Case Report

Idiopathic gingival fibromatosis in association with aggressive periodontitis and candidal infection: A unique case report with 7-year follow-up

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Abstract:
Idiopathic gingival fibromatosis, also called idiopathic gingival overgrowth (IGO), is a rare benign condition that occurs either in isolation or as a part of a syndrome. The overgrowth, if excess, impedes oral functions such as mastication and speech and causes cosmetic disfigurement. Diagnosis and treatment becomes challenging if the overgrowth is massive and accompanies other associated pathologies. This case reports concurrent occurrence of three pathologies, i.e., IGO, aggressive periodontitis, and candidal infection in a 20-year-old healthy male patient. The surgical procedure performed involved internal bevel gingivectomy combined with open-flap surgery. Seven-year follow-up revealed no recurrence of overgrowth and stable periodontal condition.

Key words: Aggressive periodontitis, candidal infection, gingivectomy, idiopathic gingival fibromatosis, open-flap surgery

INTRODUCTION

Gingival fibromatosis refers to a rare, benign, slowly progressive, fibrous enlargement of the keratinized gingiva. It is referred to as idiopathic gingival overgrowth when the cause is unknown. This condition is also termed as hereditary gingival fibromatosis when an inherited genetic predisposition is established.[1] It usually exhibits itself as an isolated disorder or to be a feature of several multisystem disorders such as Laband, Rutherfurd, Cross, and Prune belly.[2] The other synonyms used are elephantiasis gingivae, gigantism of the gingiva, congenital macrogingivae, and hypertrophic gingivae.

On the other hand, aggressive periodontitis is a type of rapidly progressive periodontal disease which can be further classified as localized or generalized based on the distribution of bone loss. Generalized aggressive periodontitis (GAgP) is a subtype of periodontitis that mainly affects younger patients and is characterized by episodic and rapid loss of periodontal supporting tissues.[3] The susceptibility of an individual to this type of periodontal disease is induced by a variety of host factors which include familial aggregation, single-nucleotide polymorphisms, polymorphonuclear neutrophils, antibodies to bacteria, smoking, stress, root morphology, and herpes virus infections.[4] These two pathologies usually occur independent of each other in the oral cavity and are rarely associated with each other, although recently few authors have reported their concurrent occurrences.[5-12]

Meanwhile, Candida species are opportunistic pathogens that are commonly found on the surface of the tongue, palate, and buccal mucosa in healthy individuals. These organisms are said to cause disease in hosts who are compromised by underlying local or systemic pathological processes.

As the occurrence of three such pathologies in the same patient has not yet been documented, hereby we attempt to present such a case of
idiopathic gingival fibromatosis (IGF) in association with GAgP and Candida infection in a 20-year-old healthy adult male. The treatment plan included internal bevel gingivectomy with open-flap debridement.

**CASE REPORT**

A 20-year-old young male reported to the Department of Periodontology with the chief complaint of massive gingival enlargement accompanied by difficulty in mastication and cosmetic disfigurement. History revealed that the enlargement had begun as a localized swelling initially in the lower left back tooth region and then involved the other areas and became generalized in about 2–3 months. His medical/drug history/family history was noncontributory.

Extraoral examination showed slight facial asymmetry [Figure 1], whereas intraoral examination revealed massive gingival enlargement of the upper and lower jaws. The gingiva appeared pink with bulbous and nodular type of enlargement. Palpation revealed overly firm and fibrous consistency. The enlarged tissue presented with pseudoclefts in between and almost extended to cover the crowns of posterior teeth. The anterior teeth appeared displaced. Supragingival plaque and calculus was evident but was minimal [Figure 2a-d]. Bleeding on probing was slight. As the enlargement impeded a detailed periodontal examination, an orthopantomograph (OPG) was advised to discern bony changes. Radiographic examination revealed moderate-to-severe bone loss with a combination of horizontal and angular type of bone loss with most of the dentitions. Angular-type bone loss was found with 15, 16, 17, 34, 35, 36, 37, 45, and 46 regions, whereas horizontal bone loss was evident with remaining teeth as depicted in OPG [Figure 3].

Based on the clinical and radiographic findings, a tentative diagnosis of IGF in association with GAgP was made. Further investigations included a routine hemogram, qualitative analysis of neutrophil defects, and histopathological examination.

After the hematological parameters were found within normal limits, a part of the fibrous tissue was excised under local anesthesia and sent for histopathological examination [Figure 4].

**Histopathology**

Histopathological report revealed stratified squamous epithelium with underlying subepithelial tissue showing moderate proliferation of fibrocollagenous tissue with infiltration of plasma cells, eosinophils, and eosinophilic bodies suggesting fibromatosis of the gingiva. In addition, the report also revealed the presence of Gram-positive budding yeasts, when stained with periodic acid–Schiff stain, suggesting the presence of fungal infection. The histopathologist further recommended to send the scrappings from the biopsy site which later on, upon isolation, showed the presence of Candida albicans [Figure 5]. Hence, the diagnosis was reported as gingival fibromatosis with secondary fungal infection and isolated organisms as C. albicans.

**Neutrophil activity analysis**

For the assessment of neutrophil defects, venous blood was drawn in a test tube containing ethylenediaminetetraacetic acid and was sent to the laboratory. Qualitative analysis of neutrophils activity was performed for the following tests: chemotaxis, respiratory burst, intracellular killing, and phagocytosis. The tests revealed a defect in chemotactic activity and abnormal respiratory burst.

