statistical significance was set at $P<0.05$.

Results

Total 370 children, 206 (55.7%) were male and 164 (44.3%) were female, out of which 104 cases (28.1%) had cleft lip only, distributed as following: 82 cases (22.2%) with unilateral cleft lip and 22 cases (5.9%) with bilateral cleft lip. One hundred twelve cases (30.3%) had cleft lip and palate, 90 cases (24.3%) of which were unilateral, and 22 cases (5.9%) were bilateral. The highest number of clefts belonged to cleft palate comprising 154 cases (41.6%) of total patients (Table 1). There was significance difference found in between gender with the type of cleft with commoner involvement in males as compared to females.

Table 1: Association of the gender with the incidence of type of cleft

Cleft Type	Male N (%)	Female N (%)	Total Number	Chi Square, P value
Unilateral cleft lip	52(63.4)	30(36.6)	82	
Bilateral cleft lip	16(72.7)	6(27.3)	22	16.14,
Unilateral cleft lip and palate	54(60.0)	36(40.0)	90	
Bilateral cleft lip and palate	16(72.7)	6(27.3)	22	0.003
Cleft Palate	68(44.2)	86(55.8)	154	
Total	**206**	**164**	**370**	

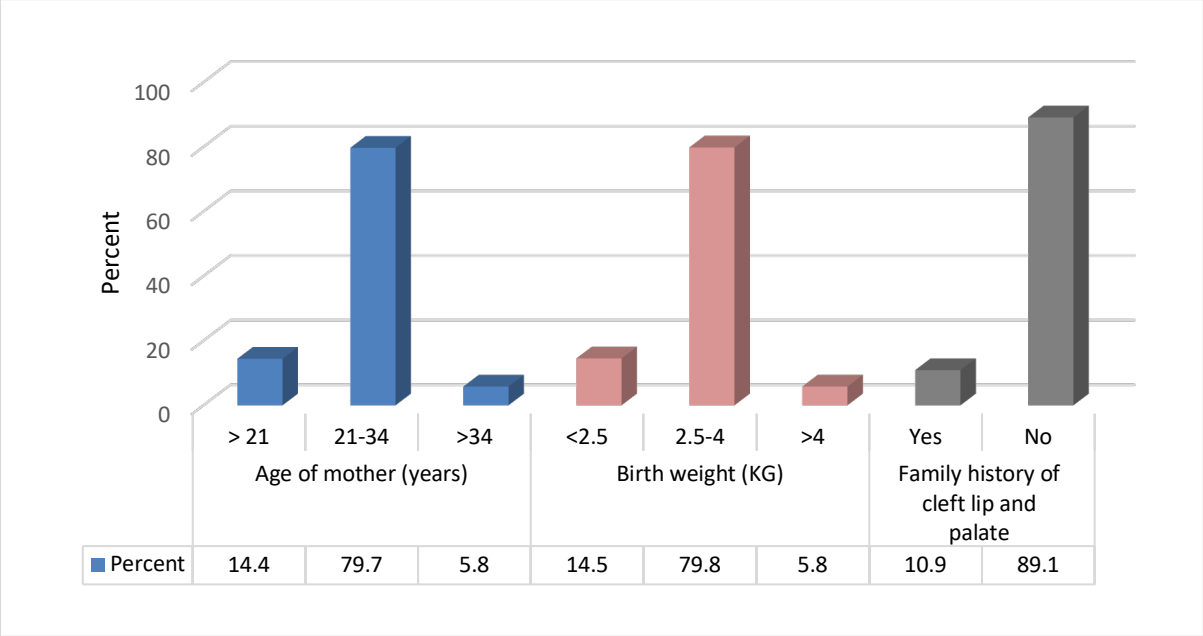

Figure 1: depicts that the percent distribution of associated risk factors

In which majority of mothers belongs to age group 21-34 years. Very few percent of mothers were above 34 years age. Most of children had weight at the time of birth were in between 2.5 kg to 4 .0 kg. Approximately eleven percent of the children had family history of cleft lip and palate, also seen among the associated factors for cleft lip and palate.

