

Original Article

A clinical and demographic profile of the cleft lip and palate in Sub-Himalayan India: A hospital-based study

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ABSTRACT

Objectives: To study the cleft lip and cleft palate in the poverty stricken Sub-Himalayan Garhwal region of India, being a commonly seen congenital abnormality and scarcity of studies about the demography of cleft in this region. **Design:** A prospective cohort observational case series was performed on 4657 cleft patients at a Tertiary care Hospital in Dehradun, India, over a period of 5 years. **Outcome measures:** The authors investigated the differences between age and sex with cleft status and family history of clefts, birth order, religion, socioeconomic status, parent literacy, source of information for treatment, haematological investigations showing the status of infection and coagulation in such children and satisfaction after treatment. **Results:** Seventy-two percent parents of cleft lip patients were illiterate, and only 8% were graduates, the majority of patients were from the low socioeconomic class. The siblings of 1.1% of the cleft patients had similar deformity. Anemia was seen in 83.16% cases which was commonly microcytic hypochromic type and eosinophilia was seen in 25.50% of cases. In the coagulation profile, International Nationalized Ratio was found to be raised in 52.12%. Almost 95% of the families were fully satisfied by the treatment and results. **Conclusion:** This study will provide baseline information on the status of these less privileged cleft patients in this mountainous region for future reference to health workers.

KEY WORDS

Cleft lip; cleft palate; haematological; sub-Himalayan region; socioeconomic

INTRODUCTION

The cleft of lip and alveolus, hard and soft palate are the most common congenital abnormalities of the craniofacial structure. Worldwide incidence

of the cleft lip and palate is one in 600 (1:600).^[1] The overall worldwide prevalence of the cleft lip with or without the cleft palate was 9.92 per 10,000. The prevalence of the cleft lip was 3.28 per 10,000, and that of the cleft lip and palate was 6.64 per 10,000,^[2] birth prevalence of clefts is somewhere between 27,000 and 33,000 clefts per year^[1] Lowest incidence occurs in Native American tribes of Montana, USA, which is 1:2076.^[3] The term cleft lip and palate inadequately describes the potential complexities of the deformity which may involve nose, lips, alveolus or palate. As a consequence breathing, appearance, dentition, dental occlusion facial growth, speech and hearing can all be

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affected leading to psychosocial implications.^[4]

It may not be the end of life but for children with cleft problem, the problem goes beyond the obvious disfigurement of face and extends to repeated infections, social stigma, and mental impairment that affect the speech, hearing, and teeth formation. These children are teased about their cleft-related features such as speech, teeth, and lip appearance and lose self-confidence.^[5] Research has shown that attractive children are seen by others as brighter, as having more positive social behavior and receive more positive treatment than their less attractive cleft counterparts. These children suffer with emotional “burn out” in adolescence. Therefore, it has been suggested that these patients should be included in national policies for integration of handicapped people, in agreement with programs of human rights, establishing a collaborative action between state and society. This would assure their inclusion in the socioeconomic and cultural context and equal opportunities in society, without privileges or paternalism.^[6]

Association of clefts with haematological abnormalities is seen such as anemia, eosinophilia and defects of the clotting mechanism.^[7] A common clinical observation among children with cleft palate is high prevalence of recurrent respiratory infection of the upper and lower airways^[8] and also, eosinophilia has been seen to be associated with cleft lip and cleft palate.^[9]

The condition in tropical countries like India is worse due to poverty and illiteracy. The difficult mountainous region of Northern India has suffered even more because of improper health facilities in the past. It is only since few years that this newly formed state of Uttarakhand, India, has developed some centers of excellence, but still not within easy reach of the general masses of mountains. In addition, due to lack of awareness, the patients of cleft remain untreated or mistreated by some quacks.^[10]

As this is a geographically and economically different region than others, the demographic, prenatal and clinical profile of the clefts of this region will also differ. There are no such studies so far published from this region, thus this study was undertaken to form a clinical picture of patients specific to the Sub-Himalayan Garhwal region of India for the benefit of patients and the treating doctor.

