IDIOPATHIC GINGIVAL FIBROMATOSIS ASSOCIATED WITH GENERALIZED AGGRESSIVE PERIODONTITIS COMBINED WITH PLASMA CELL GINGIVITIS: A RARE CASE REPORT

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Abstract

Gingival hyperplasia is a rare condition but it is important for cosmetic and mechanic reasons and because of its potential as an indicator of systemic disease. Gingival fibromatosis may exist as an isolated abnormality or as part of a syndrome. Aggressive periodontitis, another genetically transmitted disorder of the periodontium, typically results in severe, rapid destruction of the toothsupporting apparatus. The increased susceptibility of the host population with aggressive periodontitis may be caused by the combined effects of multiple genes and their interaction with various environmental factors. Plasma cell gingivitis (PCG) is reportedly an uncommon oral condition with distinctive clinical and microscopical features. The condition is characterized by diffuse gingival enlargement, erythema and sometimes by desquamation.

We present a rare case of a nonsyndromic idiopathic gingival enlargement associated with generalized aggressive periodontitis. The patient's clinical features, treatment received, the histopathologic presentation and proper management of the condition are discussed.

Keywords: gingival hyperplasia; fibromatosis; Aggressive periodontitis ; Plasma cell

Introduction:

Gingival enlargements are quite common and may be either inflammatory, non inflammatory or a combination of both. Idiopathic gingival hyperplasia is a rare condition of undetermined etiology described variously as fibromatosis gingivae, gingivaematosis, hereditary gingival fibromatosis idiopathic fibromatosis, familial elephantiasis and diffuse fibroma. Diffuse gingival enlargement is also found to be associated with syndromes like Cross syndrome, Rutherford syndrome, Ramen syndrome, Zimmerman Laband syndrome and Juvenile hyaline syndrome[1].As the fibrous hyperplasia is significantly enhanced by poor oral hygiene, periodontitis may be associated with the fibrosis. This entity was first reported in 1856 by Goddard and Gross under the rather descriptive term, "fungus excrescence of the gingiva "Gingival hyperplasia is a rare condition but it is important for cosmetic and mechanic reasons or possibility of a part of a systemic disease. In some pathological conditions, gingivitis caused by plaque accumulation can be more severe. In puberty and pregnancy, hyperplasia of the gingival tissues may be due to poor oral hygiene, inadequate nutrition, or systemic hormonal stimulation. Gingival enlargements are also seen in several blood dyscrasia e.g. leukaemia, thrombocytopenia, or thrombocytopathy Other etyologic factors are listed in table 1. A Progressive fibrous enlargement of the gingiva is a feature of idiopathic fibrous hyperplasia of the gingiva. Characteristically,

this massive enlargement appears to cover the tooth surfaces. While the cause of the disease is unknown,there appears to be a genetic predisposition. Gingival fibromatosis may exist as an isolated abnormality or as part of a syndrome . Table 2 gives an overview of syndrome related gingival overgrowth[2].

Rarely fibrous hyperplasias have large numbers of plasma cells scattered throughout the subepithelial stroma. This plasma cell gingivitis is an uncommon benign condition of gingiva presumed to be an unusual allergic reaction to an antigen,often flavouring agents and certain toothpastes.

Aggressive periodontitis is currently considered to be multifactorial disease that develops as a result of complex interactions between specific host genes and the environment, leading to development of a severe and rapidly progressive form of periodontitis.Generalized aggressive periodontitis usually affects people under 30 years of age. They have a poor serum antibody response to infecting agents and pronounced episodic destruction of the attachment and alveolar bone.The loss of attachment affects at least 3 permanent teeth other than first [3]

Visuals aspect	Cause
Gingivitis	Bacterial plaque
More severe gingivitis diabetes	Bacterial plaque and uncontrolled
Puberty or pregnancy epulides	Bacterial plaque and puberty or pregnancy
Drug-induced gingival overgrowth phenytoin, Diltiazem	Bacterial plaque and medicine
Enlarged, oedematous, soft and tender, easily bleeding gingivitis	Leukaemia
Gingival enlargment and spontaneous bleeding	Thrombocytopenia and thrombocytopaty

Table 1. *Causes of gingival hyperplasia*[2]

