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CASE REPORT

Unilateral gingival fibromatosis

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ABSTRACT

Increase in size of gingiva is termed as gingival enlargement or gingival overgrowth; characterized by expansion and accumulation of the connective tissue. Enlargement of gingiva can be classified according to etiology, pathogenesis, location, and distribution. Many forms of gingival Hyperplasia are of unknown etiology and termed as idiopathic gingival fibromatosis. Gingival fibromatosis is classified as two types according to its form. The localized nodular form is characterized by the presence of multiple localized enlargements in the gingiva and the other one is most common symmetric form results in uniform enlargement of the gingiva. Our aim in this case report is to present a very interesting case of patient with the unusual clinical forms of unilateral gingival fibromatosis and to discuss the histopathological and clinical features in comparison to similar enlargements.

Key words
Fibromatosis, gingival, unilateral enlargement

INTRODUCTION

Gingival fibromatosis represents the fibrous hyperplasia of the gingival tissue. Clinical examination reveals enlargement of buccal and lingual or palatal gingival tissue. Many forms of gingival fibromatosis are of unknown etiology and termed as idiopathic gingival fibromatosis. However, several authors' use various terms such as gingivomatosis and elephantiasis to describe these lesions. Gingival fibromatosis frequently presents itself as a generalized and irregular enlargement of the attached and marginal gingiva both in the facial and lingual aspects. Various sorts of drugs (immunosuppressant's, anticonvulsants, and calcium channel blockers)[1] or genetic disorders[2,3] can lead to gingival enlargement. However, numerous non-familial examples of this condition are recognized and classified as idiopathic.[4] Gingival fibromatosis, also known as elephantiasis gingivae, hereditary gingival hyperplasia, idiopathic fibromatosis, and hypertrophied gingivae, is a rare condition characterized by slow, progressive enlargement of the gingivae.[5,6]

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The enlargement may be associated with one or more teeth, involve one or more quadrant(s), or may be generalized. The lingual or palatal gingivae are typically increased in the thickness when compared to the buccal side. In the localized form, gingival fibromatosis may remain dormant and may suddenly extend to involve other segments of one or both jaws. One distinctive but not uncommon pattern involves the posterior maxillary alveolar ridge. This is usually seen to be bilaterally symmetrical, extending posteriorly and palatally from the posterior alveolar ridges. The etiology of gingival fibromatosis is thought to be familial or idiopathic. The familial variant may occur as a remote finding or be associated with the hereditary syndromes.[7]

Our aim in this case report is to present patient with unusual clinical forms of ipsilateral gingival fibromatosis and to discuss the histopathological and clinical features in comparison to similar enlargements.

CASE REPORT

An 18-year-old female patient reported with the chief complains of gingival enlargement of maxillary and mandibular left region since 1 year [Figures 1-3] associated with mobile teeth of the same side. The gingival overgrowth has produced facial asymmetry. Patient was unable to eat from the left side. In addition, there was bleeding from left region (Maxilla and Mandible) while brushing and eating. Upon intraoral examination, Massive, Bulbous gingival enlargement was seen on left

upper and lower quadrant of maxillary and mandibular arch. There was no enlargement of the gingivae on contra lateral side. The color of the gingivae on the affected side was pinkish. The enlargement in the maxillary arch was extended from maxillary left central incisor to maxillary left 3rd molar covering almost all the surfaces of teeth. As well as in the mandibular arch, it was extended from mandibular left lateral incisor region to 3rd molar covering almost all the surfaces of teeth. Maxillary and mandibular right side was apparently normal. The involved teeth on the affected side were mobile in addition abundant local deposits were present. The family history was negative for any occurrence of gingival fibromatosis.

Radiographic investigations [Figure 4] showed severe bone loss associated with maxillary and mandibular left region. Right side of the mouth was normal. Mandibular left 1st molar was missing. Considering severe clinical attachment loss (more than 5 mm in maxillary and mandibular left side) the diagnosis of generalized severe chronic periodontitis was established, which was considered to be secondary to the gingival enlargement. Hematological profile was normal. No skeletal deformity was evident. Mental maturity of the patient was normal. Patient was not having any systemic disease.