MATERIALS AND METHODS

This work was carried out at a Medical College Hospital,

Dehradun, over a period of 5 years. The study was approved by the Research and Ethics Committee of the hospital. All cases of the cleft lip and palate attending the plastic surgery outpatient department were included in this study. A total number of 4657 patients including primary and secondary cleft lip and palate were investigated. The details of the subjects in terms of history, clinical features and haematological investigations were recorded on an investigator-designed proforma, after obtaining an informed consent from the patient. The authors investigated the differences between age and sex with cleft status and family history of clefts, birth order, religion, socioeconomic status, parent literacy, source of information for treatment, haematological investigations showing the status of infection, coagulation in such children and satisfaction after treatment.

International classification of the cleft lip and palate was used for description purposes. Hb%, TLC and DLC were performed by an automatic analyser (MS-9, Melet Schloesing Laboratories), Absolute eosinophil count, bleeding time and clotting time were measured manually, while prothrombin time was measured by using a semi-automated coagulometer. All the standard reference ranges of haematological parameters were taken from standard reference (Wintrobe).

After a thorough postoperative assessment, the patients were generally discharged within 2–3 days and then called for follow-up on seventh postoperative day for suture removal and then monthly up to 3 months.

Subjective analysis of patients, above 5 years of age and family, in terms of their satisfaction after surgery was also recorded on an investigator designed form, at the time of their discharge from the hospital.

The data collected were then tabulated and analyzed.

RESULTS

Most of the data are summarized in [Tables 1-4, Figures 1-2]. In this study, out of 4657 patients the highest incidence was in the age group of 1–10 years, i.e. 58.65%.

The majority of the cleft patients presenting for cleft treatment were males, the male: female ratio in our study was 2:1 [Figure 1].

In this study, the monthly family earning in 60.58% cases

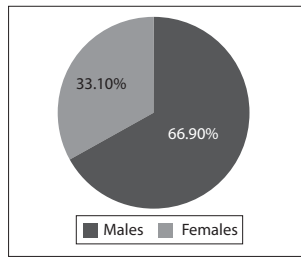


Figure 1: Sex ratio of patients

was below 1000 rupees, 21.15% were between 1000 and 1999 rupees, 0.65% was in the range of 2000–2999 rupees and 17.62% cases were above 3000 per month.

Most of our cases of the study gained information about treatment facilities for the cleft from newspapers, advertisements and news articles which have been the most popular source of mass information in this Garhwal region [Figure 2].

This study shows that 51.95% of parents of children with clefts were absolutely illiterate. A total of 24.67% of parents had attended only primary school while 15.38% of parents went to secondary school. Only a small percentage, i.e., 8% was graduates. Maternal antenatal history: In our study, 45.5% of women did not take any iron and folate supplementation while 54.5% regularly supplemented their nutritional requirement throughout gestation.

Prenatal history of anemia emerged out to be present in maximum cases when gestational problems in mothers of cleft children were studied [Table 3].

Bilateral clefts were present in 19.26% cases whereas 50.37% cases had left-sided clefts, 30.37% cases had right-sided clefts, also it was observed that complete cleft lip was present in 70% cases and incomplete cleft lip was present in 30% of cases. The isolated cleft lip (52.24%) was much higher than the isolated cleft palate (13.47%) or cleft lip associated with cleft palate (34.29%).

Patient–family satisfaction following cleft repair

93.26% cases enjoyed esthetic and functional satisfaction while the rest reported some sort of dissatisfaction for reasons such as scar problem, lengthy treatment and follow-up, projecting teeth or improper tissue alignment in cases of cleft lip repair and in terms of control of nasal regurgitation, speech and improve self-esteem in cases of cleft palate repair.

Anemia was seen in 83.16% cases which was commonly

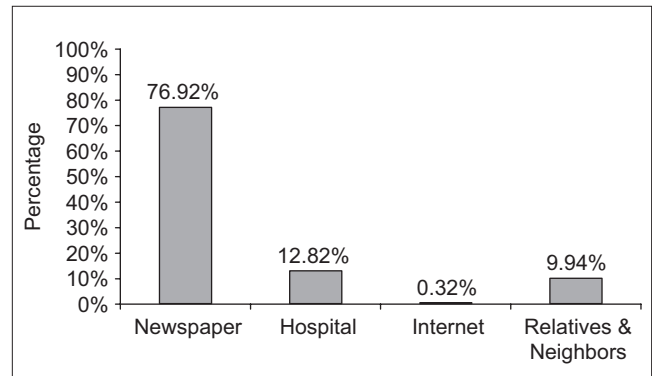


Figure 2: Source of information of treatment facilities

microcytic hypochromic type, eosinophilia was seen in 25.50% of cases. TLC was normal in 74.34% cases while leucocytosis was seen in 23.66% and leucopenia in 2.00%. The absolute eosinophil count was raised in 20.60% cases.

