

MANAGEMENT OF A RARE CASE OF IDIOPATHIC GINGIVAL FIBROMATOSIS WITH GENERALIZED AGGRESSIVE PERIODONTITIS

Deshmukh, Rajendra Shankar and Patil, Anjali Madhukar

¹Department of Oral Pathology and Micro, C.S.M.S.S. Dental College and Hospital, Aurangabad, Maharashtra, India

²Department of Periodontology, C.S.M.S.S. Dental College and Hospital, Aurangabad, Maharashtra, India

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Abstract

Gingival fibromatosis is a heterogeneous group of enlargement characterized by progressive increase in submucosal connective tissue elements. Many Cases are iatrogenic and some are inherited or idiopathic. Idiopathic gingival fibromatosis, is a benign slow growing proliferation of the gingival tissue, is genetically heterogeneous. This condition is usually part of a syndrome or rarely an isolated disorder. Aggressive Periodontitis, another genetically transmitted disorder of the periodontium, typically result in severe rapid destruction of the tooth supporting apparatus. Gingival overgrowth as a clinical characteristic of Idiopathic gingival fibromatosis causes many dental complications which worsens patient's adaptation in daily emotional, social and functional requirements. Here we present a rare case of a nonsyndromic idiopathic gingival fibromatosis associated with generalized aggressive periodontitis in a 23 year old female. The diagnosis was made based on history, Clinical examination, radiographic findings and histopathology. Gingivectomy was carried out in all four quadrants under local anesthesia. No recurrence was observed during 2 years follow up and patient showed remarkable esthetic and functional improvement.

Keywords: Aggressive Periodontitis, Gingival fibromatosis, Gingivectomy, Idiopathic gingival fibromatosis, Syndrome.

I. Introduction

Gingival fibromatosis is slowly progressive gingival enlargement caused by collagenous overgrowth of the gingival fibrous connective tissue.^[1] Many Cases are iatrogenic some are inherited while others are Idiopathic .^[2] Gingival enlargement can be due to plaque accumulation, due to poor oral hygiene, inadequate nutrition , or systemic hormonal stimulation.^[3] Gingival enlargements are also seen in several blood dyscrasias e.g. leukemia , thrombocytopenia or thrombocytopathy.^[4] Idiopathic gingival fibromatosis is a rare hereditary condition that has no definite cause.^[5] Investigations are in evolution to establish the genetic linkage and heterogeneity associated with it.^{[6] [7]} This condition may manifest as an autosomal dominant or, less commonly, an autosomal recessive mode of inheritance , either as an isolated disorder or as part of a syndrome.^{[8] [9]} The syndromes associated with gingival fibromatosis and the clinical features are presented in "Table" 1. Autosomal dominant forms of gingival fibromatosis which are usually nonsyndromic , have been genetically linked to the chromosomes 2p21- p22 and 5q13- q22.^[10] In modern

times, a mutation in the son of sevenless-I (SOS-I) gene has been suggested as a possible cause of isolated (nonsyndromic) gingival fibromatosis.

However no definite linkage has been established. [11]

A typical case of idiopathic gingival enlargement presents large masses of firm, dense, resilient insensitive growth that covers the alveolar ridges and extends over the teeth. [12] Associated Clinical problems include poor esthetics, prolonged retention of deciduous teeth, abnormal occlusion, inadequate lip closure, difficulty in eating and speaking. [1] Aggressive periodontitis comprises another group of genetically inherited disease that represent a severe and rapidly progressive form of periodontitis. Characteristically, these diseases tend to exhibit familial aggregation and there are no factors in the medical history that seem associated with the condition. The disease appears to be the result of a defect in the immune response rather than plaque and calculus deposition. It has been shown by many investigators that patient with aggressive periodontitis display functional defects of PMN, monocyte or both, but without any systemic manifestations. This results in reduced defensive ability against some of the periodontal pathogens. [13] Generalized aggressive periodontitis usually affects people under 30 years of age. They have poor serum-antibody response to infecting agents and pronounced episodic destruction of the attachment and alveolar bone. The loss of attachment affects at least 3 permanent teeth other than first molars and incisors. [14] Here we report an unusual case of a nonsyndromic, idiopathic gingival fibromatosis associated with generalized aggressive periodontitis.

II. Case Report

A 23 year old female reported to the department with a chief complaint of swollen gums involving all her teeth since last 3 years. The enlargement was slowly progressive and painless. There was also difficulty in mastication and while brushing the teeth. Her family history was non-contributory. There were no relevant findings in her medical history. She did not give any history of drug intake, pregnancy and systemic disease that could contribute to the gingival enlargement.

2.1 clinical features:

Extraoral examination revealed a symmetric face with bimaxillary protruding profile and lip incompetence. Bulbous gingivae were seen through the lips.