Syndrome	Symptoms other than gingival overgrowth	Heredity
Rutherfurd Syndrome	Corneal dystrophy	Dominant
Cross Syndrome	Microphthalmia, mental retardation, pigmentary defects	Recessive
Ramon Syndrome	Hypertrichosis, mental retardation, delayed development epilepsy, cherubism	Recessive
Laband Syndrome	Syndactily, nose and ear abnormalities, hyperplasia of the nails and terminal phalanges	Dominant

Table 2. *An overview of gingival overgrowth related with a syndrome*[2]

Case Report

A 16-year-old female presented with gradual and progressive enlargement of both upper and lower gingival tissues for one and half years, preventing proper speech, articulation, mastication, and causing inadequate lip apposition and poor esthetics. Patient gave a history of surgical excision of similar lesion two years back. There was no history of fever, prolonged medications, anorexia,weight loss, seizures, or hearing loss. Family and postnatal history was non-contributory. The patient did not have any history of epilepsy or any type of physical or mental disorder .However, patient did give a history of use of tooth paste since two and half years prior to which she was using datun.

On Extraoral examination, the patient had incompetent everted lips and a convex profile. An intraoral examination revealed generalized, gross, nodular enlargement of the gingivae involving the upper and lower arches, which were pink in color, and had a firm and fibrous consistency. The teeth were barely visible as they were buried deep within the enlarged gingiva (Fig.1)The patient had generalized tooth mobility with a severe pathologic migration.Hematological investigations were within normal limits

correlated with an absence of any history of systemic disease. Full-mouth periodontal charting, including assessment of probing depth and clinical attachment level, revealed deep pockets throughout the mouth, and scanty plaque and calculus deposits. The radiographic findings, which corroborated those of the clinical examination, revealed severe generalized alveolar bone loss(Fig.2). Based on all these findings, a provisional diagnosis of unusual gingival enlargement with generalized aggressive periodontitis was made. Histopathological examination revealed thickened and acanthotic overlying epithelium with elongated rete ridges. The underlying connective tissue showed dense collagen fibre bundles with numerous fibroblasts. Intense chronic inflammatory cell infiltration predominantly composed of plasma cells was also evident.(Fig. 3 & Fig. 4)

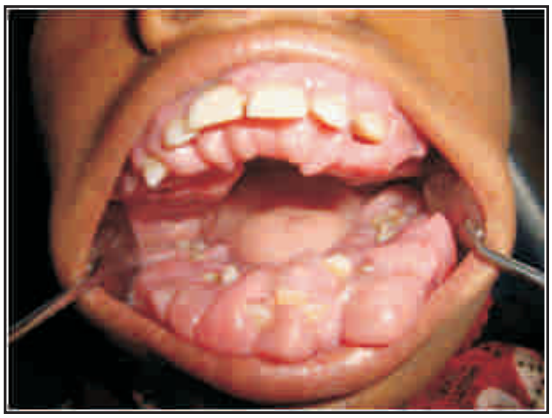

Fig 1: An intraoral examination revealed generalized, gross, nodular enlargement of the gingivae involving the upper and lower arches.

