ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 4, Issue – 6, December - 2021, Page No. : 422 - 425

Diagnosis and management of nonsyndromic hereditary gingival fibromatosis in a 9 year old girl: Report of a rare case

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Citation of this Article: Dr. Karan Thakur, Dr. Munnasha Rastogi, Dr. Amit K Sharma, "Diagnosis and management of nonsyndromic hereditary gingival fibromatosis in a 9 year old girl: Report of a rare case", IJDSIR- December - 2021, Vol. – 4, Issue - 6, P. No. 422 – 425.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Overgrowth of keratinized gingival tissues is a common condition and is described under variety of names. Causes of such enlargement can be medications, hereditary and/or local irritating factors. Hereditary gingival fibromatosis is a rare condition characterized by various degree of gingival overgrowth. It usually develops as an isolated disorder but can manifest with multisystem syndrome. We are here presenting a case of a 9 year old girl who presented with severe enlargement of gingiva covering almost the entire crown involving both maxillary and mandibular arches. Differential diagnosis includes drug induced and idiopathic gingival enlargement. Excess gingival tissue was removed by full mouth gingivectomy and sent for histopatholoical examination. Postoperative course was uneventful and there was significant improvement in patient's esthetics. A 12 month postoperative period shows no recurrence.

Keywords: Hereditary gingival fibromatosis, Gingivectomy, Idiopathic gingival enlargement.

Introduction

Overgrowth of keratinized gingival tissues is a common condition and is described under variety of names. Causes of such enlargement can be medications, hereditary or local irritating factors.

Hereditary gingival fibromatosis (HGF), also known as idiopathic gingival enlargement, Hereditary gingival hyperplasia, idiopathic fibromatosis and hypertrophied gingiva is a rare (1 in 75000) hereditary condition characterized by slow, progressive enlargement of the gingiva. Males and females are equally affected.[1]

Hereditary gingival enlargement (HGF) occurs isolated or as a part of multisystem syndrome. It may present itself as an autosomal dominant which has been linked to chromosome 2p21-p22 and 5q13-q22 or recessive mode of inheritance. Although involvement of SOS-1 gene has been suggested recently. Families are affected across generations and a positive family history is always present in HGF. Hereditary gingival fibromatosis is gradually progressive benign enlargement that affects the marginal, attached and interdental gingiva. Histopathologically, it implies an increase in both extracellular matrix and cell numbers.[2]

HGF can present in two forms, a nodular form affecting the dental papillae or a symmetric uniform gingival enlargement. HGF is seen as pale pink enlargements, firm and leathery in consistency.[3] Here we report a case of non syndromic hereditary gingival fibromatosis in a 9 year old girl which was treated using quadrant wise conventional surgical technique.

Case report

A 9 year old female patient accompanied by her mother reported to the Department Of Pedodontics And Preventive Dentistry, Himachal Dental College Sunder Nagar with chief complaint of generalized gingival swelling initiated slowly and has assumed the present size within 3 years. The family history of affected persons was determined by questioning of the index case and it was confirmed that there were other family members who also had symptoms of gingival overgrowth and that was subsequently confirmed by clinical examination. No further relevant medical history was present.

On intra oral examination there was presence of generalized firm swelling of gingiva which covered the cervical and the middle third of the clinical crowns of all the teeth. A complete hemogram was conducted with all the parameters within the normal range. An incisional biopsy was taken for histopathological exam 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. On the basis of histopathological report a confirmed diagnosis of gingival hyperplasia was made.

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

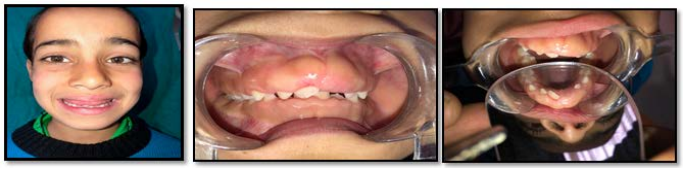

Fig 1: Preoperative Extra oral and Intraoral Photographs.

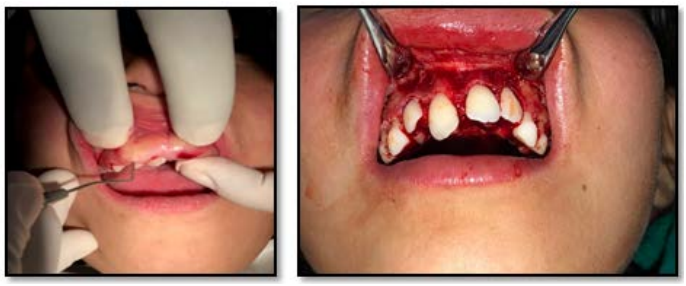

Fig 2: Photograph during procedure.

