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Nonsyndromic Hereditary Gingival Fibromatosis: A Case Report

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Abstract

Generalized gingival enlargement can be caused by a variety of etiological factors. Hereditary gingival fibromatosis (HGF) is a rare genetically inherited condition, which manifests itself by fibrous gingival overgrowth. It can develop as an isolated disorder or can be a feature of a syndrome. This paper presents a case report of a 14-year-old female who presented with a generalized gingival overgrowth, involving the maxillary and mandibular arches and covering almost the whole dentition. Patient's medical and drug history is not contributory. All the investigations were within the normal physiological limits. The enlarged gingival tissue is surgically excised by conventional gingivectomy procedure and through periodontal flap technique. Postoperatively, patient's esthetic and function was improved and there was no recurrence of the condition upto 3 months follow up period.

Keywords: Hereditary gingival fibromatosis, gingival hyperplasia, gingivectomy, idiopathic gingival enlargement.

Introduction

Gingival fibromatosis denotes the fibrous hyperplasia of the gingival tissue. It can present as Hereditary Gingival Fibromatosis, which may appear as an isolated entity or as part of a genetic disease or syndrome, as drug-induced gingival overgrowth (DIGO, GO) or as idiopathic gingival fibromatosis (IGF).[1] The other names of this condition are gingivomatosis, gingival enlargement, gingival hyperplasia, gingival overgrowth (GO), elephantiasis gingivae, familial elephantiasis, gigantism of the gingiva, and congenital macrogingivae.[2]

Goddard and Gross first reported HGF in 1856, the mode of inheritance is believed to be autosomal dominant although some cases of a recessive mode of inheritance have been also reported. [3,4,5] Chromosome 2p21-p222 and 5q13-q22 have been genetically linked to the autosomal-

dominant forms of gingival fibromatosis and are usually not associated with any syndrome. Recently, a mutation in the son of sevenless-1 (SOS-1) gene has been suggested as a possible cause of isolated (nonsyndromic) gingival fibromatosis, but no definite linkage has been established.[6]

Hereditary gingival fibromatosis (HGF) is a benign, nonhemorrhagic, slowly progressive hyperplasia of the maxillary and mandibular gingiva, affecting marginal, attached gingiva and interdental papilla. Usually occurs with the eruption of the permanent teeth, but more rarely with the primary dentition or at birth.[7] It covers the part of the teeth or the entire crown and can be localized or generalized, with a variable degree of severity. This results in diastemas, teeth displacement, or retention of primary or impacted teeth, and may also cause difficulty in speech, lip closure and chewing but above all, at the ages at which it appears, it can affect the patient's esthetics and psychology.[8] It is known to occur in both genders with equal distribution and affects 1 in750,000 individuals. The prevalence of the disease is unknown, but the number of cases can occur within the same family. [9,10,11] 20% of these cases appear with no family history of the same condition.[12]

The hyperplastic gingival tissue is normal in color, firm and fibrotic in consistency and nonhemorrhagic. [10,13] Due to the excess of gingival tissue, the presence of pseudopockets and plaque accumulation, Periodontal problems, such as bleeding and bone loss, might occur. [14,15,16]

Histologically the HGF is characterized by, the presence of thick acanthotic overlying epithelium which has elongated rete ridges. The bulbous connective tissue is relatively avascular and has numerous fibroblasts, densely arranged collagen fibre bundles and mild chronic inflammatory cells. [9,17]

HGF does not resolve naturally. The only viable treatment is through surgical excision of the excess fibrous tissue. The rate of recurrence can be reduced by performing the surgery after the complete eruption of permanent teeth.[9]

In this article, we report a case of a 14-year old girl diagnosed with non-syndromic hereditary gingival fibromatosis along with surgical management and to emphasize the importance of early management of gingival enlargement as if it is left untreated may lead to pathologic, functional and esthetic problems.

