

Hereditary Gingival Fibromatosis - A Case Report

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Abstract Hereditary gingival fibromatosis is characterized by a slow benign enlargement of gingival tissue. It causes teeth being partially or totally covered by enlarged gingiva, causing esthetic and functional problems. It is usually transmitted both as autosomal dominant trait and autosomal recessive inheritance although sporadic cases are commonly reported. This paper reports three cases of gingival fibromatosis out of which one was in a 15 year old girl treated with convectional gingivectomy.

Keywords Hereditary gingival fibromatosis, Inherited disease, Gingivectomy

1. Introduction

Hereditary gingival fibromatosis (HGF) or Idiopathic gingival fibromatosis is a rare, benign, asymptomatic, non-hemorrhagic and non-exudative proliferative fibrous lesion of gingival tissue occurring equally among men and women, in both arches with varying intensity in individuals within the same family [1]. It occurs as an autosomal dominant condition although recessive form also does occur. Consanguinity seems to increase the risk of autosomal dominant inheritance. It affects the marginal gingival, attached gingival and interdental papilla presenting as pink, non-hemorrhagic and have a firm, fibrotic consistency [2].

It also shows a generalized firm nodular enlargement with smooth to stippled surfaces and minimal tendency to bleed. However, in some cases the enlargement can be so firm and dense that it feels like bone on palpation [3]. Here we present three cases of gingival fibromatosis out of which one was in a 15 year old female patient.

2. Case Reports

A 15 year old female patient came to the department of periodontics at CDC hospital, Lucknow (U.P), India with chief complaint of generalized gingival swelling since 3 years (Fig. 1). On accounting detail history the patient revealed that the swelling initiated slowly and has assumed the present size within 3 years. On further communication with the patient it was revealed that her mother was also

having the gingival enlargement before the patient's birth and she got operated in the village government hospital. No further relevant medical history was present.



Figure 1. Gingival enlargement

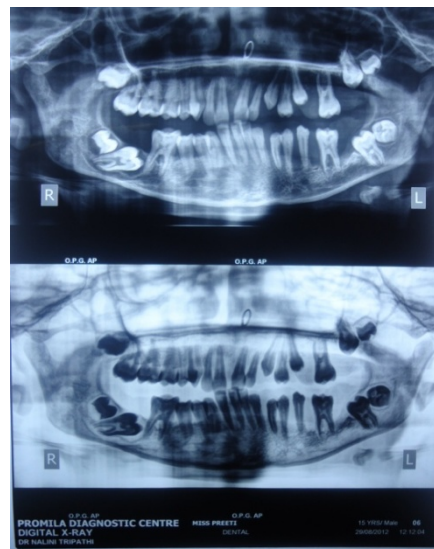


Figure 2. OPG showing impacted molars

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On intra oral examination there was presence of generalized firm swelling of the gingiva covering cervical and middle third of the clinical crown in anterior teeth and incisal surface in posterior teeth. A complete hemogram was conducted with all the parameters within normal range. An Orthopantomograph (OPG) has been taken which showed the presence of impacted molars in all the four quadrants along with bone loss and drifting of teeth (Fig. 2). An incisional biopsy was taken for histopathological which showed fibromuscular tissue covered by stratified squamous surface epithelium. The stroma shows irregular fibrosis and infiltration by chronic inflammatory cells with no evidence of malignant change (Fig.3). On the basis of histopathological report a confirmed diagnosis of gingival hyperplasia was made.