The coagulation profile

Normal bleeding time was seen in 70.14% of the patients, normal clotting time in 75.06% and prolonged prothrombin time in 15.8%. International Nationalized Ratio (INR) was found to be raised in 52.12% cases.

Associated congenital anomalies

Down's syndrome was seen in 0.32% of the cases, Pierre Robin syndrome in 1.28%, tongue tie in 1.32%, hypospadias 0.96 and congenital amniotic bands in 0.64.

DISCUSSION

Cleft deformities are a challenge to the surgeons since very long period. The cleft lip was presumed to be a curse of God in ancient times, but strange enough, the population living in the remote hilly areas of the Garhwal region still believes the same. The children born with these deformities are called as “Khandu” (incomplete) by the rest of the society. This study was undertaken for these unfortunate patients, where there are no documented data available in the difficult mountainous region of Garhwal.

The age groups under study in some of the previous studies were 1 day to 26 years, and 10 months to 11 years. However, they did not specify the commonly affected age group. It is evident that the majority of cleft patients report for treatment by 10 years of age, as also seen in this study.^[11,12] The sex wise analysis of the number of cleft cases reveal that two-thirds of them were males and one-third were females and such proportions were consistently

Table 1: Age distribution of cases

Age (years)	Percentage cases
0–1	14.78
>1–2	28.84
>2–3	9.0
>3–5	7.02
>5–10	13.78
>10–20	20.19
>20–30	5.42
>30–40	0.96

Table 2: The religion wise distribution of cleft cases

Religion	Percentage
Hindu	74.35
Muslim	24.36
Sikh	0.96
Christian	0.11

Table 3: Gestational problems in mothers of cleft children

Prenatal history	Percentage of cases
Anemia	41.34
High fever	8.97
Drugs	11.86

Table 4: Religion wise distribution in general population and cleft population

Religion	General population (Lakhs)	No. of cleft cases	Ratio
Hindus	72.12	3462	1:2080
Muslims	10.12	1135	1:881
Christians	0.27	5	1:5400
Sikhs	2.12	45	1:4444

noticed in the study by Orkar *et al.* in 2002.^[11] Zhou *et al.* observed that the male: female ratio was 2:1. The study done by David *et al.* shows similar results.^[13] There was no significant difference in sex distribution; the male to female ratio was 1.1:1. However, females had a higher incidence of cleft palate than males.^[14]

In our country where female feticide is so common, people usually refrain from bringing females to hospital. However, the M: F sex ratio of 2:1 seen in our study may not be the true representation of sex wise distribution of clefts because the fair sex in majority of Indian Society unfortunately is given the second preference as compared to males. It is assumed that many of female cleft patients are not brought to the hospital for treatment at all.^[11,15]

In this study, it was noted that the incidence of the isolated cleft lip was much higher than the isolated cleft palate or cleft lip associated with cleft palate, while Richard and

William found the isolated cleft lip in 15% cases, the cleft lip and palate in 45% cases and the isolated cleft palate in 40% cases. Zhou *et al.* similarly found in their study that unilateral clefts were more common than bilateral, with unilateral-to-bilateral ratio of 10.4:1. They noticed that left-sided defects were more common than those of right-sided. A study conducted by Blanco-Davila also suggested that clefts of the left side occurred more often than the right side and boys were affected more commonly than girls.^[15-17]

In our patients we observed that Hindus were most affected, followed by Muslims and then others. No such a religion wise distribution for clefts has been studied in earlier research studies. Table 4 depicts the religion of the general population in this state and that of the cleft.^[18]

Upon assessing data collected by Uttaranchal Government, it was observed that the frequency of the cleft in Muslim population was higher than the Hindus, Christians or Sikhs as seen in the religion wise distribution of cleft cases. However, this study being a hospital-based research this ratio may not be a true representation of the religion wise distribution of clefts in the society.

