



Developmental Defects of Enamel in Children with and Without Cleft Lip: A Case Control Study

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ABSTRACT

Orofacial clefts is the most common craniofacial birth defect and the fourth common congenital malformation in humans which require multidisciplinary care. Enamel defects are commonly seen in deciduous and permanent teeth especially maxillary incisors in an individual with cleft lip, palate, and alveolus. Hence a study was conducted to analyse the developmental defects of enamel in children with cleft lip and compare with children without cleft lip. Retrospective data collected from 89,000 case records from June 2019 to March 2020 were taken for the study. Based on the inclusion and exclusion criteria, the present study consisted of 6 children divided into two groups: children with cleft lip and children without cleft lip. In both groups, presence of developmental defects of enamel in the tooth were verified and data was tabulated. The data was subjected to Mann-Whitney test using SPSS software. Children in both the groups (with and without cleft lip) did not have any developmental defect of enamel. Within the limitations of the present study, there was no difference in the occurrence of developmental defects of enamel in children with and without cleft lip.

Keywords: Cleft lip, Central incisor, Developmental Defects, Enamel, Opacity

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INTRODUCTION

Orofacial cleft (OFC) is the most common craniofacial birth defect in humans. Orofacial clefts exhibit both ethnic and geographic variation. The estimated prevalence is 1.7 in 1000 live births in India. Incidence of cleft lip and palate (CLP) varies from 0.25 to 2.29 per 1000 births in India [1-3]. The craniofacial structure development is a coordinated process which involves the growth of multiple independently derived embryologic prominences called primordia. Incomplete fusion of primordia during 4th to 8th week of embryological life which leads to cleft lip, cleft of primary or secondary palate or both [3,4].

Clefts can be caused by various factors which include infection, toxicity, poor diet, hormonal imbalances, and genetic interference. Among these factors, genetics play an important role in cleft lip and palate. Previous study conducted by Shaw et al. [5] presented evidence that women above 35 years of age had a doubled risk of having a child with cleft lip/palate, and women above 39 years of age had a tripled risk of having a child with cleft lip/palate. Consanguineous marriages also have an increased risk of developing cleft lip or cleft palate in the children. Dental complications include congenitally missing teeth, neonatal teeth, ectopic eruption, supernumerary teeth, anomalies of shape and size of tooth, macrodontia, microdontia, fused teeth, enamel hypoplasia, deep bite, cross bite which can be anterior or posterior, crowding and spacing of the teeth [6].

Enamel defects are commonly seen in deciduous and permanent maxillary incisors in patients

with cleft lip, and palate and has been associated with the cleft especially when the alveolus is involved. Depending upon the macroscopic appearance, defects of enamel may be classified into enamel hypoplasia and Hypomineralised enamel. Among these two defects, hypoplasia is a quantitative defect whereas Hypomineralised enamel is a qualitative defect which is seen as an abnormality in enamel translucency [7-10]. Defects in enamel formation may cause aesthetic alterations and compromise tooth enamel structure. Severe defect leads to early enamel loss, consequently, results in tooth wear and impaired functioning. Less mineralized enamel or enamel with an irregular surface may become more susceptible to caries development thereby leading to pulpal involvement [11-13]. Such defects are commonly noticed in children with cleft lip and/or palate [13]. However, studies conducted in the south Indian population are limited. The present study was done to assess the presence of developmental defects of enamel in patients with cleft lip and compare with children without cleft lip and to create awareness among the cleft lip patients.

MATERIALS AND METHODS

This is a retrospective study. This study was carried out in a hospital-based university setting. This study was evaluated and ethically approved by an institutional ethical review committee. Retrospective data collected from 89,000 case records from June 2019 to March 2020. Informed consent was obtained from the parents or guardian before starting the treatment. Inclusion criteria: Children with cleft lip, children aged from 3 years to 18 years, children with at least one or two erupted teeth, complete photographic and written records regarding the complete intra-oral examination of the patient. Age and gender matched controls i.e. children without cleft lip, were taken according to the relevant cases obtained from the inclusion criteria. The exclusion criteria were incomplete and censored dental records, children below the age of 6 months and improper photographs.