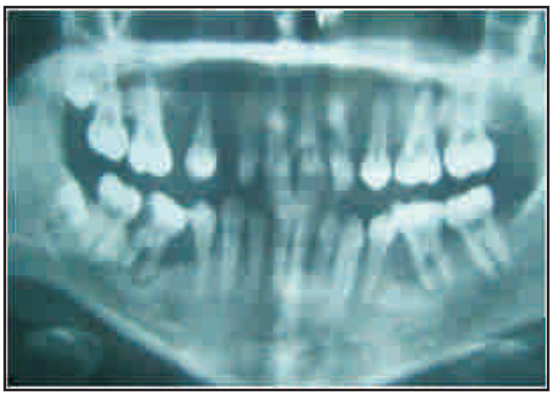

Fig 2: Radiographic findings revealed severe generalized alveolar bone loss

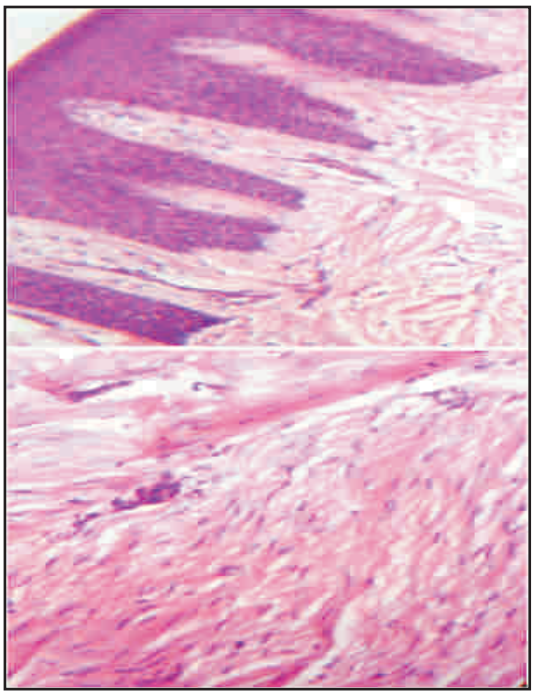

Fig3: Thickened and acanthotic overlying epithelium with elongated rete ridges and underlying connective tissue showed dense collagen fibre bundles with numerous fibroblasts

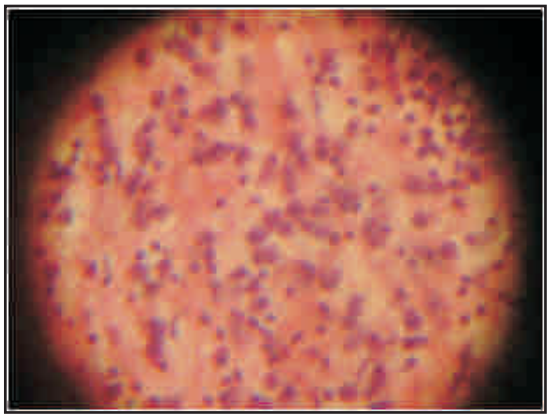

Fig 4: Chronic inflammatory cell infiltration predominantly composed of plasma cells.

Discussion

Gingival hyperplasia is a bizarre condition causing esthetic, functional, psychological, and masticatory disturbance of the oral cavity. Causes of gingival enlargement can be due to plaque accumulation, due to poor oral hygiene, inadequate nutrition, or systemic hormonal stimulation Gingival enlargements are also pragmatic in several blood dyscrasias such as leukaemia,

thrombocytopenia, or thrombocytopathy. A progressive fibrous enlargement of the gingiva is a facet of idiopathic fibrous hyperplasia of the gingiva. In modern times, 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. Gingival overgrowth varies from mild enlargement of isolated interdental papillae to segmental or uniform and marked enlargement affecting one or both of the jaws[4]. The mode of transmission is mainly autosomal dominant. The first polymorphic marker for hereditary gingival fibromatosis (HGF) phenotype in chromosome 2p21. Many cases are sporadic with no familial background.[5,6]

Gingival hyperplasia can occur after therapy with drugs like phenytoin, cyclosporine, nifidepine and nitrendipine. Long term use of these drugs has to be ruled out. The incidence of gingival enlargement caused by phenytoin, an anticonvulsant used in the treatment of epilepsy varies from 3 to 84.5%. whereas, cyclosporine a fairly potent immunosupressive agent used to prevent organ transplant rejection and to treat several disease of autoimmune origin induced gingival enlargement in 30% of the cases.Nifidepine, which is a calcium channel blocker used in the treatment of acute and chroniccoronary insufficiency, including angina pectoris and refractory hypertension and nitrendipine an analogue of nifedipine have also been reported to induced gingival enlargement.[7,8]

Aggressive periodontitis is typically characterized by familial aggregation because of evidence of genetic predisposition that was derived from segregation analysis of affected families. Mendelian inheritance occurs, and autosomal (dominant and recessive) transmission and X-linked transmission have been proposed. Presence of generalized aggressive periodontitis in a case of otherwise nonsyndromic gingival fibromatosis open savenues for research into the existence of a predictable association between the two entities. A case of hereditary gingival fibromatosis associated with generalized aggressive periodontitis reported earlier indicated the possible emergence of a new syndrome, but no definite genetic linkage could be established.[3]

The syndromes associated with GF include Murray-Puretic-Drescher syndrome (multiple hyaline fibromas), Rutherfurd syndrome (corneal dystrophy), Zimmermann-Laband syndrome (ear, nose, bone, and nail defects with hepatosple nomegaly), Jones' syndrome (progressive deafness), Cross syndrome (microphthalmia, mental retardation, athetosis, and hypopigmentation); Cornelia de Lange syndrome (primordial growth deficiency, severe mental retardation, anomalies of the extremities, and a characteristic face) , and Ramon syndrome (association with cherubism). A syndrome associated with hearing deficiencies, hypertelorism, and supernumerary teeth has been reported by Wynne *et al* and Takagi *et al*. Other associations include hypothyroidism, chondrodystrophia, and diffuse osteofibromatosis (GF with osteofibrosis) . Our patient demonstrated no clinical features fulfilling any of these possible syndromes [9].

