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Developmental Defects Of Enamel In Children With Cleft Lip And Palate: A Case Control Study

Research Article

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Abstract

The occurrence of dental anomalies may result from both environmental and genetic factors. Enamel defects are commonly seen in humans who require multidisciplinary care. Enamel defects are seen commonly in both primary and permanent dentition especially maxillary incisors in individuals with cleft lip and palate. Hence, a study was conducted to assess and analyse the developmental defects of enamel in children with cleft lip and palate and also in children without cleft lip and palate.The retrospective data were collected by obtaining and analysing the 89000 dental case records of the university from June 2019 to March 2020. The present study consisted of 20 children divided into two groups; children with cleft lip and palate and children without cleft lip and palate. Control group was age and gender matched to the case group. None of the children with cleft lip and palate had developmental defects In both the case and control group, presence and absence of developmental enamel defects were noted. Within the limitations of the study, there was no evidence of developmental enamel defects in children with cleft lip and palate.

Introduction

The occurrence of dental anomalies may result from both environmental and genetic factors. Previous studies indicate that there is a higher prevalence of dental anomalies in children with a cleft condition than in the general population. Studies have also shown that both genetics and the surgical repair of the palate influence the occurrence of dental anomalies in the cleft population [1, 2]. Both sets of dentition may be affected and occur more frequently on the cleft affected side of the maxilla [3]. Children with cleft lip and palate are more prone to dental caries due to enamel and insufficiency and poor oral hygiene.Early childhood caries (ECC) is one of the common oral health condition existing throughout the global associated with several risk factors [4].

Dental caries if left untreated, can lead to caries involving the pulp. Pulpectomy is considered to be the treatment of choice for primary dentition with pulpal involvement [5]. Pulpectomy is the choice of treatment for symptomatic decayed primary teeth and is a challenging and one of the procedures that consumes more time in pediatric dentistry [6, 7]. Pulpectomy procedure is performed in primary teeth to avoid extraction and to maintain its form and function [8, 9]. Fluoride when present in optimal quantities is known to be helpful in caries prevention by various mechanisms but more predominantly by deposition of calcium fluoride crystals which is more resistant to demineralisation [10, 11]. The commonest types of dental anomalies have been reported to be absent or supernumerary teeth, enamel dysplasia and discoloration and delayed tooth development. In children with cleft lip and palate, the lateral incisors in the alveolar region have the highest prevalence of dental development disorder. This may cause functional and aesthetic issues for the child and complicating factors for dental and orthodontic treatment. While abnormalities are most commonly caused by defects in specific genes, pre and post natal aetiological event have also been implicated in anomalies in tooth dimensions, morphology, position, number and structure. Cleft lip and palate is one of the most common developmental disturbances of the orofacial structures [10]. There is a quite a marked racial variation in the prevalence and incidence of cleft lip and palate [12, 13]. Cleft lip and palate is more frequent in males,

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however cleft palate alone most frequently affects females [14, 15]. Depending on the embryological characteristics, varieties of cleft lip and cleft palate may be categorized as those involving the lip and palate, those in which palate is alone affected and congenital missing of the palate [16, 17].

Study conducted by Shaw et al., [18] in 1990 presented an evidence that women above 35 years old has a doubled risk of having a child with cleft lip and palate and women above 39 years of old had a triple risk of having a child with cleft lip and cleft palate,and also consanguineous marriages have an increased risk of developing cleft lip and palate children [6, 19]. Dental complications include congenitally missing teeth, neonatal teeth,ectopic eruption supernumerary teeth,anomalies of tooth size and shape, microdontia, macrodontia, fused teeth,enamel hypoplasia, malocclusion such as deep bite, crossbite which can be anterior or posterior, crowding and spacing of teeth [20-22]. 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 [23, 24].

Materials and Methods

This retrospective study was conducted under a university setting. Ethical approval for this study was granted by the institute's ethical committee (SDC/SIHEC/2020/DIASDATA/0619-0320). Consent to use treatment records for research purposes were obtained from patients/guardians at the time of patient entry into the university for dental needs. The retrospective data were collected by obtaining and analysing the 89000 dental case records of the university from June 2019 to March 2020. The inclusion criteria for the current study were children between the age of 3 years old to 17 years old, presence of cleft lip and palate, complete photographic and written records regarding the cleft lip and palate. Age and gender matched controls i.e.children without cleft lip and palate, were taken according to the relevant cases obtained from the inclusion criteria. The exclusion criteria were patients above 18 years of age and below 3 years of age, incomplete and censored dental records and absence of photographic evidence of cleft lip and palate. The selected case and control group were examined by three people; one reviewer, one guide and one researcher. The patients' case sheets were reviewed thoroughly. Digital entry of clinical examinations and intra oral photographs of selected subjects were assessed and this included the assessment of every patients'. The examiner was trained to add data on the developmental enamel defect for both case and control group by tabulation using excel software. Data analysis was done using SPSS PC Version 23.0 (IBM;2016) software for statistics. The incidence of developmental defects of enamel for both case and control group was compared by Mann-Whitney U-Test.The results were statistically non-significant when the p value was more than 0.05.