Total cases acquired for this study were patients 6 which includes 3 children with cleft lip and 3 children without cleft lip (age, gender matched controls). Selected case and control group were

examined by three people: one reviewer, one guide and one researcher. Patient's case sheets were reviewed thoroughly. Cross checking of data including digital entry and intraoral photographs was done by an additional reviewer, and as a measure to minimize sampling bias, samples for the group were picked by the simple random sampling method. Digital entry of clinical examination and intraoral photographs were assessed. For both groups, presence, or absence of developmental defects of enamel were noted by a researcher, entered Microsoft excel (MS Excel) and then transferred into Statistical Package for the Social Sciences (SPSS) Software for statistical analysis. A correlation test (Mann-Whitney test) was done between the children with cleft lip and children without cleft lip. The difference was statistically significant when the p-value was less than 0.05.

RESULTS AND DISCUSSION

The final study sample size included a total of 6 children with 3 children with cleft lip (case group) and 3 children without cleft lip (control group). In this study, the control group was matched based on age and gender as like the case group (Figures 1 and 2). Absence of developmental defect of enamel was noticed in all children in both the groups i.e. children with and without cleft lip. On comparison of the results using Mann-Whitney test, the results were not statistically significant (p-value=1) (Figure 3).

Dysfunction of ameloblasts results in changes

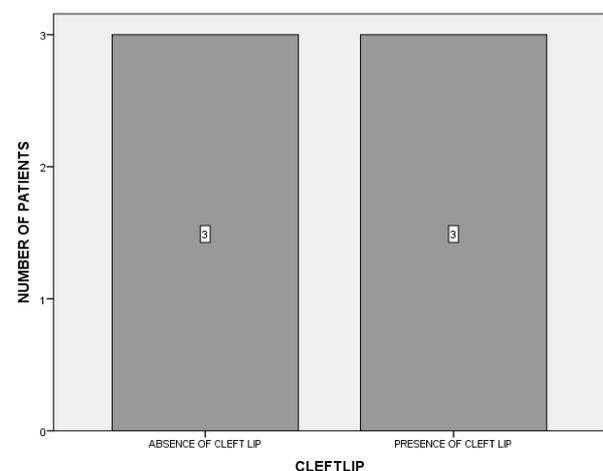


Figure 1: Bar graph represents the distribution of cases in case (children with cleft lip) and control group (children without cleft lip). (Y-axis represents number of patients; X-axis represents presence or absence of cleft lip) Note the equal distribution of cases in both the case (children with cleft lip) and control group (children without cleft lip).

in the appearance of the enamel in permanent dentition. These developmental defects of enamel (DDE) may range from slight change in the tooth colour to a complete absence of enamel. Developmental defects of enamel cause tooth sensitivity and an increased risk of dental caries [14]. Assessment of such enamel defects would be necessary for early detection and warning of

caries risk in such individuals which would help the practitioner to provide preventive measures [7,8].

The results of the present study show that children with and without cleft lip did not show any developmental defects in the enamel of the teeth. This was contradictory to majority of the studies which suggests a prevalence in children with cleft lip [15-17]. However, there were a few studies that supports the results of the current study with minimal presence of defects in the enamel of children with cleft lip [18,19]. This difference could be due to the smaller sample size, that would have affected the results of the current study in a unidirectional manner.

Fluoride use has been recommended to prevent dental caries [20]. Decreased concentration of fluoride also results in increased incidence of dental caries [21]. Prevalence of enamel defects increases with increasing fluoride level in drinking water. Good attitude of parents reflects as a good oral health in children and vice versa [22]. Preservation of primary teeth in the dental arch is important to guide the eruption of the permanent teeth in the optimal position. Grossly decayed primary teeth which are extracted before exfoliation causes space in the dental arch which causes malocclusion if space maintainer was not given [23,24]. Bacteria play a vital role in the initiation and progression of dental caries

