CASE REPORT

An Unusual Presentation of Hereditary Gingival Fibromatosis with Maxillary Buccal Alveolar Bone Exostosis: A Case Report

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ABSTRACT

Hereditary gingival fibromatosis is a rare condition. The enlargement characterized by slow and progressive enlargement of attached gingiva as well as the gingival margin and interdental papilla. It can occur as an isolated disorder and also as a part of syndrome. This case report present a case of 18 year old male who presented with generalized gingival overgrowth with maxillary buccal alveolar bone exostosis. This non-syndromic case was treated by modified gingivectomy technique with osteoplasty. The post-operative result was uneventful, and the patient appearance improved considerably. After completing the treatment regular follow up visits are necessary in order to evaluate oral hygiene and stability of periodontal treatment.

KEYWORDS: Hereditary Gingival Fibromatosis, Modified Gingivectomy Technique, Osteoplasty

INTRODUCTION

Gingival fibromatosis (GF) is an abnormal growth of gingival tissues caused by an increase in submucosal connective tissue elements Many cases reported are iatrogenic; some are inherited while others are idiopathic.[1] The etiology and pathogenesis of gingival fibromatosis are not well established; however it can occur due to factors like plaque accumulation, caries, hormonal stimulation, drugs, blood dyscrasias, or idiopathic.

Hereditary gingival fibromatosis is a rare type of gingival enlargement which may be generalized or localized. In this type the enlarged gingiva have a normal color which is firm in consistency with stippled surface texture. Buccal and lingual tissues in both the maxilla and the mandible can be involved. There can be mild to severe degree of enlargement. It usually begins with eruption of the primary or permanent dentition and rarely present at birth. The gingival overgrowth leads to effects like diastemas, malpositioning of teeth and long term retention of primary teeth. More severe lesions causes both esthetic and functional problems because of coverage of crowns of teeth.[2] The enlarged tissue to may get traumatized during mastication. Sever growth usually leads to an abnormal swallowing pattern, speech impediments and difficulty in mastication. All these factors will favour the accumulation of plaque and material alba and cause interference with maintenance of oral hygiene, which further complicates the existing hyperplastic tissue.[3] Gingival fibromatosis may present in some genetic disorders like Hurler syndrome, Sturge Weber syndrome, Wilson syndrome Zimmerman – Laband syndrome, Maroteaux-Lamy syndrome, KlippelTrenaunay syndrome, Goltz syndrome, Ramon syndrome, Scheie syndrome, Regional Systemic hyalinosis Hurler/ scheie, Odontodysplasia, Jones syndrome Hunter syndrome, Rutherfurd syndrome, Sly syndrome, Cross Syndrome, I- Cell disease, Neurofibromatosis type I, Anderson – Fabry disease, Alpha Mannosidosis, Schinzel – Giedion syndrome, Ligneous periodontitis, Niemann – Pick disease, Menkes Kinky hair disease, Cowden syndrome and many more.[4]

An exostosis is a localized, peripheral overgrowth of the bone which is benign in nature. The etiology of exostosis is unknown. It may be present as a nodular, flat or pedunculated protuberance located on the alveolar surface of maxillary and mandibular bones. In the jaws, depending on the anatomic location they are named as torus palatinus, torus mandibularis (lingual premolar area), or buccal bone exostoses (BBE). BBE are found less commonly than tori. BBE occurs along the buccal aspect of the maxilla or mandible, usually in the premolar and molar areas. These exostoses are usually found at the time of periodontal diagnosis and treatment. Buccal exostoses may be traumatized and interfere with oral hygiene procedures. Also, Buccal exostoses are significant with regards to prosthodontics because they may interfere with denture insertion.[5]

Here we report a case of an 18-year-old male patient with non syndromic hereditary gingival fibromatosis (HGF) with maxillary buccal alveolar bone exostosis with its management.

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CASE REPORT

An 18-year-old- male reported to the Department of Periodontics, Govt. Dental College and hospital, Patiala, with complaint of swollen gums, speech and masticatory difficulty and poor esthetics. According to the patient gingival enlargement had started at the time of eruption of permanent dentition with slow increase in size; which leads to delayed eruption of permanent dentition. He did not give any history of drugs intake, fever, anorexia, weight loss, seizures, hearing loss, nor having any physical or mental disorder. His family history was of significance since, according to the patient, his father, and his nephew had the gingival enlargement but both of these cases did not report to the hospital for treatment.