Case Report

A 14-year-old female patient was referred from the Department of Pedodontics to the Department of Periodontics, Rajarajeswari Dental College and Hospital, Bangalore, for the management of the gingival enlargement. The patient complained of swollen gums, difficulty in speech and mastication and was concerned of her aesthetic appearance. The patient gives a history of enlarged gingiva since the time of eruption of permanent teeth and it was not associated with pain. There was no relevant familial history of a similar condition. There was a history of consanguinous marriage in all three generations. The patient was systemically healthy and did not have any history of drug intake. Her overall general health was noncontributory. Her mother gave no history of any drug intake during her gestational period and had normal parturition without any complications.

Detailed family history was taken and pedigree chart (figure 1) was prepared including the four latest generations. The history of this family revealed the consanguinous marriages in all four generations. But none of the family members had a history of gingival enlargement.

On extra oral examination, the patient exhibited a convex profile with incompetent lips. Intraoral examination revealed generalized gingival enlargement involving both

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maxilla and mandible and both buccal and lingual/palatal sides, covering ¾ th of the crown anteriorly and the entire crown posteriorly. The gingiva was hyperpigmented, firm and fibrotic in consistency, with increased stippling (figure 2 and 3). There were generalised pseudo pockets with no bleeding on probing and mobility of teeth. Hard tissue examination reveals a high arched palate, generalised fluorosis of the teeth, generalised spacing between the teeth.

The panoramic radiographic examination revealed a normal height of bone and routine blood examination showed values within the normal range. Based on the above findings the provisional diagnosis of idiopathic gingival enlargement was given.

Histopathological examination revealed an abundance of collagen, hyperplasia of overlying epithelium with elongated rete ridges. Relatively avascular connective tissue along with scanty inflammatory cell infiltrate was found. The histopathologic features were suggestive of gingival fibromatosis.

The treatment planned to remove hyperplastic tissue was internal bevel gingivectomy with a full thickness flap in the maxillary arch, due to large areas of gingival enlargement and external bevel gingivectomy in the mandibular arch. The treatment plan was explained to the parent and informed consent was obtained. The surgical intervention was carried out under the infiltration of local anaesthesia, using a surgical scalpel. Excision was carried quadrant by quadrant at an interval of 10days. In the maxillary arch, after anaesthetizing the area, with a # 15 blade, initial scalloped internal bevel incision is made 3mm coronal to the mucogingival junction, including the creation of new interdental papillae and a full-thickness flap is elevated (figure 4 and 5). The base of the papilla connecting the facial and lingual incision is incised. The curettes were used to remove the excised marginal and interdental tissues. Tissue tabs are removed, the root surface is thoroughly scaled and planed. The flap is replaced at tooth bone junction and sutured with interrupted technique (figure 6) and the area is covered with the periodontal dressing. External bevel gingivectomy was carried out in both quadrants of the mandibular arch (figure 7). The patient was prescribed antibiotics (Moxikind 250mg, thrice daily) for 5 days and analgesic (Ibugesic plus 200mg, twice daily) for 3 days postoperatively. A 0.2% chlorhexidine gluconate rinse was prescribed twice a day for 2 weeks and oral hygiene instructions were given.

Sutures and packs were removed after 10 days. After completion of the gingivectomy procedure in all the quadrant, laser bleaching with help of opalescent bleach was carried out in 3 sittings to treat discolouration of teeth due to fluorosis. During the procedure, the tooth was isolated with the help of cotton rolls, margins of the gingiva were protected using the gingival barrier. Using single tufted brush Opalescent® (Potassium nitrate and fluoride) was applied to each tooth. On wearing the protective eyewear (both patient and operator) each tooth was irradiated with an 810-nm diode laser in a contact mode for 5 min with the power setting of 3w (figure 8). The procedure was repeated 3 times in each visit for 3 visits in an interval of 1 week between each visit.

Postoperatively, the healing was satisfactory. The function and esthetic appearance of the patient was improved. Significant reduction in enlargement, adequate exposure of tooth crown along with the whitening of the tooth was achieved. The patient was put on a recall visit at 1, 3, 6 months and 1-year interval. Currently, the patient is under follow up. At 3 months follow up, oral examination showed good oral hygiene and normal probing depth with no significant recurrence of the condition (figure 9).