Results and Discussion

The final study sample size included a total of 10 patients with the patients with cleft lip and palate (case) and 10 patients without cleft lip and palate (control). In this study the control group was matched based on age and gender as similar to the case group (Figure 1 and 2). None of the children with cleft lip and palate had developmental defects. But developmental enamel defects were seen only in two (20%) children without cleft lip and palate.

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

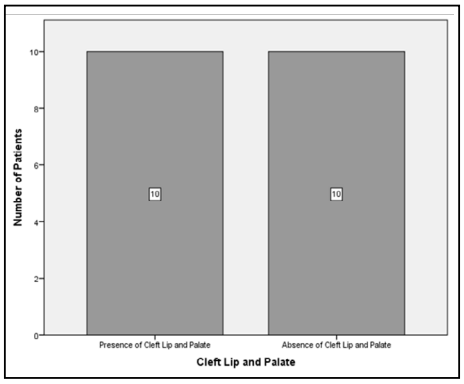


Figure 2. Bar graph representing the gender distribution of cases in the case group (children with cleft lip and palate) and the control group (children without cleft lip and palate). X- axis represents presence or absence of cleft lip and palate and Y- axis represents the number of patients. Note the equal distribution of cases based on gender in both the groups.

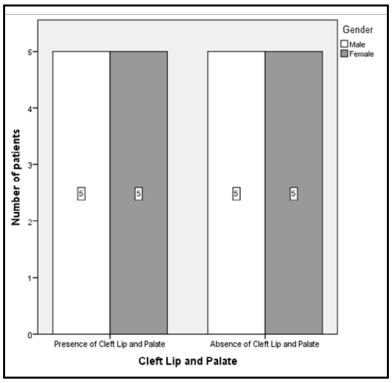
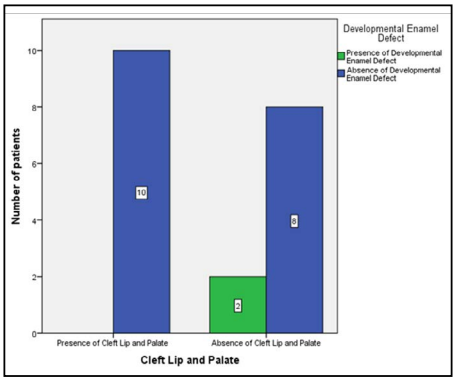


Figure 3. Bar graph representing the association of absence and presence of developmental enamel defect in children with cleft lip and palate. X- axis represents presence or absence of cleft lip and palate and Y- axis represents the number of patients. Developmental enamel defects were noticed (green) in children without cleft lip and palate when compared to children with cleft and palate. (Mann-Whitney U test; p-value =1- not significant).



This difference was not statistically significant. (Mann- Whitney U test; p-value =1). The developmental enamel defect that was noticed in children without cleft lip and palate was enamel hypoplasia.

Enamel is formed by cells called ameloblasts. These are the secretory cells which are particularly sensitive to changes in their environment during the process of enamel formation. Dysfunction of ameloblasts results in changes in the appearance of enamel in permanent dentition. These developmental defect of enamel may range from slightest change in the tooth colour to a complete absence of enamel [25]. Developmental defects of enamel causes tooth sensitivity and increased risk of dental caries [26].

Previous study conducted by Pijari and Lanning in 1995 [27], found that irradiation of 0.72 to 1.44 Gy to dental arches caused an increase in the number of enamel opacities along with the acute lymphoblastic leukemia. In another study conducted Ahalussa S [28], reported that dioxin being a causative agent for developmental defects of permanent first molars. Tacial et al., [29] reported that there is a high incidence of enamel defects on the cleft side for both deciduous and permanent dentition. Ruit et al reported that there is a significant increase in the incidence of enamel defects in patients with complete cleft lip and palate. J.R Chappel, reported that 24% of Cleft lip patients had enamel hypoplasia.

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 and disadvantage of the study was this was a unicentric study with geographic limitations, limited sample size and has lower external validity. The dietary factors, feeding and oral hygiene factors were not taken into consideration while interpreting the results. Future scope of this study includes larger sample size which is not confined to a particular geographic area and to assess the oral mucosa and changes by clinically examining the cleft lip and palate patients.

Conclusion

Within the limitations of the present study, the children without cleft lip and palate had higher chances of having developmental defects of enamel compared to children with cleft lip and palate. These defects need to be diagnosed carefully in children with cleft lip and palate and thereby treating them at earlier stages.

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