Examination

Extraoral examination revealed a convex profile.

An intraoral examination revealed generalized enlargement of gingiva with involvement of both the maxillary and mandibular arches. The gingiva was pink, with a firm and dense consistency and covered more than half of the crown surfaces. The enlargement had influenced the position of his teeth, in such a way that the dentition appeared with diastema and malpositioning [Figure 1]. Transgingival probing revealed the bony overgrowth on the buccal aspect of the maxillary posterior region.

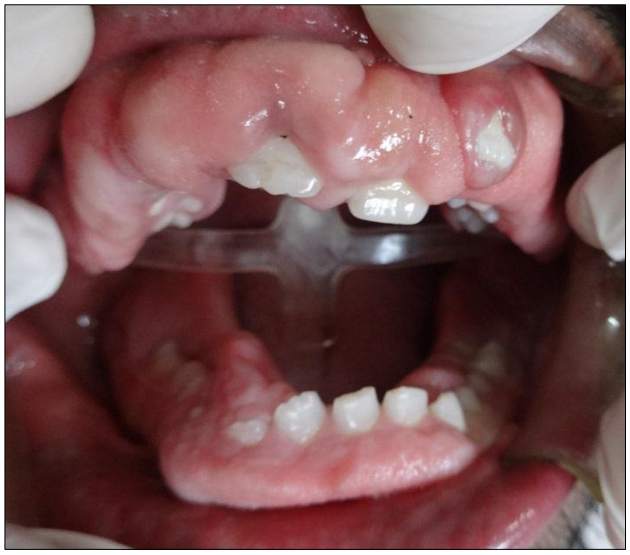

Figure 1: Preoperative photograph showing generalized gingival enlargement

Investigations

A panoramic radiograph was taken, which revealed normal bone height and retained deciduous canines in maxillary right and left region [Figure 2]. Routine blood investigations were done which showed normal results.

Treatment

After explaining the patient the potential risks and benefits, the informed consent was obtained.

Keeping in mind the desire of the patient for better esthetics and his unpleasant situation, surgical treatment

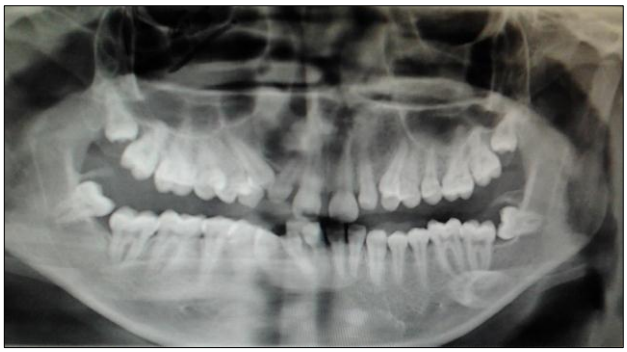

Figure 2: Panormic radiograph showing retained deciduous canines in maxilla

for complete excision was planned. Modified gingivectomy technique followed by osteoplasty of buccal alveolar exostosis was done under local anaesthesia quadrant wise on maxillary arch and retained deciduous canines were extracted at the same time which were present buccally to the permanent canines and modified gingivectomy technique was performed on mandibular arch. The biopsy samples of the gingival tissue was submitted for histologic evaluation. The gingival histopathology revealed moderately dense collagenous connective tissue with collagen bundles arranged in a haphazard manner. Connective tissue was relatively avascular along with few inflammatory cell infiltrate. The overlying epithelium was hyperplastic with elongated rete ridges [Figure 3], and bone histopathology shows normally looking bony trabeculae and calcification. The histopathologic features were suggestive of hereditary gingival fibromatosis with maxillary buccal alveolar bone exostosis.

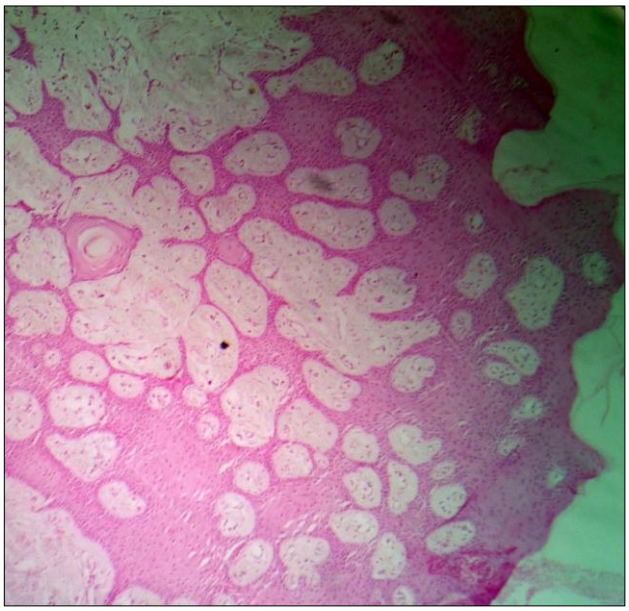

Figure 3: Histologic Section showing thick parakeratinized stratified squamous epithelium with dense collagenous connective tissue (H & E, 10x)