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Discussion

In the present case report, a 14-year-old female was diagnosed with HGF based on the correlation between the clinical features, familial history, and histopathological findings. The genetic mode of transmission in HGF can be either autosomal dominant or autosomal recessive. [18,19,20]

In autosomal dominant form, the children may suffer from mental retardation and epilepsy. Most autosomal recessive forms are without defects, other than gingival enlargement. Consanguinity has been observed in the recessive form.[21] The mode of genetic transmission in this patient was autosomal recessive form and all three generations had a history of consanguinous marriage.

HGF can occur as an isolated nonsyndromic condition or be associated with a syndrome. Zimmerman–Laband syndrome (defects of bone, ear, nail, and nose, accompanied by hepatosplenomegaly), Cowden syndrome (multiple hamartomas), Murray–Puretic–Drescher syndrome (multiple dental hyaline tumors), Rutherfurd syndrome (corneal dystrophy) and Cross syndrome (hypopigmentation with athetosis) have been occasionally associated with HGF.[22] A thorough evaluation of the patient, in this case, revealed good general health and mental status, no association with any of the clinical features associated with the above syndromes.

The differential diagnosis of gingival enlargement includes drug-induced gingival enlargement and gingival enlargement associated with hormonal changes (pregnancy).[23] The present case did not have a positive drug history or hormonal changes.

Gingival enlargement can be localised or generalised. Based on the extent of tissue involved, HGF has two forms, nodular (localised, mostly seen in maxillary tuberosity and mandibular molar area) and symmetric forms (generalized, which is more common and both areas are equally affected) [24]. In this case, the enlargement was generalised involving both maxillary and mandibular arch, covering ¾th of the crown in the anterior region and the entire crown in the posterior region. The gingival enlargement usually begins at the time of eruption of permanent teeth, which is consistent with the present case [7].

The affected gingiva in HGF is firm, fibrotic in consistency with exaggerated stippling and pink in colour.[24] The same was present in this case, but the colour of the gingiva was black due to melanin hyperpigmentation. This also helped to differentiate the condition from that of the drug-induced gingival enlargement and inflammatory gingival enlargement where the tissue is often movable and red in colour.

The gingival enlargement had influenced the position of the teeth, leading to generalised interdental spaces.[9] The patient had difficulty in speech and mastication. The patient was also concerned about aesthetics.

The histopathological features, in this case, had the typical histopathological features of HGF [25], which consists of acanthotic and hyperkeratotic epithelium with elongated rete ridges, avascular connective tissue with numerous collagen bundles.

In order to address the aesthetic and functional impairment, surgical intervention was necessary. It consists of excision of the enlarged tissue to restore the gingival contours. The most widely used method of removing excess gingival tissue is the conventional external bevel gingivectomy with gingivoplasty, particularly when there are pseudopockets with no attachment loss. Internal bevel gingivectomy along with periodontal flap technique may be preferred for the treatment of gingival enlargement if there are large areas of gingival overgrowth or attachment loss and osseous defects.

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In the present case, gingivectomy was performed in the mandibular arch and in the maxillary arch gingivectomy plus periodontal flap procedure was done. The surgery was done quadrant by quadrant at an interval of 10 days. Also, the patient underwent laser bleaching with opalescent bleach to lighten the fluorosis stains.

Recurrence is a common feature over the varying period. The chance of recurrence is less if the treatment is delayed until the complete eruption of permanent teeth. The post-surgical healing, in this case, is uneventful. The patient's speech, masticatory function and esthetic was improved. No recurrence of the condition was noticed at 3 months follow up. Further evaluation should be carried out in future follow up period.

Conclusion

Early diagnosis and management of the rare condition of hereditary gingival fibromatosis is necessary in order to prevent, the influence of the gingival enlargement in causing pathological migration, malocclusion, functional and esthetic complications. Also, the parents should be educated about the possible condition due to the family history of consanguinity as well as genetic inheritance. The present case of HGF is successfully managed with surgical interventions with no significant recurrence.