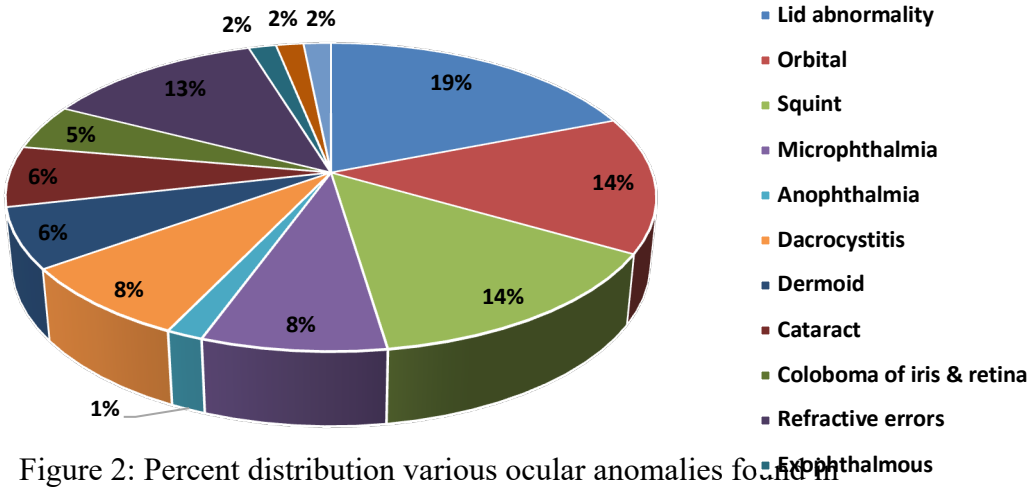

Figure 2: Percent distribution various ocular anomalies found in 36 patients

Figure 2 illustrates that the various ocular anomalies found in the study, in which most common ocular anomaly to be lid abnormalities followed by Orbital and squint. Overall, 63 ocular defects were identified in 36 patients. Lid abnormalities were the common most i.e., 19% of the total defects (12/63), which comprises lid colobomas, symblepharon, ectropion, euryblepharon and ptosis. Second common most anomalies were squint (14.3%) and orbital defects (14.3%) (Hypertelorism and Telecanthus). Abnormalities of the Microphthalmia (7.9%), refractive errors (12.7%), cataract (6.3%) and Dacrocystitis (7.9%) constituted the rest.

Table 2: Ocular anomalies in relation to type of cleft lip and palate

Ocular anomalies	Bilateral cleft lip and palate (15 patients)	Unilateral cleft lip and palate (8 patients)	Facial clefts (4 patients)	Syndromic (4 patients)	Cleft lip with alveolus (5 patients)
Squint	5	2	-	2	-
Coloboma lid	1	-	1	3	-
Microphthalmia	2	-	2	1	-
Coloboma iris	2	1	-	-	-
Symblepharon	2	-	1	1	-
Dacryocystitis	3	-	2	-	-
Limbal dermoid	2	1	-	1	-
Cataract	2	-	-	2	-
Myopia, astigmatism	4	-	-	-	-
Ptosis	1	1	-	-	-
Telecanthus	4	2	-	1	-
Euryblepharon	-	1	-		-
Congenital nystagmus	-	1	-	-	-
Hypermetropia	-	2	-	-	-
Epicanthus	-	-	1	-	-
Nystagmus	-	-	-	1	-

Table 2 shows the ocular anomalies in relation to type of cleft lip and palate. Three hundred seventy patients with CL and P detected, 36 (9.7%) had ocular anomalies. Out of 36 patients, 15 (41.7%) patients had bilateral cleft lip and palate, 8(22.2%) patients had unilateral cleft lip and palate, 4 (11.1%) patients had facial clefts, 5 (13.9%) patients had clefts with alveolus and remaining 4 (11.1%) patients had identifiable syndromes.

