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Idiopathic gingival fibromatosis with unilateral aggressive periodontitis: A case report

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Case Report

Abstract

BACKGROUND AND AIM: Until now there are a few case reports in which idiopathic gingival enlargement and aggressive periodontitis are diagnosed in combination but in none of them unilateral aggressive periodontitis was seen.

CASE REPORT: A 25-year-old female was referred with the chief complaint of swelling of the gingiva in the regions of right upper and lower jaws, for the past 7 years. No relevant medical or family history was recorded. There was not any local or environmental factor. On physical examination, generalized diffused enlargement of gingiva was observed. Radiographs showed unilateral advanced destruction of supporting bone in right upper and lower sextants. The histological analysis of excisional biopsy revealed hyper parakeratinized epithelium. Enormous distribution of inflammatory cells was seen in subepithelial tissue.

RESULTS: The combination of gingival fibromatosis (GF) and unilateral aggressive periodontitis in this rare case can be a kind of syndromic form that is uncommon in GF. There is little information in this area.

KEYWORDS: Gingival Fibromatosis, Agressive Periodontitis, Unilateral Bone Loss, Gingival Enlargement

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Enlargement of gingiva is a common feature of gingival disease. The diagnosis is based on careful history and clinical examinations. This condition can be caused by several etiologies such as dental plaque, drugs, hormones, and genetic disorders. Idiopathic gingival fibromatosis (GF) is a rare condition. The cause is unknown, and thus the condition is designated as "idiopathic". The enlargement generally begins before the age of 20 years.[1]

The etiology is familial or idiopathic. The familial variation of GF may occurs as an isolated findings or to be associated to one of several hereditary syndromes, for example; Zimmermann-Laband, Murray-Puretic-Drescher, Rutherfurd, and Cross syndrome. Syndromic forms of GF may be associated with other clinical manifestations such as hypertrichosis, growth retardation, hypopigmentation, mental deficiency, epilepsy, splenomegaly, optic and auditory defects, cartilage and nail defects and dentigerous cysts. The most common clinical findings of syndromic GF includes hypertrichosis, epilepsy, and mental retardation. The enlargement affects the attached gingiva, as well as the gingival margin and interdental papillae. The facial and lingual surfaces of the mandible and maxilla are generally affected, but the involvement may be limited to either jaw. The enlarged gingiva is pink, firm, and almost leathery in consistency.[2]

One type of periodontitis is aggressive periodontitis that affects systemically healthy individuals < 30 years old, although patients may be older. Aggressive periodontitis may

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be universally distinguished from *chronic periodontitis* by the age of onset, the rapid rate of disease progression, the nature and composition of the associated subgingival microflora, alterations in the host's immune response, and a familial aggregation of diseased individuals.[3] In this paper, we report an unusual case of idiopathic gingival enlargement associated with unilateral aggressive periodontitis. Until now there are few case reports in which idiopathic gingival enlargement and aggressive periodontitis are diagnosed in the same patients but in none of them unilateral loss of attachment was seen.

Case Report

A 25-year-old female was referred to the Department of Periodontics, School of Dentistry, Kerman University of Medical Sciences, Iran, with the chief complaint of swelling of the gingiva in the regions of the right upper and lower jaws. The past history revealed that the gingiva enlarged over the past 7 years. No relevant medical and family history was observed. The patient was not receiving any medication related to gingival hyperplasia. There was no local or environmental factor.

Intraoral examination

On physical examination, generalized severe diffused enlargement of gingiva was observed. The score of gingival enlargement in right posterior upper and lower sextants was grade III and in the other sites was grade II. Teeth 3, 4, 30 showed grade III mobility, and furcation involvement was seen in teeth 3, 30. Oral hygiene was good and there were not any calculus formation even on hopeless teeth (Figure 1).

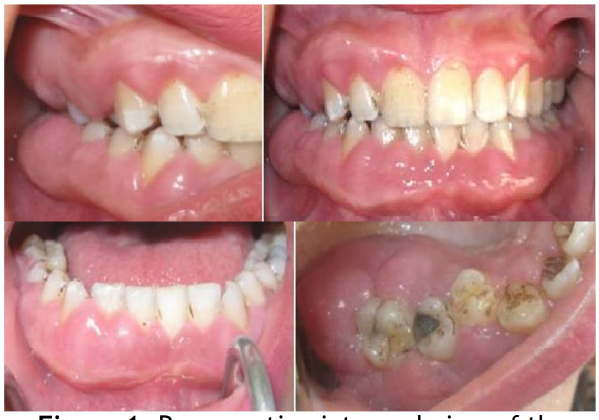

Figure 1. Preoperative intraoral view of the maxillary and mandibular arches

Radiographic findings

Panoramic and periapical radiographs showed unilateral advanced destruction of supporting bone in right upper and lower sextants. Through and through furcation involvement was seen in tooth 3, 30. There was not any bone loss in incisors or left side (Figures 2 and 3).