Considering the poor prognosis of the associated teeth, decision to extract them was taken. The following teeth were extracted before surgical management of gingival overgrowth: In left maxillary Arch: 1st, 2nd premolars and 1st, 2nd molars were extracted (3rd molar was extracted during surgery). In left mandibular arch: 1st, 2nd premolars and 2nd, 3rd molars were extracted. The healing after extraction was uneventful and there was no sign of regression in the gingival overgrowth. During extraction of teeth, incisional biopsy was taken for histopathological examination, which revealed the fibromatous changes in the gingivae confirming the diagnosis as gingival fibromatosis.

Histopathological features

Histopathology section shows a lining of stratified epithelium focally keratinized with elongated rete ridges. The subepithelial tissue was composed of interlacing bundles of dense collagen interspersed with fibrovascular tissue.

Surgical management of gingival fibromatosis

Under local ane sthesia (Lignocaine 2% with 1:80,000 adrenalin as a vasoconstrictor) External bevel gingivectomy

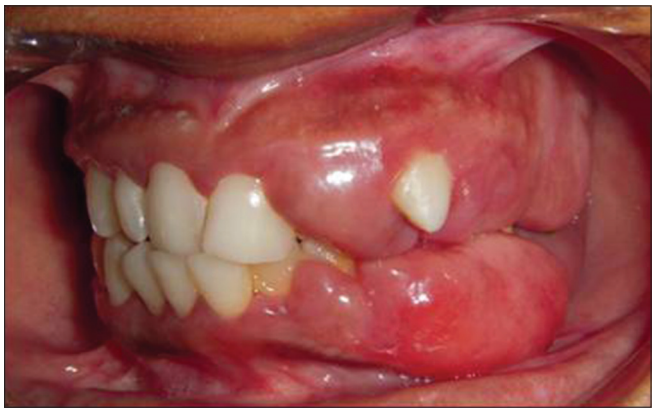

Figure 1: Pre-operative labial view showing massive and bulbous gingival enlargement on left side of maxillary and mandibular arch

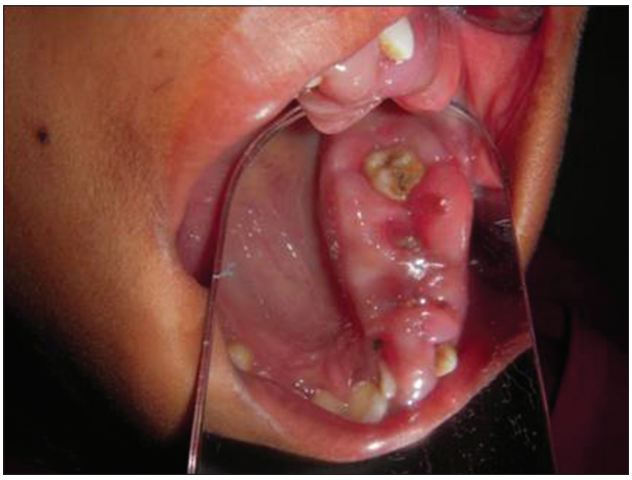

Figure 2: Pre-operative palatal view showing bulbous and massive gingival enlargement extending from maxillary left central incisor to 3rd molar region

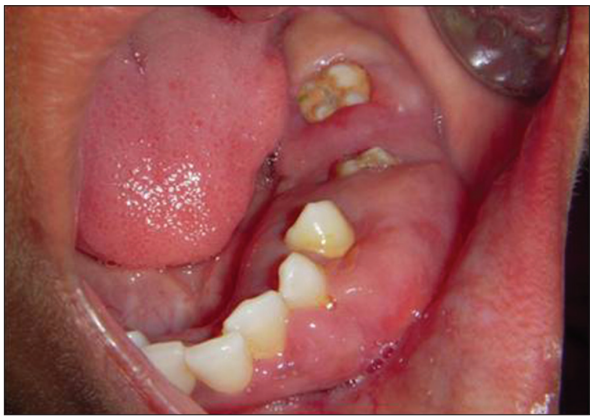

Figure 3: Pre-operative lingual view showing massive and bulbous gingival enlargement extending from mandibular left lateral incisor to 3rd molar region