Intraoral examination revealed generalized diffuse enlargement of gingivae that involved both the maxillary and mandibular arches. (Fig1 A & B) Gingival enlargement was severe in maxillary and mandibular posterior region covering almost three quarters or more of the crowns of teeth. (Fig 2 A & B). There was a slight bleeding on probing. The gingiva was pink, edematous and soft in consistency with absence of stippling. The degree of gingival enlargement was scored as grade III in maxillary and mandibular posterior regions on both the sides. Grade II in mandibular anterior region and grade I in maxillary anterior region. [15] The oral hygiene status of the patient was fair. The patient had generalized tooth mobility. There was grade II mobility with 22, 31, 34, 37, 41 and 42 and grade I mobility with 11, 12, 21, 24, 32, 33, 35 and 36. Generalized periodontal pockets (pocket depth ranging from 6-10 mm with

average 7.3 mm) were present. Grade II furcation involvement with 36, 37, 46 and Grade III with 47 was noted. Malpositioning of the teeth were observed in posterior region of maxillary and mandibular arches on left side.(Fig 2 A & B) All the hematological investigations were within normal limits. Intraoral panoramic radiograph revealed generalized alveolar bone loss. (Fig 3) Based on all these findings, a provisional diagnosis of Idiopathic gingival enlargement with generalized aggressive periodontitis was made.

2.2 histopathological investigations:

Under local anesthesia, a biopsy specimen was obtained from the hyperplastic gingiva during surgery and sent for histopathological examination. Multiple serial sections were prepared and stained with hematoxylin. & eosin. Section showed hyperplastic overlying epithelium with elongated rete ridges. The underlying connective tissue showed densely arranged collagen fiber bundles with numerous fibroblast and varying amount of chronic inflammatory cell infiltration. (Fig. 4).Based on clinical history, radiographic findings and the histopathological examination the case was diagnosed as Idiopathic gingival fibromatosis with generalized aggressive periodontitis.

2.3 management:

Full mouth undisplaced flap or internal bevel gingivectomy. [16] was carried out to treat gingival enlargement as well as aggressive periodontitis. Oral hygiene was meticulously supervised during peri-surgical and follow up period. (Fig 5-8)

2.4 Follow-up:

The case was followed 6 weeks postoperatively and then every 3 months for 2 years. The mobility of the teeth was reduced to physiologic at the end of 3 months. No recurrence was observed within 2 years (Fig. 8).

Patient is still following the follow up regime.

III. Discussion

Gingival fibromatosis is a rare benign slow growing fibrous overgrowth of the gingiva with great genetic and clinical heterogeneity. [17] It may exist as an isolated abnormality or as a part of a syndrome. As an isolated finding, it is mostly sporadic, but an autosomal dominant inheritance pattern is also possible. Rarely, autosomal recessive inheritance is found.[4] The overgrowth might be caused by several etiological factors such as administration of specific drugs, (e.g. Phenytoin, cyclosporine, nifedipine) inflammation, systemic conditions.(e.g. Leukemia). Hormonal conditions (e.g. Pregnancy, Puberty or hypothyroidism), nutritional conditions (e.g. vitamin C deficiency). A progressive fibrous enlargement of the gingiva is a feature of Idiopathic fibrous hyperplasia of the gingiva. [4] Idiopathic gingival fibromatosis is a rare hereditary condition that has no definite cause.[5]. Aggressive periodontitis is a rapidly progressing type of periodontitis and has a familial tendency suggesting a genetic predisposition.[12]

In the present case, patient had no history of any systemic disease, hypertrichosis, mental retardation, epilepsy or medication which could contribute to gingival overgrowth. she also did not give history of

pregnancy. General physical examination of the patient revealed no syndromic association which could contribute to gingival overgrowth. The clinical, histopathological features and systemic examination excluded the diagnosis of neoplastic enlargement. Gingival enlargement also is a symptom of scurvy; however our patient did not have any other signs of scurvy such as petechiae, echymoses or spontaneous bruising of the extremities. All the hematological investigations were within normal limits. Panoramic radiograph of our patient demonstrated generalized alveolar bone loss. Histopathological examination revealed hyperplastic epithelium with elongated rete ridges and densely arranged collagen bundles. These histological findings of our patient were consistent with those for fibrous gingival hyperplasia. The diagnosis of Idiopathic gingival fibromatosis with generalized aggressive periodontitis was made based on patient's medical and family history, clinical presentation, radiographic findings and histopathological examination. Similar cases of Idiopathic gingival fibromatosis associated with aggressive periodontitis have been reported by Chaturvedi R.(2009), ^[11], Sanadi RM (2010) ^[18], Fatema S. , Preeti M. (2012) ^[19] and Casavechia P,Uzel ml , Kantari A, Hasturkh , Dibots, Hart (2004) ^[20]. The patient in the present case was troubled by esthetics and chewing function hence gingivectomy was carried out in all four Quadrants. Patient was kept under observation to see any kind of recurrence. No recurrence was observed during 2 years of follow up period and patient showed remarkable esthetic and functional improvement.

IV. Conclusion

Present case was an Idiopathic gingival fibromatosis with generalized aggressive periodontitis and its management. Idiopathic gingival fibromatosis with generalized aggressive periodontitis is an uncommon entity. The unusual coexistence of these two pathological processes presents challenge for the dental professionals and could be evaluated as a new syndrome in the near future. However, further research is needed to establish a syndromic association between the two conditions based on genetic and linkage studies.

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