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Legends Figure

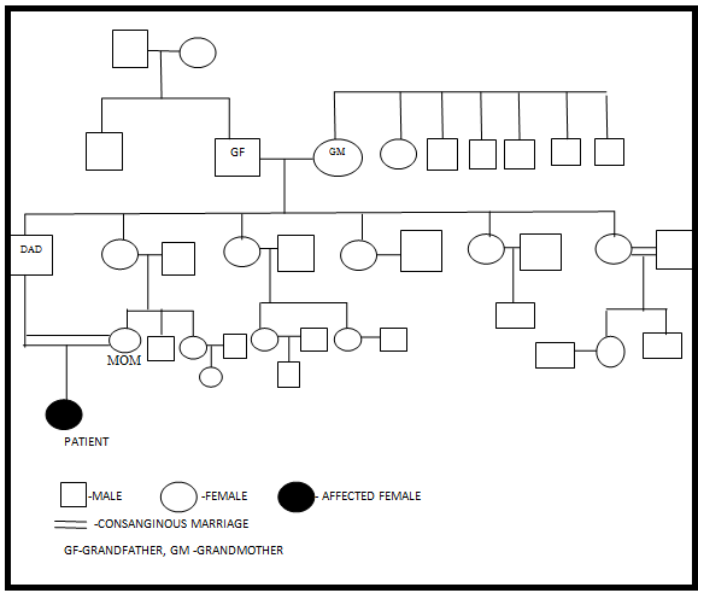

Figure 1: Pedigree of Proposita

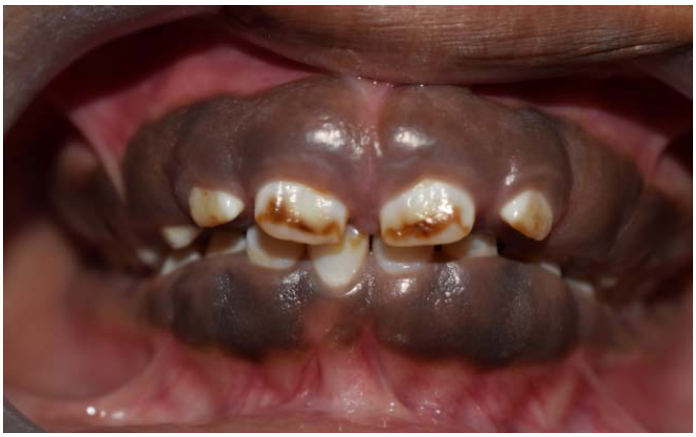

Fig 2: pre-operative photograph showing gingival hyperplasia (frontal view)

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Fig 3: **A** and**B**, lateral view showing the gingival enlargement involving almost the entire crown

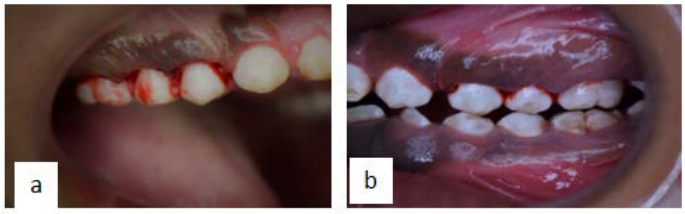

Fig 4: A and B, internal bevel gingivectomy done in both sides of the maxillary arch