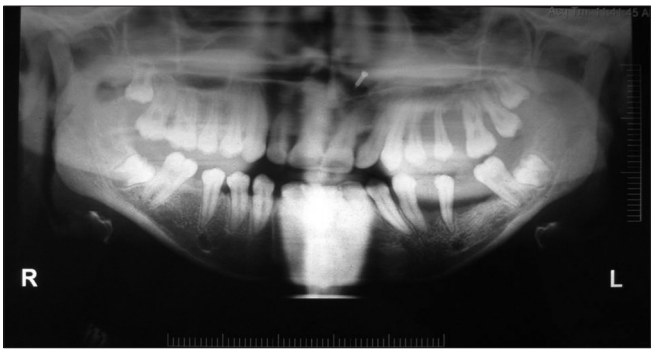

Figure 4: Orthopantomograph showing severe bone loss associated with left maxillary and mandibular arch

incisions were taken (First external bevel cum horizontal incision and then flap was reflected) to remove gingival overgrowth, then the fibrous connective tissue between buccal and palatal flap (buccal and lingual in the mandibular arch) was removed, 3rd molar was extracted in the maxillary arch and the flaps were approximated by simple interrupted sutures using 3-0 Mersilk suture (non-resorbable). All post-operative instructions and medications (Antibiotics [Penicillin] for 7 days and Analgesics for 3 days) were given. Patient was recalled after 7 days for suture removal. The healing was uneventful after 4th week [Figures 5 and 6]. After 4 weeks, patient was sent for prosthetic management (either Implant or Cast Partial Denture). The regular follow-up of the patient was done at 3rd and 6th month and was instructed for regular maintenance visit after every 6 months to observe any recurrence.

DISCUSSION

Increase in size of gingivae is termed as gingival enlargement or gingival overgrowth characterized by expansion and accumulation of the connective tissue. Enlargement of gingivae can be classified according to etiology, pathogenesis, location, and distribution. Hereditary gingival fibromatosis (HGF) is a rare gingival lesion that presents as localized or generalized enlargement of the attached gingivae and is characterized by pink, firm, and fibrotic lesion with little tendency to bleed. These distinct types of enlargement observed may resemble to the other types of gingival enlargements due to leukemic infiltration[8] and the use of drugs such as phenytoin, cyclosporine, and calcium channel blockers.[1] Furthermore, the overgrowth of the gingivae could have been a feature of several multisystem syndromes, such as Laband syndrome (ear, nose, bone, and nail defects with hepatosplenomegaly),[9] Rutherfurd syndrome (corneal dystrophy, mental retardation, and impairment of dental eruption by radicular resorption), or Cross syndrome (microphthalmia, mental retardation, athetosis, and hypopigmentation).[10] Additionally, the similar clinical appearance of these lesions to HGF might have led to misdiagnosis. These complications in the clinical diagnosis necessitate a detailed examination of the patients. Past medical history can eliminate the probability of drug-induced enlargements. Histopathological investigation may also help in clarifying the existence of neoplastic leukocytes, which are abundant in leukemic gingival hyperplasia. As to our consideration, the most difficult part in the differential diagnosis could be the presence of HGF, which is frequently associated with hypertrichosis, mental retardation, epilepsy, and familial disturbances.[6] The reported lesions were located not only in the palatal region but also in the buccal gingivae of ipsilateral of both jaws.

Royer *et al.*[11] reviewed hypothetical mechanism of gingival fibromatosis which include

- Direct fibroblast stimulation
- Production of an altered less easily degraded collagen
- Increased activity of enzyme prolyl hydroxylase which is important in polymerization of collagen.[12,13] It may be due to the patient's inherited propensity for proliferation of gingival connective tissue in response to minor irritations. Once started, the increase in size of the gingivae is conducive to further local irritation and the process becomes progressively more severe. This tendency would be at its height during the growing years of the individual. Removal of these irritants by means of strict oral hygiene and gingivectomy would modify the response and might even prevent regrowth, but at the worst, lead to slow relapse. In the present case, no relapse was observed. The enlargement has no tendency towards continued growth when the teeth were extracted as in our case.