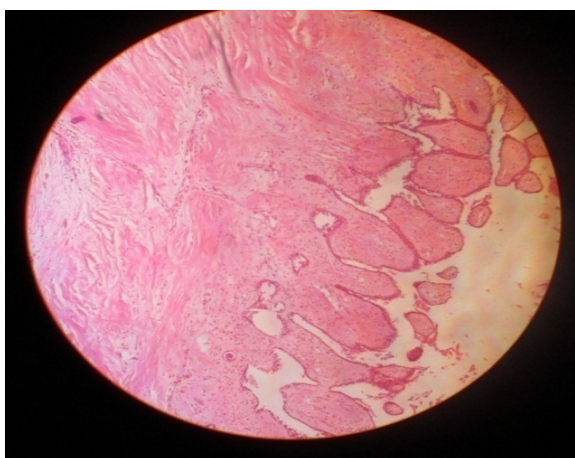


Figure 3. Showing irregular fibrosis & inflammation



Figure 4. Showing the surgery performed under LA

The treatment plan consisted of gingivectomy which was performed under local anesthesia after getting patient consent (Fig. 4). The patient was prescribed antibiotics orally for ten days and was kept under follow up for six months. There was no untoward consequences and relapse reported by the patient post operatively during the follow up (Fig. 5). Earlier the treatment plan was SRP (scaling and Root Planing) with chemotherapeutic agent for 14 days but the outcome was not satisfactory as the enlargement didn't reduced nor was it progressive. Electrocautery and LASER

therapy also didn't work because in the case of electrocautery layerwise cutting is performed and the tissue is too thick to be cut layerwise and moreover it is time consuming for the patient as well as surgeon. Similarly was the reason with LASER, it can't be used at the increased watt as desired for the oral tissue and also due to the presence of submandibular and sublingual glands. The conventional scalpel therapy is used because with it the whole tissue can be cut at one time and with single incision.



Figure 5. Post operative showing no recurrence

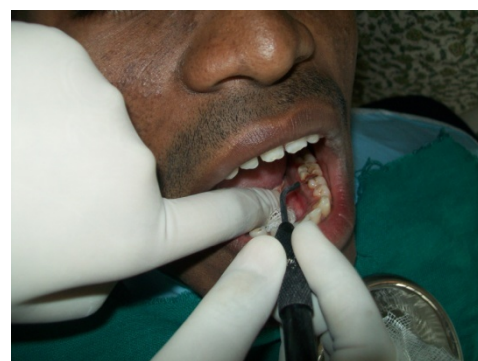


Figure 6 & 7. Showing localized gingival enlargement treated with electrocautery in cutting and coagulation mode

A 25 year old female patient came with the chief complaint of localized gingival enlargement in the lower left molar region since 6 months. On intraoral examination there was presence of enlarged tissue with relation to interdental papilla between 45 and 46 i.e. lower left second premolar and first molar. It was hard in consistency and extending to the occlusal surface of molar. No relevant medical and family history was given by the patient. On obtaining the patient

consent the treatment was done with removal of enlarged tissue with electrocautery in the cutting and coagulation mode (Fig 6). The patient was kept on antibiotic and chlorhexidine mouthwash for 15 days after which there were no untoward consequences reported by the patient.

A 27 year old male patient reported with chief complaint localized gingival enlargement in the lower left molar region since one year. On intraoral examination there was presence of enlarged tissue on lingual side with relation to interdental papilla between 45 and 46 (lower left second premolar and first molar). It was also hard in consistency and extending to the occlusal surface of molar. No relevant medical and family history was given by the patient. On obtaining the patient consent the treatment was done with removal of enlarged tissue with electrocautery in the cutting and coagulation mode (Fig 7). The patient was kept on antibiotic and chlorhexidine mouthwash for 15 days after which there were no untoward consequences reported by the patient.

3. Discussion

Hereditary gingival fibromatosis is also associated with Rutherford, Laband, Murrey-Puretic-Drescher, Cross and Ramon syndrome. It is also seen in Lysosomal storage disease [3]. It is a rare benign fibrous growth of gingiva with genetic and clinical heterogeneity. It manifests as an isolated form which is mostly sporadic or with an syndrome. The etiological factors contributing for gingival fibromatosis reported were administration of certain drugs (Phenyton, Cyclosporine, Nifedipine), inflammation, systemic conditions (leukemia), hormonal conditions (Pregnancy, puberty or Hyperthyroidism) and nutritional conditions like Vitamin C deficiency [4].

In the present case the patient has not reported any systemic disease or any dependency on medication. On clinical examination neither syndromic signs were revealed nor the hemogram did showed any abnormality. It was assumed that the patient may be suffering from scurvy but there were no signs of petechiae, echymosis or spontaneous bruising of the extremities.

