

Bimaxillary unilateral gingival fibromatosis with localized aggressive periodontitis (eating the tooth at the same table)

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Abstract

This case reports a unique presentation of two different clinical entities amidst few similarities and differences. Usually, aggressive periodontitis and gingival fibromatosis occur independently. Their simultaneous occurrence is rarely found. This report deals with the clinical features and management aspect of such a case.

Keywords: Aggressive periodontitis, idiopathic gingival fibromatosis, pubertal onset

Introduction

Gingival fibromatosis, otherwise known as gingival hyperplasia or gingival overgrowth, may occur due to a variety of etiological and pathological factors. These include inflammation, use of systemic medications, presence of systemic diseases or conditions, neoplasia or due to pseudotumors. In rare cases (1 in 750,000) people, the overgrowth can be hereditary, when it is called hereditary gingival fibromatosis (HGF). This term is synonymous with idiopathic gingival fibromatosis (IGF) as in most cases the etiology remains unknown.^[1]

IGF can occur as an isolated disease affecting only gingiva. It can occur as a part of syndrome or chromosomal abnormality and both autosomal dominant and recessive forms have been described.^[1-3] It may present itself in localized or generalized form. The symmetrical generalized form of IGF is more common.^[1] Clinically, it is a slowly progressive benign gingival enlargement of keratinized gingiva. The enlarged gingiva may be normal in color or erythematous. The consistency might be nodular or uniformly fibrous. The enlargement may

potentially cover the exposed tooth surfaces to the extent of causing functional and esthetic impairment. Alveolar bone is usually not affected, but periodontal problems might result due to pseudo pocketing.^[1]

Histologically, the fibrous connective tissue presents bundles of coarse collagenous fibers and a high degree of differentiation of young fibroblasts and scarce blood vessels. The epithelium is dense with elongated papillae and hyperkeratosis.^[2]

Various procedures are available for removal of fibromatosis gingiva. The usual treatment option includes external bevel gingivectomy using scalpel unless complicated by bony defects where in flap surgery is carried out. Other modes include removal by electrosurgery or with the application of lasers.

On the other hand, aggressive periodontitis is characterized by the rapid rate of disease progression seen in otherwise healthy individual with the absence of large accumulation of plaque and calculus with a family history suggestive of a genetic trait. It can occur in localized or generalized forms. Localized aggressive periodontitis is characterized by: Circumpubertal in onset, robust serum antibody response to infecting agents and localized first molar incisor presentation with interproximal attachment loss on at least two permanent teeth one of which is a first molar and involving no more than two teeth other than the first molars and incisors.^[4]

Management of patients diagnosed with aggressive periodontitis include non-surgical, surgical with regenerative or resective therapy and antimicrobial therapy. Non-surgical therapy involves educating the patient about the disease, its risk factors. Regenerative techniques include the use of bone grafts, barrier membranes and wound healing agents, use of enamel matrix protein. Local drug deliveries, full mouth disinfection and host modulation are other modes of treatment options.

The above mentioned pathological entities are known to occur independently. Concurrent occurrence is rare and reports are scarce.

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Hence, we report management of such a case that presented these two different pathologies occurring concurrently within the same side of the jaw.

Case Report

A 15-year-old female patient accompanied by her uncle was referred to the Department of Periodontics, ACPM Dental Collage and Hospital, Dhule, (Maharashtra) for the complaint of progressively enlarging gums. According to her, gums started growing about 1 year back beginning in the upper left back tooth region and gradually progressively involved all the teeth on that side. Similar enlargement of gums was also noticed with the lower back tooth region of that side. As the patient had difficulty in chewing from that side she continued to chew from the opposite side. Over a period of time, as the enlargement took a massive form and caused difficulty in mastication and compromised her esthetics, the school authorities brought it to the notice of her parents and they approached for treatment for it.

On further communication, the patient's past medical history and dental history was non-significant and was reporting for the first time to dental hospital.

Extraoral examination revealed a slight asymmetry of the face on the left side [Figure 1]. Intra-orally massive gingival enlargement was noted in the maxillary as well as mandibular gingiva, but only on the left side whereas the right side gingiva appeared normal. The gingival enlargement extended from distal aspect of maxillary left lateral incisor to second molar [Figure 2]. The mandibular gingiva enlargement extended from distal aspect of lower lateral incisor up to the second molar and almost encroached the occlusal surface leaving a small view of it. The degree of enlargement was categorized as grade 3 (Angelopoulos 1971) [Figure 3].

The color of the gingiva appeared pink with consistency being firm and fibrous with no evidence of stippling on the affected side. Traces of plaque, food debris and calculus were noted on the affected side. Bleeding on probing was minimal/evident/present. Periodontal examination revealed deep periodontal pockets in relation to 25, 26, 27, 35, 36 and 37. Mobility of grade 3 degree was found with 26 and 27 and grade 1 degree mobility with 25 and 35. An orthopantomograph was advised to rule out bony changes. Along with this a complete blood examination along with hormonal analysis of estrogen and progesterone were advised.