In the majority of cleft cases, the family's income was less than Rs. 1000/-per month and on considering the income level of Rs. 1750/- per month as the government identified "poverty line", it may be stated that more than three-fourths of families of persons with clefts belonged to below the poverty line status. The association of clefts with the low socioeconomic class has also been corroborated by Irma and Puho *et al.* in the mothers of cases with orofacial cleft. Parents below the poverty line are naturally deprived of nutritious diet, periconceptional folic acid and multivitamin supplementation during the antenatal period. This could be a contributory factor for occurrence of clefts in poor families.^[19-21]

Gupta *et al.* showed that the patients of cleft are brought for surgery later than the ideal surgical time, which also may be due to ignorance, poverty or unawareness about the fact that cleft anomaly can be corrected by surgery. Without repair, these children would have suffered from facial disfigurement, feeding problems, social isolation and abnormal speech.^[22] These patients are unable to afford the treatment offered by the tertiary hospitals, but in our study such a large number of poor patients could undergo surgery because of "The Smile Train Project" which is a funding agency.

The educational status was also very low among the parents of cleft families. It was noticed that only 8% were graduates. Illiteracy can be a reason for consanguineous marriages and multiple conceptions. Frequent pregnancies can lead to anemia contributing to cleft deformity. The literacy rate in Uttarakhand is 79.63% as per 2011 population census as compared to the families of cleft population as seen in this study which is 51.95%.^[23,24]

In this region, the majority of the terrain is hilly regions which are difficult to reach. Parents of a large number of cleft patients in our study gained the information regarding the availability of treatment facilities for cleft deformity from newspaper advertisements and news articles. This shows that the level of mass awareness and concern about treatment of cleft disorders is generally low in this population.

In our study the majority of women did not take any folate supplementation. Andrew *et al.* emphasized that the periconceptional daily supplementation of multivitamins including physiologic doses (<1mg) of folic acid or folic acid alone cannot reduce the birth prevalence of cleft lip and cleft palate. Only the high pharmacological doses (6mg) of folic acid alone in the critical period of the primary and secondary palate development are effective for the reduction of orofacial clefts.^[25]

In this study, prenatal adverse factors associated with cleft lip and palate were anemia, drugs and high fever. Puha *et al.* confirmed the orofacial cleft inducing effect of phenytoin, carbamazepine, oxytetracycline and thiethylperazine and suggested a possible association between orofacial clefts and oxyphenolol and amoxicillin. Lack of multivitamins, inappropriate use of drugs and problems such as high fever and anemia during pregnancy are likely etiological factors in the development of cleft deformity.^[21,23,25]

Down's syndrome and few others are the associated congenital anomalies observed by Orkar *et al.* and they were similar to our results. Associated congenital anomalies should be suspected in the patients of cleft deformity, so as to have better care and management of the patient.^[11,26]

Association of clefts with haematological abnormalities is seen such as anemia, eosinophilia and defects of the clotting mechanism.^[7] There seems to be a reasonable agreement on the minimum haemoglobin of 10g%, with only 14 (12.5%) centers willing to operate between 8 and <10g%.^[27]

In this study, the majority of our patients being poor and illiterate were malnourished and living in unhealthy conditions. This accounted for a derangement of their haematological profile, the most important of them being anemia especially microcytic hypochromic type and alteration in the coagulation profile was seen in the majority of cases, and the INR was also found to be raised in more than half the cases. Eosinophilia and lots of patients with eosinophil count close to higher normal limits were seen and was probably due to local climatic factors and co-incidental worm infestation or allergy which can be attributed to this geographical region where allergies are common.^[7,28] Absolute eosinophil count (AEC) was therefore considered to be a more suitable parameter in these patients for evaluation of these specific granulocytes. Whatever the reason, the raised values are significant and it delays the surgical management by 5–7 days, which again is unaffordable by these poverty stricken patients and their attendants. Considering all this, there should be cleft counseling and continued cleft care for the complete complex of deformities.^[29]

CONCLUSION

This study will provide baseline information on the status of cleft patients in the Sub-Himalayan Garhwal region of India for future reference to researchers, doctors and paramedical workers working for the upliftment of these low privileged children and adults. This study will also lay the groundwork for a better understanding of clefts that present challenges for management. This study invites further work aimed at problems in clefts and betterment of cleft care services especially in such regions of tropical countries.

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