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RESEARCH ARTICLE

IDIOPATHIC GINGIVAL FIBROMATOSIS - A CASE SERIES

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Abstract

Unusual Cases with the complaint of swollen gums in the region of left upper and lower back teeth since 2years of are presented. They had moderate to severe, unilateral diffuse gingival enlargement in the maxilla and mandible. The diagnosis was made based on clinical and histopathological examination after ruling out drug and family history. Surgical removal of the hyperplastic tissue was done.

Introduction:-

The gingival tissues in the healthy mouth almost completely fill the interproximal spaces between teeth, beginning near the contact area and extending apically and laterally in a smooth curve. However, there is frequently an increase in the size of the gingiva so that soft tissue overfills the interproximal spaces, balloons out over the teeth and protrudes into the oral cavity. Gingival fibromatosis (GF) is a slowly progressive, benign, non-bleeding, painless, localized or generalized overgrowth of the maxillary and mandibular keratinized gingiva.[1] It can lead to diastema, malocclusion, delayed eruption of permanent dentition or prolonged retention of primary dentition, causing aesthetic and functional problems. Hereditary GF (HGF) is a rare disorder; which has been also termed: Elephantiasis of gingiva, Gigantism of gingiva, Fibromatosis gingivae, multiple epulides, Hypertrophic gingivitis.[2] It has prevalence of about one in 1,75,000 individuals transmitted either as an autosomal dominant or rarely, an autosomal recessive trait.[3] 2p21 -p22 is one of the two loci demonstrated to be responsible for it.[4]

In many instances there are family histories of the disease being associated with a hereditary developmental anomaly, apparently transmitted by a dominant gene, other cases deviate from this pattern and in some instances this pattern cannot be established. It is not always possible to identify the exact etiologic factors for gingival enlargement and classify them into a particular type, such cases are considered to be Idiopathic. It appears as an isolated disorder or may sometimes be associated with other conditions like epilepsy, hypertrichosis and mental retardation. [5,6] It may also develop as a part of syndromes like Cowden's syndrome [7], Zimmerman-Laband syndrome [8] and Murray-Puretic Drescher syndrome. [7] The onset usually coincides with the eruption of the permanent dentition though some cases have even been reported in the deciduous dentition. Different clinical variations are seen depending on the genetic heterogeneity.[9]

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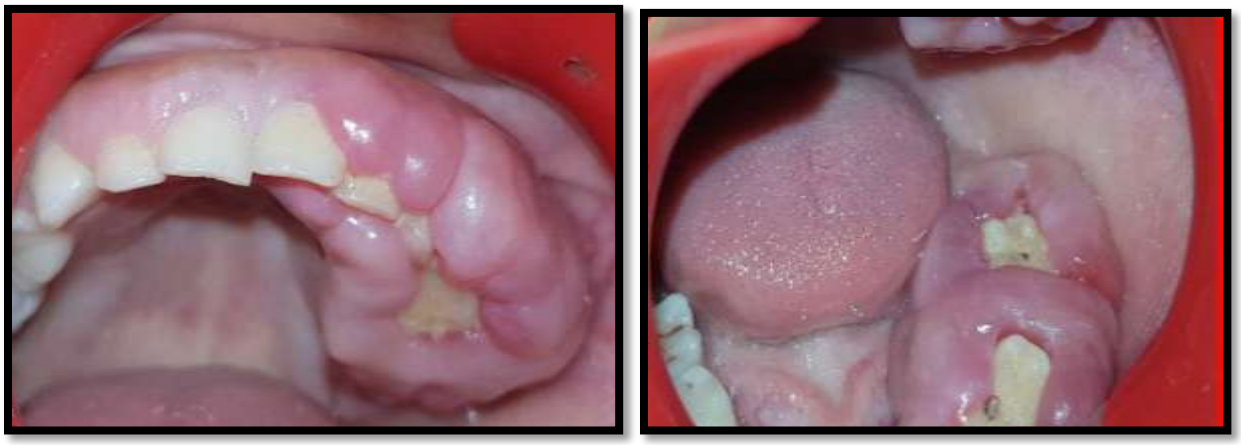
Oral manifestations may vary from minimal involvement of only the tuberosity area and the buccal gingiva around the lower molars to generalized enlargement inhibiting eruption of teeth. The hyper plastic gingiva usually is pale-pink, non-hemorrhagic with a firm and fibrotic consistency and presents a characteristic 'pebbled' surface. The condition has been classified into two types, Nodular form characterized by presence of multiple tumors in the dental papillae and other form which is symmetric resulting in uniform enlargement of gingiva and represents the most common type. There may be combination of both the types. [10] The hyperplastic gingival tissues appear normal at birth but begin to enlarge with eruption of primary teeth. Histologically IGF is described as a moderate hyperplasia of the epithelium with hyperkeratosis and elongation of the rete pegs. The increase in the tissue mass is primarily the result of an increase in thickening of the collagen bundles in the connective tissue stroma.[11]