Blood examination reports revealed all parameters to be within the normal limits including hormones (estrogen and progesterone). Radiographic examination revealed severe angular bone loss localized around maxillary first and second molars and moderate bone loss around mandibular first and second molars, but of the left side only [Figure 4]. Based on onset of age, presence of minimal local factors, clinical, radiographic

and non-significant medical history and a diagnosis of IGF with localized aggressive periodontitis was made.

A biopsy was advised and sent for histopathological examination, which confirmed the features of fibrous gingival overgrowth. The section was stained with hematoxylin and eosin, which revealed thickened epithelium with elongated retepegs that penetrated deep into the connective tissue. Dense collagenous tissue bundles arranged in parallel were found scattered throughout the connective tissue. Few fibroblasts, inflammatory cells, blood capillaries were also found [Figure 5].

The usual treatment for IGF consists of surgical excision of gingiva the treatment for aggressive periodontitis initially consists of Phase 1 therapy that comprises of scaling and root planing, advise on oral hygiene instructions, administration of antibiotics, drugs amoxicillin (500 mg) and metronidazole (400 mg) for 5 days, along with anti-inflammatory drugs (ibuprofen and paracetamol) and use of chlorhexidine mouthrinses The same was carried out for the patient. The patient was discharged with oral hygiene instructions and recalled after 4 weeks.

At the end of 4 weeks, internal bevel gingivectomy quadrant wise along with open flap debridement was carried out. This procedure eliminates the pocket reduces the bulk of tissue and makes plaque control much easier. External bevel gingivectomy remains the method of choice if no bony involvement exists. Maxillary first and second molars were extracted as their prognosis was hopeless. Interrupted sutures were placed and periodontal dressing given [Figure 6]. Healing after 2 weeks at suture removal was satisfactory. Patient was recalled again at the end of 3 months, during which time there were no signs of recurrence. The clinical picture of the jaws at the end of 3 months [Figure 7]. The patient was advised to get the missing teeth replaced. Follow-up at the end of 1 year showed no recurrence of gingival overgrowth [Figure 8].

Discussion

IGF is a rare disorder characterized by proliferative fibrous overgrowth. If not in isolation it can be a feature of several syndromes such as Laband, Rutherford, Jones, Cross, Ramon, Costello. It has also been associated with family with hearing loss, hypertelorism, cherubism, growth hormone deficiency and generalized aggressive periodontitis. Whether the coexistence of these two separate entities represents a new syndromic form is not known two entities.^[1]

Here, we reported a case of non-syndromic idiopathic unilateral bimaxillary gingival fibromatosis with localized aggressive periodontitis.

The differential diagnosis of gingival overgrowth includes



Figure 1: Extra-oral view showing asymmetry of face

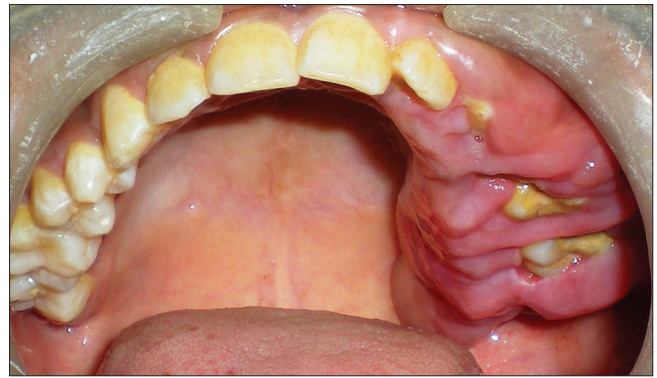


Figure 2: Unilateral enlargement of the maxillary arch



Figure 3: Unilateral enlargement of the mandibular arch



Figure 4: Orthopantomograph showing severe angular bone loss in relation to 26 and 27

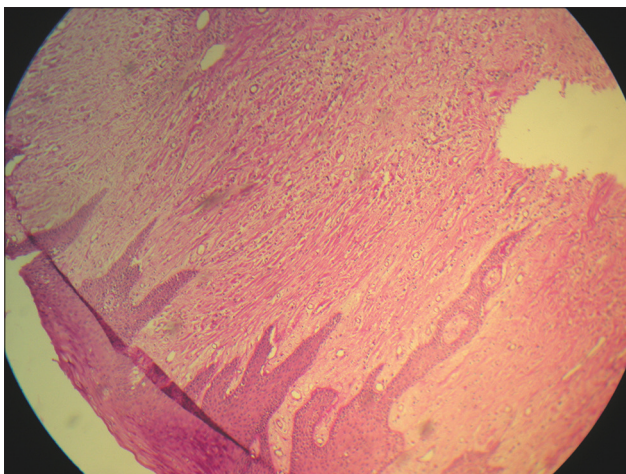


Figure 5: Histopathological picture showing enormous bundles of collagen fibers

inflammatory overgrowth, medication induced overgrowth and systemic diseases/conditions induced overgrowth.