Direct fibroblast stimulation hypothesis is in question. Recent studies suggested that fibroblasts from fibrotic tissues remain activated, even in the absence of continuous stimulation.[14-16]

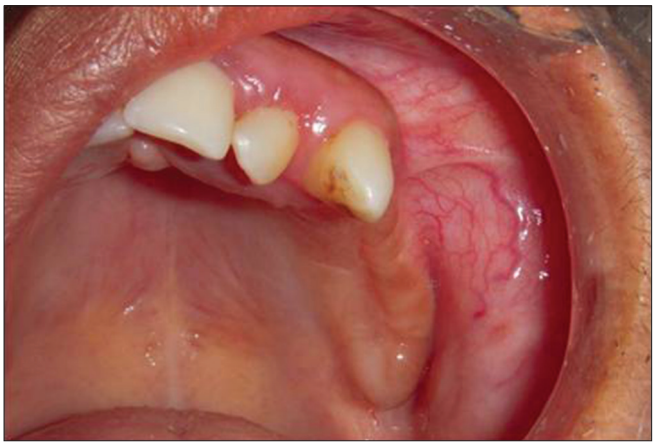

Figure 5: Post-operative palatal view showing uneventful healing of maxillary arch after 4 weeks

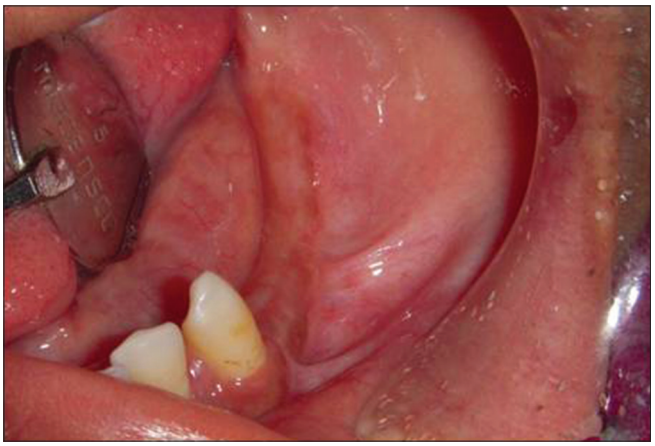

Figure 6: Post-operative lingual view showing uneventful healing of mandibular arch after 4 weeks

Histologic features of gingival fibromatosis has been focused on the connective tissue alterations, which showed an increased amount of collagen fiber bundles running in all directions associated with the few fibroblasts and the epithelium showing elongated rete pegs.[17] This histologic feature was noted in the present case.

The fibrous connective tissue presents bundles of coarse collagenous fibers, a high degree of differentiation in premature fibroblasts, and scarce blood vessels. Moreover, the epithelium with elongated papillae and hyperkeratosis is observed.[18,19] Small calcified particles, islands of osseous metaplasia, ulceration of the overlying mucosa, deposition of amyloid, and islands of odontogenic epithelium have also been reported in the previous cases.[20]

The gingival enlargement is mainly due to an increase and thickening of mature collagen bundles in the connective tissue stroma. The nodular appearance can be attributed to the thickened hyper parakeratinized epithelium. Various modalities of treatment had been proposed including radical treatment with extraction of the involved teeth, which was reported not to favor a recurrence of the growth.

There are various procedures available for removal of gingival enlargement including, surgery, electrocautery, and use of a carbon dioxide laser. If no carbon dioxide laser is available, the most effective method for removing large quantities of gingival tissue, especially, when there is no attachment loss and all the pocketing is false, is the conventional, external bevel gingivectomy.[20] Ramer *et al.* advocated quadrant by quadrant gingivectomy with periodontal pack placement for 1 week, followed by 0.2% chlorhexidine oral rinse twice a day for 2 weeks after each surgery. Careful, non-surgical, anti-infective therapy can achieve a marked improvement in many cases and complete resolution of the enlargement in some cases.[21,22] If periodontal surgical measures are necessary, external gingivectomy of the tissue can be performed conventionally with a scalpel or alternatively by laser.[23] Gingival enlargement can recur even after successful treatment hence, regular and thorough follow-up is important.

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