Case 1:
A 3 year old girl accompanied by her parents reported to Department of Pediatric and Preventive Dentistry, NPDCH, Visnagar with the complaint of swollen gums in the region of left upper and lower back teeth since 2 years. Difficulty in chewing and pain was also reported. Family and post-natal history was non-contributory and patient did not have any history of epilepsy or any type of physical or mental disorder. She was undergoing treatment for anemia with iron supplement. Developmental milestones and other systems of the child were normal. The patient has unilateral mastication habit with right side since childhood. The right hand used for brushing.

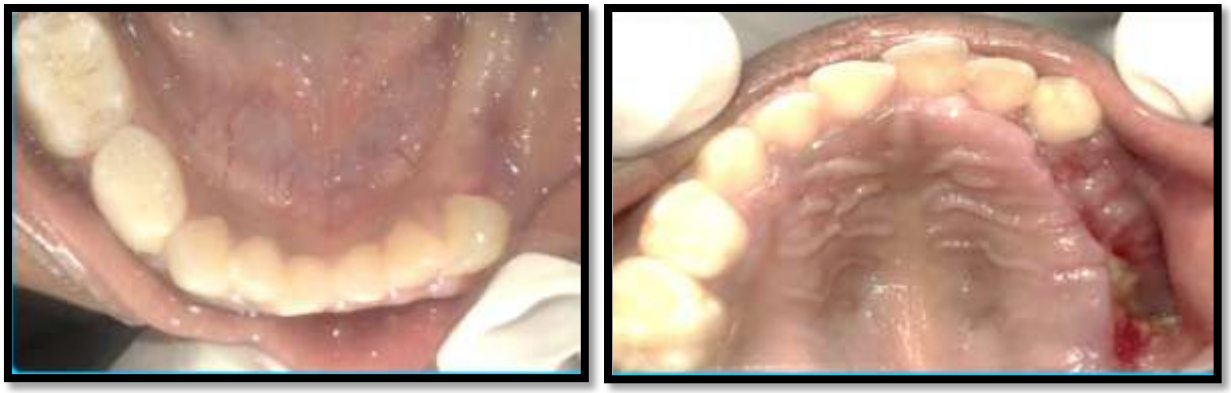
On extraoral examination child had incompetent evened lips and convex profile and the enlarged gingiva could be palpated over left cheek. The overlying skin appeared to be normal. Left submandibular lymphadenopathy was noticed.

Intraoral examination revealed unilateral diffuse, nodular enlargement of gingiva extending from Primary canine to primary second molars in both arches on left side. The teeth were almost completely covered and displaced. The colour of the gingiva appeared to be normal. Calculus deposits were seen on exposed tooth surfaces. Gingiva was pale-pink, firm and fibrous consistency with pebbled surface. Panoramic radiograph revealed crestal bone loss with displacement of teeth and no abnormality in number, size and structure of the teeth and all the deciduous teeth had erupted from the alveolar bone. Patient was subjected to a thorough medical examination. Patient was found to be anaemic (Hb 8 gm %) and hormonal investigations were normal.

