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Idiopathic Gingival Fibromatosis and its Management in Children: A Rare Case Report

Sachin Parmar[1], Anuradha Agrawal[2], Madhu Singh[3], Shivani Mishra[4]
[1]*Post Graduate Student, Department Of Pediatric & Preventive Dentistry, Govt. College Of Dentistry, Indore, MPMSU, India.*
[2]*Associate professor, Department Of Pediatric & Preventive Dentistry, Govt. College Of Dentistry, Indore, MPMSU, India.*
[3]*Professor & Head, Department Of Periodontics, Govt. College Of Dentistry, Indore, MPMSU, India.*
[4]*Medical Officer, District Hospital, Barwani, MP, India.*

Abstract:
Idiopathic Gingival Fibromatosis is a progressive fibrous lesion of the gingival tissue that causes aesthetic and functional trouble. Etiologyof Gingival Fibromatosis is diverse including genetic, inflammatory, drug induced etc. This case report discourse the identification followed by management of a case of idiopathic gingival fibromatosis in a 5 -year old female. Generalized diffuse gingival enlargement of both maxillary and mandibular arches covering all surfaces of teeth was seen, sparing incisal and occlusal third of few teeth resulting in compromised mastication and speech since last three years. On the basis of clinical examination, history given and tissue biopsy, idiopathic gingival hyperplasia was considered to be the final diagnosis. Keeping patient's age in mind, gingivectomy was performed in all quadrant under general anesthesia. Patient was asked to be on regular follow-up to check if any recurrence.

Key Word*:Gingival Enlargement, Gingival Fibromatosis, Gingivectomy*

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I. Introduction

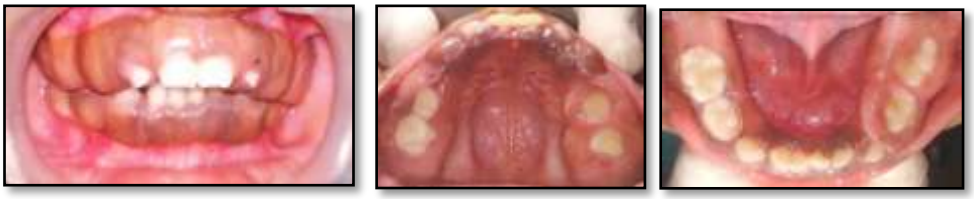
Gingival fibromatosis (GF) is a diverse class of disorders which is marked by progressive gingival enlargement due to submucosal connective tissue overgrowth.Gingival enlargement (GE) have various etiological factors including genetic, inflammatory, drug induced etc. while some GE are of unknown cause and termed as idiopathic. Exact pathogenesis of GE is not clear, however it is strongly associated to local factors (dental plaque, calculus), individual susceptibility and the effect of chemical substances. It is generally asymptomatic till the hyperplastic tissue cover the masticatory surface of tooth which get traumatized while mastication. Sometime because of huge GE, patient gradually develops an abnormal swallowing pattern, alteration in speech and difficulty in mastication.

Gingival Enlargement, also known as gingivamatosis or fibromatosis can take place as a single pathology or can be linked with several syndromes. GE does not have particular etiology, it can either be due to heredity, hormonal imbalances (e.g. pregnancy or growth-hormone related), drug induced, inflammatory, syndromes associated or can be oral manifestation of systemic disease.GE with no definite cause, is termed as Idiopathic GE. It is rare with prevalence of 1 in 7,50,000 peoples.(1) Characterized by enlargement of gingiva which is generally firm , pale pink and leathery with pebbled surface.

Histopathological features include, epithelial hyperplasia and elongated rete ridges with increased keratin production and collagen bundles. Generalized GE in children may result in over-retained deciduous teeth, delayed eruption of permanent teeth, difficulty in maintaining oral hygiene, malocclusion, compromised mastication, speech and aesthetics.This case report highlights management of a rare case of idiopathic gingival enlargement (GE) in child, along with its follow-up.

II. Case Report

A 5 year old female child along with her parents reported to the department with complaint of swollen gums in upper and lower arches (fig. 1). On taking history it was found that this problem persists from last 3 years. GE causes compromised speech, poor aesthetic and difficulty during mastication to child. Family history of a similar illness was ruled out, also no relevant history of any ongoing medication, seizures or any other mental or physical impairment was mentioned.



A **B** **C**

Fig. 1 Preoperative photographs (A) Frontal View (B) Maxillary occlusal view (C) Mandibular occlusal view

Extraoral examination reveals incompetent lip and convex facial profile. Intraoral examination revealed generalized GE involving both arches. GE was diffused, pale pink with melanin pigmentation in color, firm and fibrous in consistency. Teeth were scarcely visible as gingiva covers nearly incisal and occlusal third. There was no positive finding related to prompt gingival bleeding, linear ulcerations, mucosal pallor and petechiae. There was fair oral hygiene with least amount of plaque and absence of calculus and other inflammatory factors.

All the hematological investigations were performed to exclude any systemic involvement and findings were within the limits. OPG revealed all the primary teeth and developing permanent teeth bud with intact alveolar bone (fig. 2).

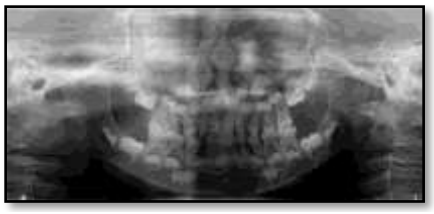


Fig. 2 OPG showing presence of all deciduous teeth and permanent tooth bud

Under general anesthesia gingivectomy was performed with external bevel incision given first to get fine contour and superior aesthetics. Kirkland gingivectomy knife was used for incision over facial and lingual gingival surface, Orban periodontal knife for interdental areas. Incisions were followed by electrocautery to attain proper hemostasis and to reshape the gingival tissue (fig. 3).