**Treatment**

As a part of Phase I therapy, scaling and root planing was carried out along with advice of oral hygiene instructions [Figure 6]. The patient was recalled after 15 days for re-evaluation of overgrowth. After 2 weeks, the surgery was carried out quadrant wise under local anesthesia. It involved excision of the gingiva using internal bevel gingivectomy with open-flap debridement. No attempt was made to regenerate the bony defects since they were noncontaining defects. The flaps were sutured with silk sutures, and periodontal dressing was given [Figure 7a-c]. The patient was prescribed a combination of antibiotics, amoxicillin (500 mg) and metronidazole (400 mg) along with anti-inflammatory drug, ibuprofen 400 mg for 7 days. The use of 0.2% chlorhexidine mouthwash was recommended. The sutures were removed after 10 days. The same procedure was carried out for all the remaining teeth. Healing was uneventful. The patient reported after three months for follow up. The clinical appearance at the end of three months [Figure 8a-c]. The patient was put on recall therapy for every 3 months initially and then for yearly follow-ups. The patient appeared satisfied with functionality of oral cavity and good esthetics. Subsequently, the patient reported every year for follow-up visits till 7 years with no untoward events. The following are the clinical photographs at the end of 2 years, 4 years, and 7 years and OPG of 2 and 4 years postoperatively [Figures 9 and 10]. Seven-year follow-up OPG could not be taken due to inevitable reasons.

**DISCUSSION**

Gingival fibromatosis and aggressive periodontitis are comparatively two different and rare clinical entities with varying etiologies and different clinical features. They usually occur independent of each other. Sometimes, both conditions represent themselves as part of syndromes.[12] The present case was nonsyndromic type. For nonsyndromic forms of gingival fibromatosis, chromosomes 2p21–p22, 5q13–q22, and Son of Sevenless-1 gene have been linked as the predisposing factors. So far, the studies have reported inconclusive evidences.[3] In the present case, genetic analysis was not performed.

Massive gingival fibromatosis covers the crowns of the teeth and causes esthetical, functional, and psychological problems, which need to be addressed meticulously for the well-being of the patient. The treatment of isolated gingival overgrowth without bone involvement is surgical excision either through the use of scalpels, lasers, or electrocautery. This pathology, if combined with bone loss, is treated with open-flap debridement along with gingivectomy.

Etiologically, genetic segregation analysis studies have contributed a major role in describing the transmission of aggressive periodontitis through autosomal dominant/recessive and x-linked forms.[4] Furthermore, it is said that not every generation is affected in recessive type. In the present case, the familial history was negative in relation to aggressive
periodontitis, as the patient’s mother and his siblings who were assessed did not present with any clinical findings such as bone loss. Hence, this case could be a recessive-type trait.

The key protector cell against any bacterial infection is the polymorphonuclear leukocyte. Any abnormal function of these cells in relation to adherens, chemotaxis, superoxide generation, phagocytosis, and bactericidal activity often results in increased susceptibility to periodontitis. Chemotaxis is a receptor-mediated event that is initiated by chemotactic factors forming a concentration gradient that directs the approach of phagocytic cells. Moreover, abnormal respiratory burst phenomenon refers to increased level of superoxide generation in response to bacterial infection. Both events cause destruction of the periodontal supporting structures. These dysfunctions have been reported in patients with aggressive periodontitis.\[4\] In the present case, an abnormality in chemotaxis and abnormal...
Fungal organisms are commonly found on the tongue, palate, bucal mucosa, and gingival sulcus of healthy individuals. Most frequently isolated and the one often associated with oral lesions is *C. albicans* species. They have been recovered from periodontal pockets in 7.1%–19.6% of patients with chronic periodontitis and also in aggressive periodontitis. A higher prevalence of these was also reported in the subgingival biofilm of human immunodeficiency virus-seropositive patients.[13] In the present case, the patient was systemically healthy and presented with clinical findings of aggressive periodontitis. The presence of *C. albicans* could have been an incidental finding.

It is also said that the exact role of *C. albicans* in the development or progression of periodontal disease is not known, but their ability to adhere to epithelial cells, colonize, and proliferate in deep gingival connective tissue with activation of local and systemic defense factors may contribute in the development and progression of different type periodontal diseases. In the present report, budding yeasts were noted incidentally by the histopathologist during microscopic examination which further led to recognition of *C. albicans*. These organisms were the cause, or the effect of such destructive lesions needs to be further elicited.

Hence, from the above studies, it can be inferred that although *Candida* species can be considered as part of normal oral flora, they can be also found in chronic, aggressive periodontitis cases and in immunocompromised individuals but by how manyfold these microorganisms are increased further to cause tissue destruction needs to be warranted. In the present case, the patient was not specifically treated for fungal infection as it seemed unnecessary by the concerned physician.

**CONCLUSION**

This is the first case reporting the triology of pathologies occurring concurrently, i.e., gingival fibromatosis, aggressive periodontitis, and *Candida* infection. The treatment approach included internal bevel gingivectomy with open-flap surgery. Seven-year follow-up...
of the patient revealed no recurrence of gingival fibromatosis with stable periodontal condition. The presence of candidal infection along with such pathologies is debatable. Patient education and motivation remain undisputable part of treatment plan.

Declaraton of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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