Treatment

Treatment included phase 1 periodontal therapy; scaling and root planning and chemotherapy of amoxicillin 500 mg/three times a day and metronidazole 250 mg/three times a day followed by periodontal surgery (Figure 4). Surgical therapy included surgical excision of enlarged tissue by internal bevel

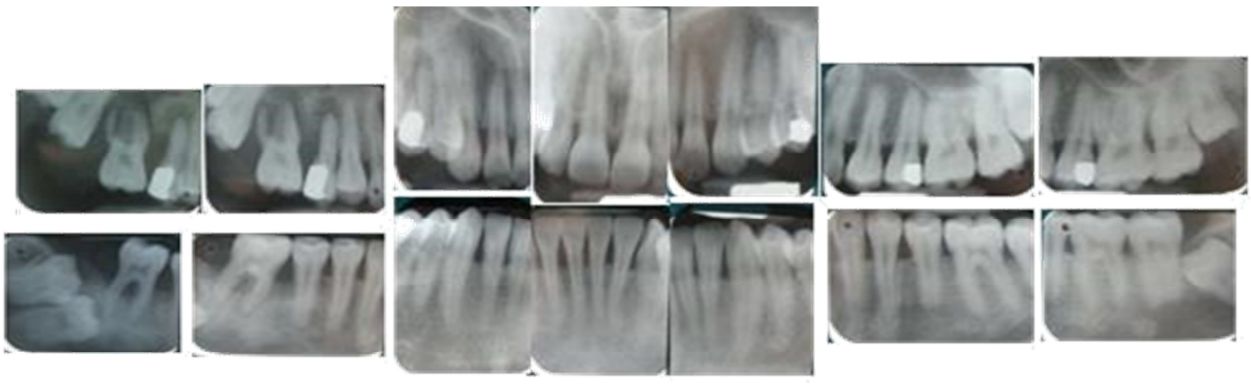

Figure 2. Full mouth periapical that revealed severe bone loss in right sextants without any bone loss in other regions

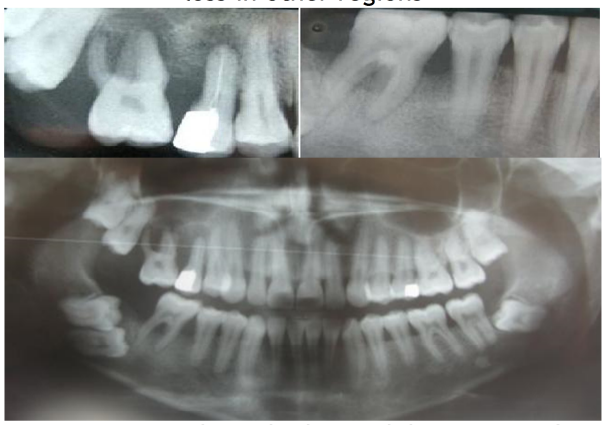

Figure 3. Unilateral advanced destruction of supporting bone in right upper and lower sextants

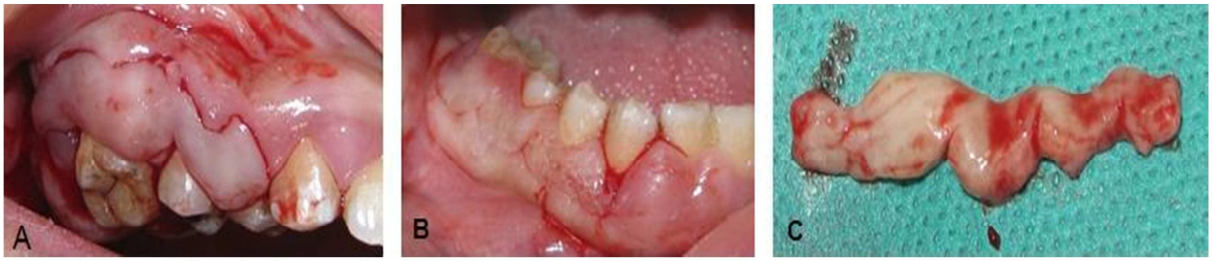

Figure 4. Surgical procedure: (A, B) internal bevel gingivectomy and (C) excisional biopsy