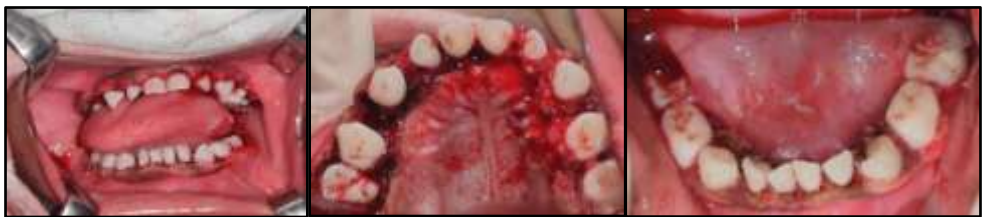


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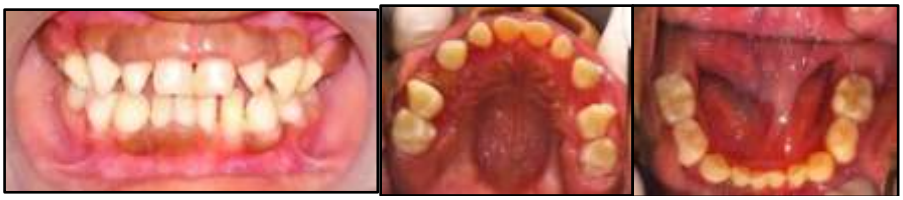
Fig.3 Operative photograph (A) Frontal View (B) Maxillary occlusal view (C) Mandibular occlusal view

Surgical dressing (Coe-Pak) was placed over gingiva for better management of postoperative pain and discomfort. Patient and parents were instructed to maintain proper oral hygiene. Histopathological examination of excised gingival tissue revealed hyperplastic gingiva with keratinized stratified squamous epithelium, Elongated interlacing rete ridges, Prominent fibrosis and focal lymphoplasmacytic inflammation noted in subepithelial tissue. Dysplastic or malignant changes were not evident (fig. 4). Hence the findings were in accordance to idiopathic GF.

Fig. 4 Histopathology of excised gingival tissue shows stratified squamous epithelium with hyperplasia and interlacing rete ridges. Subepithelial tissue shows marked fibrosis

A postoperative follow-up was done and after one month wound was completely epithelized. The patient was kept on follow-up at every 3 months interval. After 12 months follow-up, intraoral examination showed correct occlusion with no sign of further enlargement (fig. 5).



A **B** **C**

Fig. 5 A 12 month follow-up photograph (A) Frontal View (B) Maxillary occlusal view (C) Mandibular occlusal view

III. Discussion

GE in children is quite rare and only seen in cases where there is poor oral hygiene or having some associated drug history. Differential diagnosis of GE in children includes granulomatous diseases(eg.Tuberculosis, sarcoidosis, Crohn's disease etc.), AML and hereditary gingival fibromatosis.[2] Hereditary Gingival Fibromatosis tends to occur more frequently as a generalized type than Idiopathic Gingival Fibromatosis. The ratios of generalized to localized types in HGF and IGF have been reported as 15.2:1 and 1.6:1 respectively.[3] This condition can be evident as an autosomal dominant or less commonly as autosomal recessive mode of inheritance.[4]

In a study done by *Gangliano et al.* it was found that the possible mechanism for GE are excessive fibroblastic proliferation, increased production of type-I collagen, degradation of matrix metalloproteinase, formation of heat-shock protein and extracellular matrix components.[5] The hyperplastic response does not involve the periodontal ligament and occurs peripheral to the alveolar bone within attached gingiva.[6]

As per a recent report, an exacerbate proliferation and elevated growth of extracellular matrix molecules, collagen fiber (type - I) and fibronectin could contribute to the clinically increased bulk of gingiva.[7] Several authors also suggested that more the number of fibroblasts present, greater the chance for recurrence.[8]

In the given case report there was no relevant personal, medical or drug history given by patient and parents. Clinically there was non-inflammatory, generalized uniform GE having no contributing factors ultimately led to a diagnosis of a generalized idiopathic gingival enlargement.

Majority of cases of GE present with chief complaint of difficulty in mastication, altered phonation and inadequacy to maintain proper oral hygiene. Mean age group of presentation is 26 years (ranges from 10 to 65 years).[9]

The recurrence of GE varies among different families or individuals of same family and the reason for the same is still undefined. The current case was managed using two approaches, first with external bevel gingivectomy in all quadrant followed by electrocautery which result in complete exposure of submerged teeth and shown remarkable correction in facial appearance, speech and mastication. After a follow-up of 12 months no sign of relapse was seen.

A case reported by *Kavvadia et al*of a 11-year-old boy with diffuse idiopathic GE which was surgically managed and have completed follow- up for 30 months with no sign of relapse.[10] On contrary, a case of hereditary GF was presented by *Kelekis-Cholakis et al* in a 13-year-old girl. All four-quadrant gingivectomy was performed followed by an orthodontic correction, after a follow-up of 3 years complete relapse was noted. Ultimately another full mouth gingivectomy had to be done. [11]

The persistent recurrence of gingival enlargement following surgery and a permanent remolding of tissue after extraction of teeth indicate that presence of teeth and the gingival crevice environment plays an important role in the pathogenesis of GF. [12] In cases of recurrence it is documented to wait until the eruption of permanent teeth before repeating the surgery. [13]

IV. Conclusion

The treatment modalities of GE varies in accordance with its etiology but maintaining proper oral hygiene and regular follow-up yield better outcome. The current case was managed surgically followed by electrocautery result in complete exposure of submerged teeth and shown remarkable correction in facial appearance, speech and mastication.

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