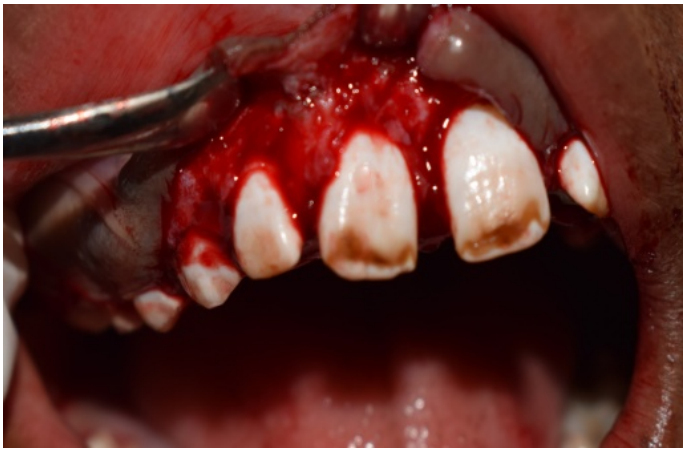

Fig 5: conventional gingivectomy plus periodontal flap technique

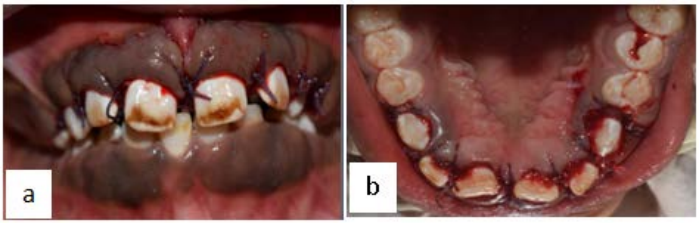

Fig 6: A,labial view, **B**, palatal view ,flaps are approximated with the help of vicryl sutures

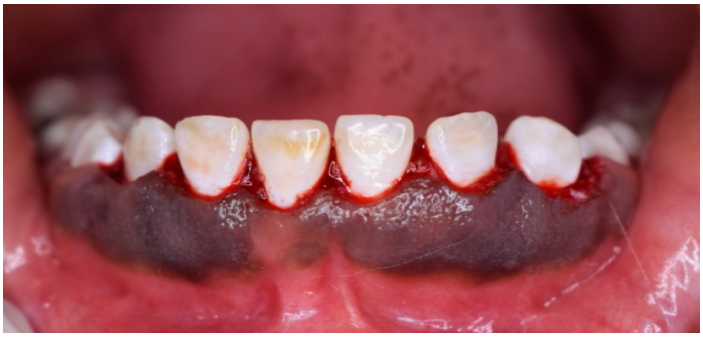

Fig 7: conventional gingivectomy

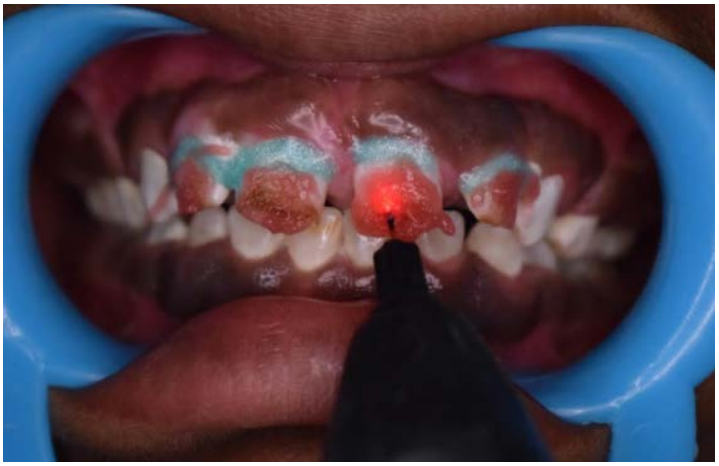

Fig 8: Laser bleaching

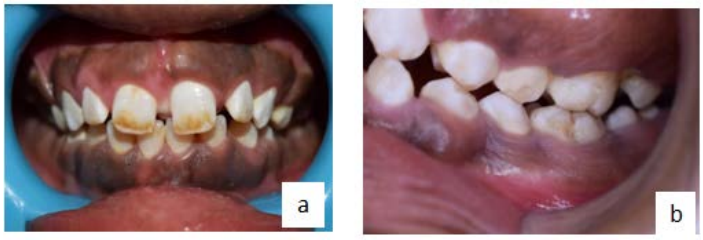

Fig 9: A, frontal view, **B** lateral view, 3 months post-operative follow up

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