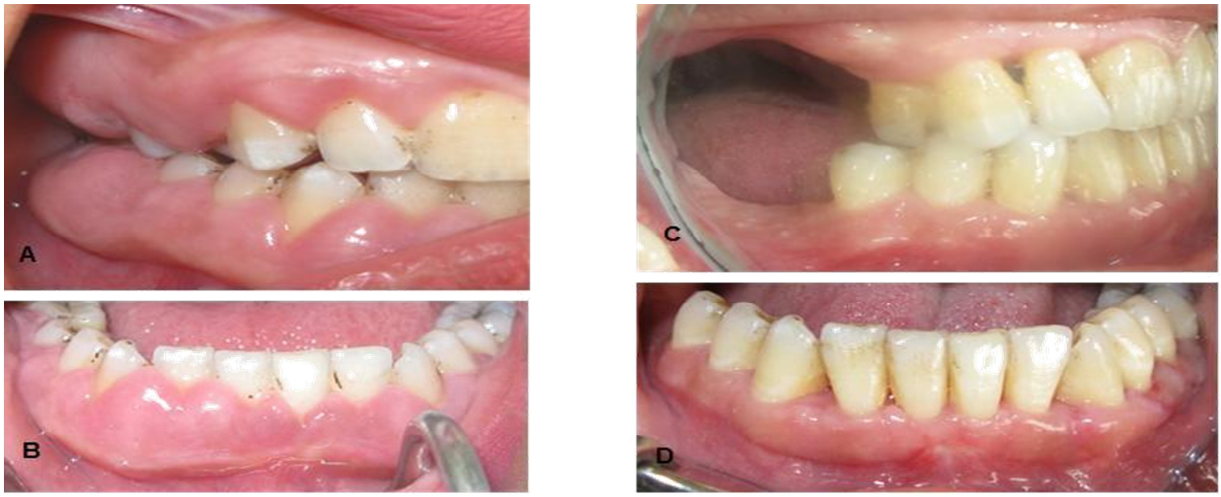

Figure 5. Photograph illustrating before (A, B) and 18 months after treatment (C, D)

gingivectomy and extraction of the hopeless teeth. Following the completion of the treatment, no signs of recurrence of gingival hyperplasia or bone resorption were observed over 18 month's follow-up (Figure 5).

Histopathologic report

An excisional biopsy of the lesion was performed during surgery (Figure 4C). Samples were fixed with 10% buffered formalin. Multiple sections with 5 µm thicknesses were obtained, stained with H&E and evaluated by light microscope (Olympus D 25, Tokyo, Japan). Microscopic evaluation of the gingival specimens revealed hyper

parakeratinized epithelium. Enormous distribution of inflammatory cells was seen in subepithelial tissue. There was large parallel collagen bundles associated with inflammatory cells in the connective tissue (Figure 6).

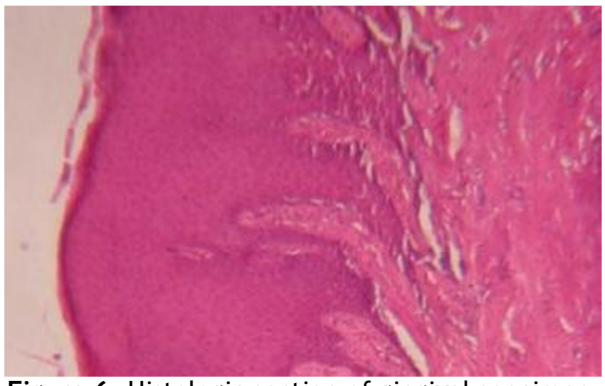

Figure 6. Histologic section of gingival specimen

Discussion

Gingival enlargement can be caused by a wide variety of etiologies. The clinicians can often diagnosis the cause by careful history, by location or by the clinical presentation. Biopsy may be needed to correctly diagnose and treat the gingival enlargement. There are several case reports that have been seen gingival enlargement associated with bone loss. Bekisz reported diffuse but unilateral gingival enlargement that in histological analysis, neurofibroma was observed.[4] Sandhu et al. reported unilateral GF with localized aggressive periodontitis.[3] In the other cases GF associated with generalized aggressive periodontitis[3-6] were reported.

Histological evaluation of these cases revealed dense, mature collagen bundles with a few fibroblasts that reached into subepithelial connective tissue. Thin long rete ridge extending into the connective tissue were also seen that were similar to our findings. In all reports, aggressive periodontitis was diagnosed but in present case unilateral aggressive periodontitis was observed where gingival enlargement was more severe than in the other side.

GF may exist as an isolated finding or as part of a more general syndrome. Syndromic type of HGF is associated with other clinical manifestations such as hypertrichosis, growth retardation, hypopigmentation, mental deficiency, epilepsy, splenomegaly, optic and auditory defects, cartilage and nail defects and dentigerous cysts. The most common clinical findings of syndromic GF include hypertrichosis, epilepsy, and mental retardation. In clinical examination on skin, eye, bone, nail and ears, none of these signs was seen in present case. The clinical expression of isolated hepatocyte growth factor is highly variable.

Gingival enlargement may be generalized to all gingival areas, or quite focal and limited in its distribution. The severity of gingival enlargement may range from slight enlargement to total coverage of the dentition. It is possible that isolated GF may result from a single gene mutation, while syndromic forms may result from alterations of multiple genes, or perhaps a gene dosage affect. It is unknown if this is due to variable clinical expression of a single gene mutation, or to allelic or non-allelic mutations.[7-9]

So the combination of GF and severe bone loss in this rare case can be a kind of unknown syndromic form that is uncommon in GF. Pattern of bone destruction in this case is similar to some disease such as histiocytosis X and Langerhans cell disease that can create same clinical findings but histopathologic and immunohistochemistry findings of excisional biopsy of gingiva and bone of socket rule out histiocytosis X. Rapid attachment loss and bone destruction, lack of inflammation despite very deep pocket and inconsistency of microbial deposits amount with disease severity in a female patient < 30 years of age lead us to diagnose aggressive periodontitis in this rare case.

It is impossible to recognize which of gingival enlargement and severe bone loss happened first but each of these diseases aggravated each other in the environment

without any local or systemic factors. Since there is little information in this area, further investigation should be carried out.

Conflict of Interests

Authors have no conflict of interest.

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