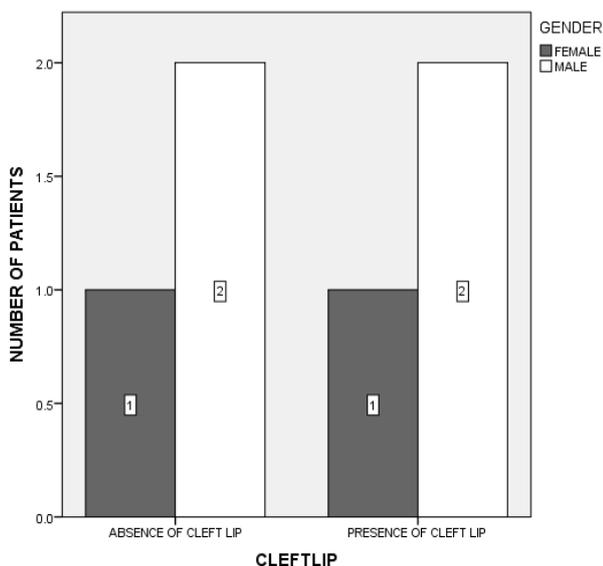


Figure 2: Bar graph represents the distribution of cases in case (children with cleft lip) and control group (children without cleft lip). (Y-axis represents the number of patients ; X-axis represents presence or absence of cleft lip; grey represents female ; white represents male) Note the equal distribution of cases in both the case (children with cleft lip) and control group (children without cleft lip).

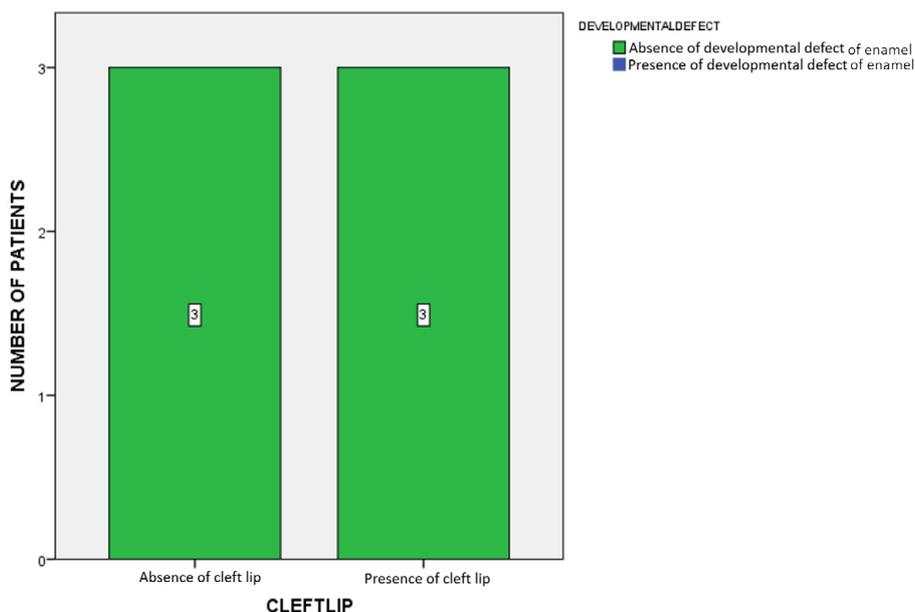


Figure 3: Bar graph represents the presence or absence of developmental defects of enamel in children with cleft lip and children without cleft lip. (X-axis represents presence or absence of cleft lip; Y-axis represents the number of cases; green represents absence of developmental defects of enamel; blue represents presence of developmental defects of enamel). None of the cases in both the groups (children with and without cleft lip) had developmental defects of enamel, which was not statistically significant. (Mann-Whitney U test; p-value=1-not significant).

which eventually causes pulpal and periapical disease [25]. Defect in enamel lead to early caries which would lead to pulpal involvement and even extraction [26-31]. Fluoride and regular toothbrushing can help in maintaining proper oral hygiene [32-34].

Maciel et al. [35] reported that there is a high incidence of enamel defects on the cleft side for both deciduous and permanent dentition. Chapple et al. [36] reported that 24% of cleft lip patients had enamel hypoplasia. Chia-An-Shen et al. [37] in 2019 reported that 87.9% of cleft lip patients had enamel defects. Amandeep Chopra et al [38] reported that the prevalence of developmental defects of enamel was higher in children with cleft. This is contradictory to our study.

Advantages of this study were that this was a case control study with age and gender matched controls to provide best results with high internal validity, reasonable data, Disadvantage of the study was that this was a unicentric study with geographic limitations, limited sample size and has lower external validity. Future scope for this study includes larger sample size which is not confined to a particular geographic area and to assess the presence of developmental defects if enamel by clinically examining the cleft lip patients.

CONCLUSION

Within the limitations of the present study, there is no evidence of developmental defect of enamel in children with and without cleft lip.

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CONFLICT OF INTEREST

The authors declare that there were no conflicts of interest.

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