Histopathological investigations after excisional biopsy revealed hyper-para keratinised hyperplastic stratified squamous epithelium with dense fibro collagenous tissue with chronic inflammatory cell infiltration. On the basis of the medical, family and drug histories and clinical findings it was diagnosed as Idiopathic Gingival Fibromatosis. Considering the age and behaviour of the patient unilateral gingivectomy procedure was done under Midazolam nasal spray. During the course of surgery lower left first and second primary molar was noticed to have grade III mobility. The patient was recalled after 15 days for check-up. There are various other procedures for removal of such tissues like electrocautery and carbon dioxide laser. The final outcome of the surgery should restore functional/masticatory and esthetic needs of the patient.



Pre-operative photograph



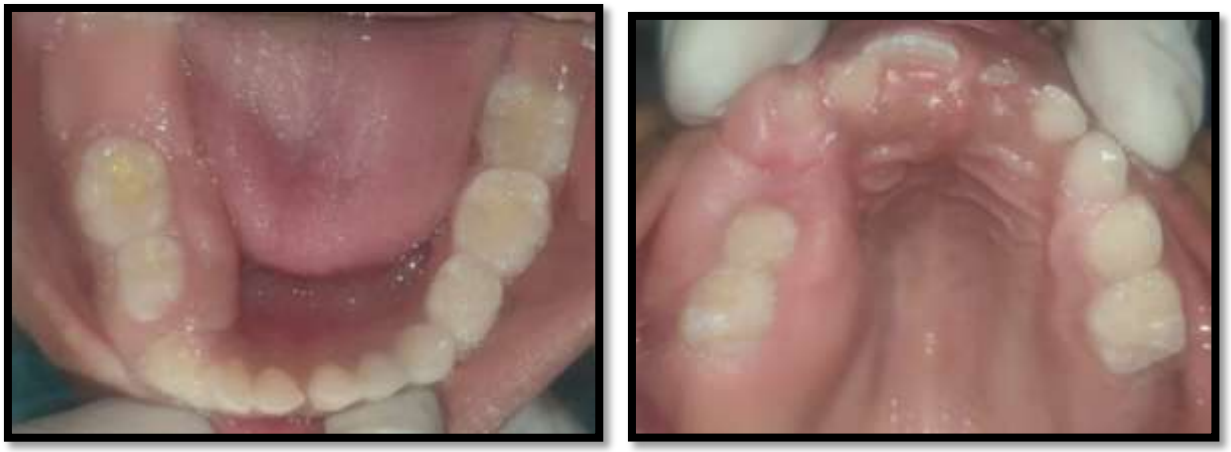
Post-operative photograph **15days follow up**

Case 2:
A 6 year old girl accompanied by her parents reported to our department with the complaint of swelling of gums in the region of right upper and lower right back teeth since 2-3 years.

The patient was slightly under built for her age. No relevant medical history of intake of drugs, nutritional deficiency or family history indicating any underlying genetic mechanism.

Examination of oral cavity revealed diffuse type of gingival enlargement involving marginal, papillary and attached gingiva on right side involving both maxilla and mandible. The enlargement extended from canine to second deciduous molar in both upper and lower jaws. Gingiva was pale-pink, firm and fibrous consistency with pebbled surface. Panaromic radiograph revealed no bone loss with displacement of teeth and no abnormality in number, size and structure of the teeth and all the deciduous teeth had erupted from the alveolar bone. Routine haematological investigations, thyroid function tests, serum calcium and phosphorus and other routine urine examinations were found to be within normal limits.

Histopathological investigations after excisional biopsy revealed hyper-parakeratinised hyperplastic stratified squamous epithelium with dense fibro collagenous tissue with chronic inflammatory cell infiltration. On the basis of the medical, family and drug histories and clinical findings it was diagnosed as Idiopathic Gingival Fibromatosis. The treatment is normally based on understanding of cause and underlying pathologic process. Since the exact cause could not established, treatment included surgical excision of enlarged tissue. Patient is appointed for maxillary arch treatment.