Post-surgical healing was uneventful. The patient was recalled at 1, 3, 6 and 12 months post-surgery. During this period, no recurrence was seen [Figure 4]. At the

time of recall visit after 3 months of surgery, the patient was suggested to undergo orthodontic treatment for the correction of diastema and malpositioning of teeth. However, the patient did not acted to it.

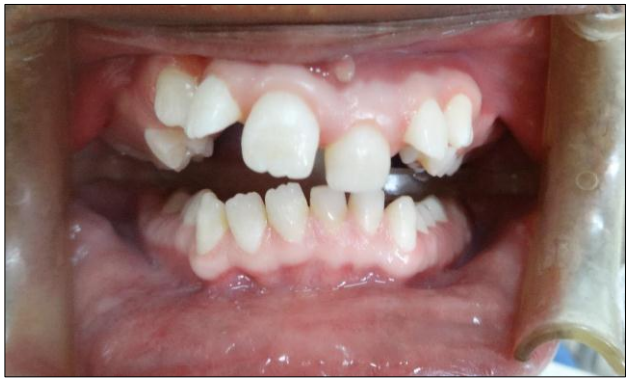

Figure 4: Post-Operative photograph showing 1 year of follow up showing no recurrence of gingival enlargement

DISCUSSION

This article reports a case of hereditary gingival fibromatosis with maxillary buccal alveolar bone exostosis and its management. The genetic mechanism of this condition is not fully understood, but many of research workers have related it to hereditary factors.[6] In our case, the patient did not had any signs or symptoms which can be related to any syndrome. The diagnosis was made on the basis of clinical features, family history, and characteristic microscopic features of the histologic samples. This type of fibromatosis can have inheritance as an autosomal dominant or autosomal recessive condition. In the present case, the gingival enlargement was a hereditary condition, probably autosomal dominant, due to its existence in father and nephew.

There are inconsistent aspects in the literature about the cellular and molecular mechanisms of the gingival fibromatosis. Some authors report that the mechanism involved is an increase in the proliferation of gingival fibroblasts,[7] whereas others report slower-than normal growth.[8] there can be chances that increased collagen synthesis rather than decreased levels of collagenase activity may be involved.[7]

The treatment of gingival fibromatosis is very important. The method for the treatment varies according to the degree of severity, when the enlargement is minimum, good scaling of the teeth and home care may be enough to maintain good oral health. As the size of gingival tissue increases which compromise function and esthetic of the patient, the need for surgical intervention usually become must.[2]

Among the suggested treatment protocols, modified gingivectomy technique, primarily a modification of the ledge and wedge technique was favored.[9] Unlike the ledge and wedge technique which consists of a primary gingivectomy followed by two incisions, our surgical technique comprised of a primary gingivectomy followed by a single internal bevel incision. This technique allows the reflection of the conventional flap to permit access to the underlying bone for osseous reconturing.

Recurrence following surgical treatment cannot be predicted. It is most frequently seen in children and teenagers rather than adults. Good oral hygienic environment is maintained by a combination of regular examinations with professional cleaning and oral hygiene instructions that delay the chances of recurrence. Surgical intervention has benefits that very well improve patient's quality of life since the removal of enlarged gingival tissue improves access for plaque control, eliminates difficulties in mastication and speaking and leads to psychological benefits due to improvement in esthetics.[10] In our case, even after 1 year of follow-up, no recurrence of gingival overgrowth was observed.

CONCLUSION

Hereditary gingival fibromatosis with buccal alveolar bone exostosis is a rare disorder. Unesthetic appearance and impaired function often demand surgical intervention, although recurrence cannot be predicted. The good esthetic result was achieved without recurrence. After completion of the treatment, regular recall visits are necessary in order to evaluate oral hygiene/ and the stability of the periodontal treatment done.

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