Regarding the etiology of IGF, based on the current knowledge, studies have provided two important experimental strategies



Figure 6: Interrupted sutures placed

in understanding the mechanism of gingival fibromatosis:

- Gingival fibroblasts cultured from affected individuals generally produce (a) high levels of transforming growth factor-beta (TGF- β), (b) high rates of proliferation (c) produce increased levels of extracellular matrix and respond to TGF- β
- Genetic linkage studies have identified a specific gene *sos1* (son of sevenless). Mutation segregating HGF This mutant product is constitutively active and contributes to the uniqueness in the metabolic pathway of gingival cells and tissues

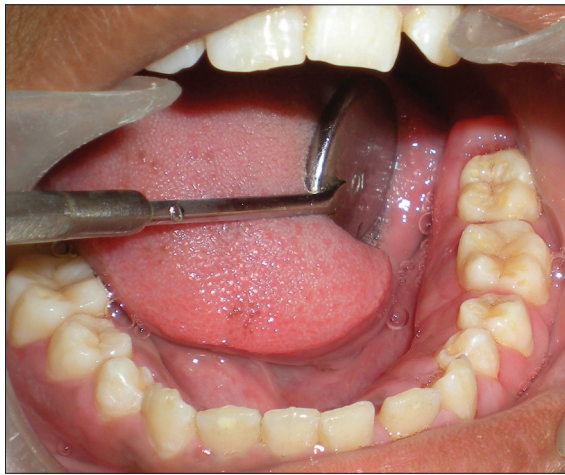


Figure 7: Post-operative view of mandibular arch after 3 months

- Modulation of apoptosis could contribute to the etiology of fibrosis in gingival tissues.^[5]

With aggressive periodontitis, the etiology appears unique with a strong genetic influence.

The similarities IGF shares with localized aggressive periodontitis are its onset around pubertal age, female predilection, hereditary background and progression of disease in the presence of minimal local factors, although secondary involvement can aggravate the pre-existing condition. Both the lesions seem to develop irrespective of effective plaque control.

Fibromatosis gingiva may hinder mastication, compromise esthetics and oral hygiene care. In the present case the patient owing to the enlargement on the left side, started to chew from the right side for almost a year. Lack of functional stimulation in this case might have led to disuse hypertrophy secondarily.

The generalized form of isolated IGF is said to be more common in male patients while the localized type is said to be more common in females.^[3] In the present case, the patient was a female. Gingival enlargement can vary from mild enlargement to such severe form that sometimes can extend covering the occlusal surfaces of crown. The enlargement may either involve one or both jaws, but bimaxillary unilateral presentation is rare as reported in the case.

IGF and aggressive periodontitis both are rare conditions. They have been usually known to occur independently. Concomitant occurrence is rare and reports regarding this are scarce. Most of information about their characteristics is based on case reports. The first of its kind was reported by Bittencourt *et al.*, in 2000.^[6] In 2007, Mahajan *et al.*, mentioned about a similar case report.^[7] In 2008 such another case was reported by Jadwat *et al.*,^[8] followed in 2010, by Vishnoi and Phadnaik.^[9] Recently, Ramachandra



Figure 8: Post-operative view of the maxillary arch at the end of 3 months

et al., reported case of gingival enlargement, along with generalized aggressive periodontitis and presence of mesiodens in a patient.^[10]

Isolated cases of gingival overgrowth have been reported since ages, but in association with aggressive periodontitis are rarely found. It is difficult to ascertain, which occurred first, localized aggressive periodontitis or gingival overgrowth since the patient gave a history of noticing the enlargement 1 year back, around upper molars of the left side and aggressive periodontitis progresses without any warning symptoms. The patient becomes aware once the masticatory function gets hampered. Since the enlargement occurred pubertal age, analysis of female sex hormones was performed which were within normal limits as depicted [Table 1].

A complete extraoral intraoral examination with radiographs, with proper medical and family history, laboratory investigations is mandatory before initiation in such a case.

In the present case, though the patient had noticed the enlargement in the maxillary left posterior aspect of the jaw 1 year back, it was brought to notice only after it took a massive form. This shows lack of awareness and negligence on the part of the patient. It is also true that aggressive periodontitis progresses silently without any warning symptoms initially. Patient becomes aware of this situation once their masticatory efficiency is hampered as happened in the present case.

Therefore, early detection is critically important in treatment of aggressive periodontitis to prevent further destruction rather than regaining what is lost. Equally, important is to create awareness and educating the patient regarding this progressive disease, its infectious role so that patient can be helped in maintaining periodontal care.

Table 1: Hormonal analysis

Hormones	Observed value	Reference value
Estradiol level in serum	158.00 pg/ml	21-251 pg/ml (follicular phase)
		21-312 pg/ml (luteal phase)
Progesterone levels in serum	4.50 pg/ml	Upto 0.3 ng/ml (follicular phase)
		12-15.9 ng/ml (luteal phase)

In the present case as the maxillary first and second molar had a poor prognosis, they had to be extracted.

Follow-up in the present was performed for a year during which there was no recurrence of gingival overgrowth; however, frequent periodontal examination is recommended and being undertaken.

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