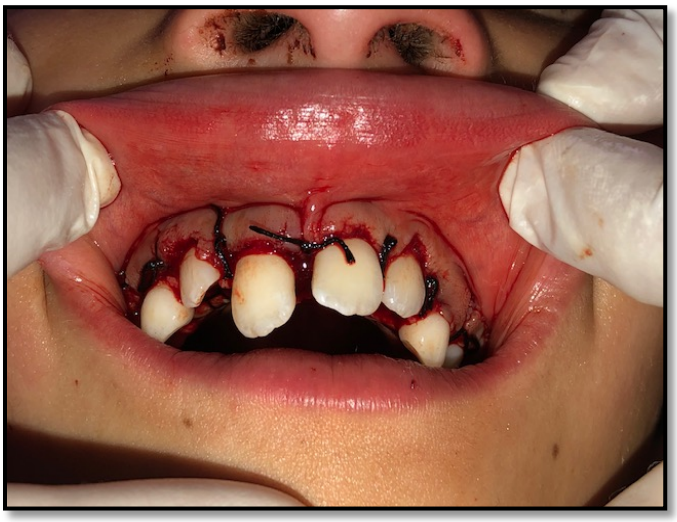

Fig 3: Postoperative picture.

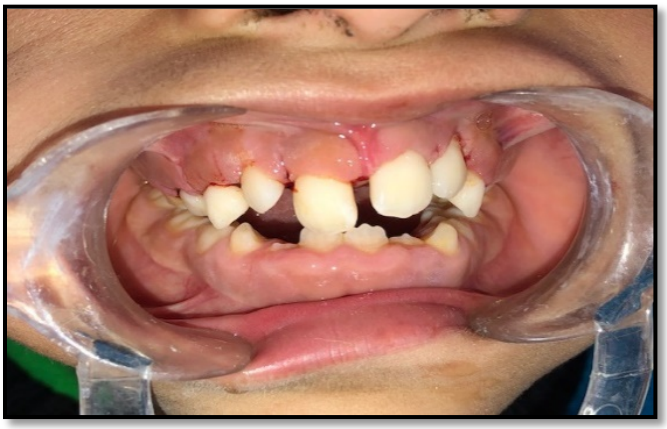

Fig 4: Follow up after two weeks

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. 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 within a syndrome. The etiological factors contributing for gingival fibromatosis reported were administration of certain drugs (phenytoin, cyclosporine, Nifedipine), inflammation, systemic conditions (leukemia), hormonal conditions (Pregnancy, puberty or hyperthyroidism) and nutritional conditions like vitamin C deficiency.[4] Genetic studies demonstrated that HGF could be caused by mutation of the SOS1 gene, however, researchers on three Chinese families and two Polish families with HGF did not detect any mutation in SOS1. Notably, the latest research revealed RE1- silencing transcription factor (REST) final axon truncating mutations caused HGF in three unrelated families. Thus, HGF may involve several genes and more studies are needed to probe the HGF related mutations.[5]

The histological findings are nonspecific with hyperkeratosis, acanthotic epithelium with elongated papillae. The abundance of fibroblasts is present in connective tissue. This increased production of extracellular matrix molecules, type I collagen and fibronectin could lead to bulk of gingiva clinically. Gingival enlargement can cause speech alteration, diastema in teeth, cessation of eruption of permanent teeth leading to difficulty in mastication, malocclusion and unaesthetic appearance. Management of gingival enlargement depends on the age of occurrence and cause of this condition. Gingival overgrowth usually begins with the eruption of permanent teeth which progresses rapidly with the stage of active eruption. In order to address patient's functional and aesthetic needs, surgical excision of the enlarged gingiva is usually necessary. Other methods of removing large quantities of gingival tissue have been used in number of studies, such as carbon dioxide laser and electrocautery. In the present case, Gingivectomy using scalpel method with wedge procedure was adopted. Conventional surgical excision has advantages over other modes as bulk of tissue can be removed in single visit, technique is not sensitive and faster healing rates have been observed. There is no definite consensus regarding the time of surgical intervention in young and adolescent patients. Emerson suggested to wait till all permanent teeth have erupted i.e.

at ages 3, 6 and 12 in order to maintain oral hygiene effectively after gingivectomy.[6]

Recurrence rate in HGF is very high after surgery, so that the patient should be followed for considerable period of time and may require repeated surgery. The appropriate time of removal of recurrent gingival enlargements varies. Emerson recommended that the best time is when all the teeth have erupted.[7]

Conclusion

HGF is a rare entity, conferring to pedodontist and periodontist which have important role in diagnosis and treatment of affected patients. Aesthetic and dental associated alterations can affect the quality of life of affected children. Hence, quadrant wise gingivectomy using scalpel method was used along with proper oral hygiene instructions in this case. Follow up of 12 months revealed satisfying results with no recurrence.

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