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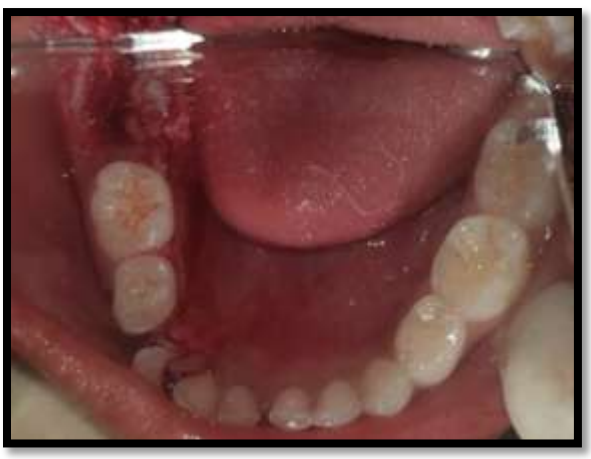
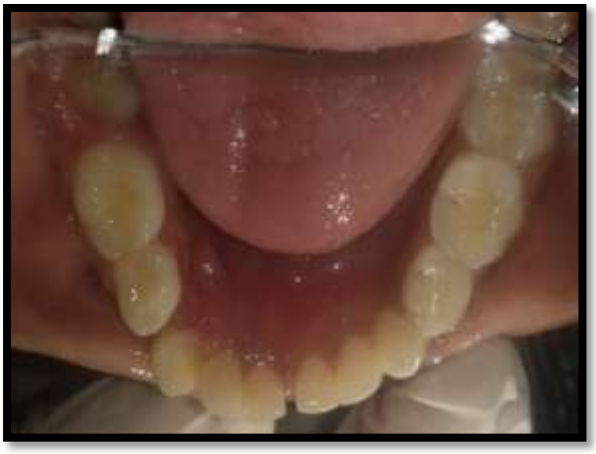


Post-operative photograph



15 days follow up

Discussion:-

As the family, medical, prenatal and drug histories were noncontributory to this case, it was termed as Idiopathic Gingival Fibromatosis (IGF). IGF manifests due to congenital or hereditary causes which is not understood accurately. Some authors have proposed mode of transmission as mainly autosomal dominant, suggesting abnormal chromosome on phenotype 2p21[12,4] Various other factors are responsible for IGF including inflammation, leukemic infiltration and drugs like Phenytoin, [13] Verapamil,[14] Cyclosporine,[15,16] Nifedapin. [17,18] It is associated with many syndromes like combination of IGF, Mental Retardation, Hypetrichosis and Epilepsy[19,20], Ru-therford syndrome (IGF and corneal dystrophy), Laband syndrome" (IGF, ear, nose, nail, bone defects with heptosplenomegaly) and cross syndrome[21](IGF microopthalmia, mental retardation, athetosis. and hypo pigmented skin). Murray-Purelie-Drescher[21] (IGF with multiple hyaline fibromas). Jones syndrome (IGF with sensorineural deafness). This patient had no clinical findings that fulfilled any of these possible syndromes. [21]

Sometimes, gingival enlargement does not occur until the eruption of the primary or permanent dentition.[22,23] It has been also suggested that IGF may be due to nutritional hormonal factors but this is not proven. Due to massive gingival enlargement an affected child usually develops abnormal swallowing pattern and experiences difficulty in speech and mastication.[13]

Histologically, gingival hyperplasia is mainly due to an increase and thickening of collagen bundles in connective tissue stroma.[24] The nodular appearance can be attributed to the thickened para hyperkeratinized epithelium.[25] Various treatment modalities have been proposed but the treatment of choice in this condition was Gingivectomy. Recurrence rate in IGF is very high after surgery and because of this the patient should be followed for considerable period of time and may require repeated surgeries. Appropriate time of the removal of recurrent gingival enlargement varies; Emerson recommended that the best time is when all the permanent teeth have erupted. [26] Appropriate time for the removal of gingival enlargement is at the age of 3, 6 and 12 years to have effective plaque control and to maintain oral hygiene after Gingivectomy procedure. This often causes further increase in the patient and parents' psychological and emotional stress. Hence, psychological counselling is a must for patients and parents.

Conclusion:-

Early diagnosis, emphasis on conservative management and a multidisciplinary treatment protocol are important factors in the management of patients with idiopathic GF. More genetic research is needed to identify their molecular basis for a more definitive line of treatment.

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