Discussion:

Cleft lip & cleft palate is one of the commonest congenital facial anomalies in Indian population. Different types of cleft-associated anomalies occur with variable frequency. Loretz, Westmorel and, Richards [15] found that anomalies of the bones and joints are the most commonly found anomalies in conjunction with cleft lip and/or palate. According to Ivy [16] and McKeown and Record [17], anomalies of the nervous system occurred most frequently, while Kraus, Kitamura, and Ooe [18] reported that brachdactyly and syndactyly were the anomalies most frequently seen in cleft lip and/or palate fetuses. Other anomalies frequently accompanying cleft palate are ocular anomalies; supernumerary fingers, toes, and teeth; malformed ears; clubbed hands or feet; tongue abnormalities; macroglossia and mandibular micrognathia [19]. Some recent studies reported that females are bornwith anomalies in addition to cleft palate defects more frequently as compared to males. According to Lutz and Moor (20) 62% of the cleft associated defects were in females, while Ivy [16] found that slightly more than 50% of the associated anomalies were observed in cleft palate males. In our present study, cleft lip and palate was higher in male patients as compared to female patients. About 28% patients had cleft lip only, 22.2% with unilateral cleft lip and 5.9% with bilateral cleft lip. One third patients had cleft lip and palate. Moreover, Adesina et al [21] found that although the incidence of cleft anomaly was highest as isolated unilateral cleft lip, other associated anomalies (28%) was highest in patients with isolated cleft palate. Similar studies reported earlier by Stoll [22] and Natsume [23]. Josef et al. however reported a higher incidence of associated anomalies among patients with combined cleft lip and palate [24]. The findings of the current study also correspond with the findings of the study conducted by Figueiredo et al. [25] in relation to family history of cleft anomalies. However, the present study showed a higher incidence of CL and P in babies born of mothers younger than 21 years old. Relatively consistent with the present study, Acuna-Gonzalez et al. [26] also found that the highest risk for CL and P was associated with variables related to family history background and family history of CL and P. The reported prevalence of associated anomalies varies widely across the literature; generally, a prevalence rate between 3% and 63% has been reported which is a reflection of varying data collection [27]. Among the ocular association various types of eyelid defects found were coloboma, eury blepharon, ectropion, Ptosis and symblepharon. Developmentofeyelid and palatine process occur almost simultaneously in the intrauterine period and consequently a defect of the palate could lead to anocular defect and resultant coloboma of the lid [28]. Ptosis of the eyelid which is a rare finding found in 2 patients in the present study. Ocular movement defects or squint is not uncommon in CL and P. In present study approximately 15% of patients had non-paralytic squint and this was attributed to the irregular shape and position of the orbital cavity and abnormal insertion of extra ocular muscles or both [29]. Five of our patients included in the study had congenital dacryo cystitis.

Because of entrapment of epidermal cells, four patients were found with limbal dermoids. Microphthalmos is a rare congenital condition where the size of the eyeball is smaller than normal. It is frequently associated with ocular anomalies

which include epibulbar dermoid, anophthalmia and colobomas of upper eye lid. Other abnormal facial findings include hypoplasia, lateral cleft lip, preauriculartags/pits, deformity of middle ear and ossicles, deformity of pinna and external auditory meatusca using conductive hearing loss and hypoplasia of temporal bone. Blepharocheinodontic syndrome isa rare autosomal dominant condition of congenital facial clefting. Previous studies have found association between cleft lip and palate and eye lidretraction, euryblepharon, lagophthalmos. Thus, the wide variation in reports on prevalence of cleft deformity and associated ocular anomalies is found in previous literature which is in concurrence with our study. d [22]. A larger sample-based study of longer duration is to be planned in future to cover some other challenges other investigators have encountered are variation in the time of presentation of these cases after birth, level of knowledge of the investigators themselves and available technology, as well as variability in the clinical expressions of these associated anomalies [31].

Conclusion

The present study found various ocular anomalies associated with cleft lip and palate. Individual approach and long-term follow-up off or each syndromic cleft lip and palate patient is necessary in order to classify defects and recognize early upcoming complications. This study proposes the need for further research; considerably more information is needed concerning the incidence, including interpopulation differences, and the type of additional congenital anomalies in cleft lip and palate.

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