A chromosomal abnormality involving 2p21-p22 and 5q13-q22 has been found in HGF. A mutation in the *son of sevenless-1* (SOS-1) gene has also been suggested as the possible cause. An unknown cause of HGF confined to the fibroblasts in the gingivae has been suggested which says that there is an increase in the proliferation of gingival fibroblasts or increased collagen synthesis rather than decreased levels of collagenase activity may be involved [5]. It was reported that HGF is characterized by excess accumulation of extracellular matrix (ECM) and fibroblasts cause remodeling of ECM, which causes disturbed collagen turnover or change in fibroblast proliferation resulting in gingival overgrowth. Likewise, oral keratinocytes modulate ECM protein expression and activity of fibroblasts; whereas keloid keratinocytes promote proliferation and production of collagen of underlying fibroblasts and inhibit apoptosis indicating the role of epithelium-connective tissue in

diseased condition. This suggests that epithelium in HGF plays a part in pathogenesis of gingival overgrowth [6].

Integrins are heterodimeric membrane glycoproteins involved in cellular processes like cell-ECM and cell-cell interactions. The gene expression of collagen and MMP-1 is regulated by $\alpha 1\beta 1$ and $\alpha 2\beta 2$ where $\alpha 1\beta 1$ reduces the expression of collagen while $\alpha 2\beta 2$ stimulates collagen and collagenase gene expression in both two and three dimensional cultures. Although, excess ECM accumulates in HGF, it is reported that these integrins play important role for this change to occur. Heat shock protein (Hsp) 47 which is the major collagen binding heat inducible glycoprotein in fibroblasts found to be involved in several fibrotic diseases such as connective tissue diseases and dermal fibrotic diseases. Its expression is found in basal and suprabasal layers of HGF epithelium [7].

As stated non-syndromic forms of HGF are transmitted as autosomal dominant traits and gene locus is present on chromosome 2p. Based on FISH analysis it was found that there are 2 gene loci on the short arm of chromosome 2 located in 2p21-2p22 and 2p13-p16 [8]. It is found that the patient with HGF shows the sign of failure of eruption of permanent molars which is consistent with our case also as the permanent molars in our case failed to erupt. Maxillary molars impaction can be due to abnormal osteofibrosis of alveolar bone while impaction of mandibular molars implies abnormal development of dental germ near to lower margin of mandible [9].

The most characteristic feature of HGF is gingival enlargement of both maxillary and mandibular gingiva. In case of maxilla the enlargement can be due to both osteofibrosis and fibromatosis while in the mandible there is only fibromatosis without the mandibular bone affected which is seen in our case also [9]. Another feature is absence of involvement of periodontal ligament and the enlargement occurs peripheral to the alveolar bone within the attached gingiva [10] which is consistent with our case also as there is focal involvement of periodontal ligament at buccal area of molar only.

The treatment modalities available for HGF include surgery, electrocautery and use of carbon dioxide laser. In the absence of carbon dioxide laser the most preferred method is conventional, external bevel gingivectomy, which should be done quadrant by quadrant with periodontal pack placement for 1 week followed by 0.2% chlorhexidine oral rinse for 2 weeks [11]. Another treatment proposed for HGF includes radical surgery with the extraction of involved teeth which can be not convincing related to patient's age and condition of teeth. To improve esthetics as well as treat periodontal disease the method of choice remains is undisplaced flap surgery [12, 13]. The classic treatment till today in developing countries is removal of excess gingival growth by scalpel but one should remember about the periodontal treatment which should be done before commencing the surgical part of the treatment plan [14]. It is reported that the patient of HGF undergoes repeated surgeries; therefore careful counseling of both parents and

patient is of paramount importance. Education and motivation of patient should be properly done and is essential for maintenance of proper oral hygiene and regular recall visits [15].

In the present case the method of treatment implied was surgery by scalpel method along with oral prophylaxis. The patient was counseled for repeated follow ups every month but it turned for 4 months only without any post operative complications and recurrence.

4. Conclusions

The present case was of hereditary gingival hyperplasia (HGF). Patients with this condition require great care and motivation. It should be remembered that even correctly performed gingivectomy may not prevent from recurrent illnesses. To lower the risk of recurrence, eliminating the irritant, correct dental hygiene and regular cleaning along with frequent controlled visits should be performed.

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