Enlargement usually begins with the eruption of the permanent dentition but can develop with the eruption of the deciduous dentition; rarely, it may present at birth or arise in adulthood 11. The age at onset is divided into the pre-eruptive period (<6 months), deciduous dentition period (6 months-6 years), mixed dentition period (6-12years), permanent dentition period before adolescence (12-20 years), and permanent dentition period after adolescence (>20 years)[10] . Maximal enlargement occurs either during the loss of deciduous teeth or in the early stages of the eruption of permanent teeth. It progresses rapidly during 'active' eruption and decreases with the end of this stage[11].

Clinically, in the plasma cell gingivitis, the gingiva appear red, friable and bleed easily; usually it does not induce loss of attachment. Histologically, a dense infiltration of the normal plasma cells in the connective tissue is a common finding. A hypersensitivity reaction to some antigens, often flavorings or spices, is generally recognized. In this case, a rapidly progressive loss of attachment was observed, so rapidly progressive periodontitis was diagnosed. Differential diagnosis of the plasma cell gingivitis could be determined by histological and ultrastructural examination. Allergens, however, could not be identified. Conventional periodontal therapy, including intensive plaque control, could not cure the plasma cell gingivitis completely but recurrence of gingival enlargement and loss of attachment could be well controlled[12].

Treatment of gingival hyperplasia is essential because it causes difficulties with mastication, speech problems, malpositioning of teeth, esthetic effects and psychological difficulties for the patient. In a case of gingival enlargement, with deep pockets and severe loss of underlying alveolar bone, an internal bevel gingivectomy with open-flap debridement is indicated. This surgical procedure was done for our patient to eliminate pockets, making plaque control easier; to reduce the bulbous gingival tissues; and to enable the regeneration or repair of the alveolar bone defect. Our patient is being regularly monitored clinically and radiographically for improvement in his periodontal condition, as well as for any recurrence of gingival overgrowth.

Conclusion

This case highlights the unusual coexistence of nonsyndromic gingival fibromatosis with generalized aggressive periodontitis and plasma cell gingivitis. Diagnosis was based on clinical, radiographic and histopathologic assessment. However, further research is needed to establish a syndromic association between the two conditions based on genetic evaluation and linkage studies.

References

1. *Indu Shekar Idiopathic gingival fibromatosis. Saudi Dental Journal 2002;14(3):143-5.*
2. *Ferhat Cekmez, Ozgur Pirgon, Ilhan Asya Tanju Idiopathic Gingival Hyperplasia. Int J Biomed Sci 2009; 5(2):198-200.*
3. *Rashi Chaturvedi.Idiopathic Gingival Fibromatosis Associated with Generalized Aggressive Periodontitis: A Case Report JCDA 2009;75(4):291*
4. *Prashant Jaju,Ankit Desai,Rajiv Desai,Sushma Jaju Idiopathic Gingival Fibromatosis :Case report and its management.Int J Dent 2009;2009.*
5. *Emerson TG. Hereditary gingival hyperplasia. A family pedigree of four generations. Oral Surg 1965;19:1.*
6. *Jorgenson RJ, Cocker ME. Variations in the inheritance and expression of the gingival fibromatosis. J Periodontol 1974;45:472-477.*
7. *Barclay S, Thomason JM, Idle JR, Seymour RA. Incidence and severity of nifedipine induced gingival Overgrowth. J Clin Periodontol 1992; 19: 311.*
8. *Brown RS, Sein P, Corio R, Bottomley WK. Nitrendipine - induced gingival hyperplasis. Oral Surg 1990; 70: 593.*
9. *Pappachan B, Narayan J,Nayak A Idiopathic Gingival Fibromatosis:A neglected case.Indian J Radiol Imaging 2002;12:335-8.*
10. *Anderson J, Cunliffe WJ, Roberts DF, Close H. Hereditary gingival fibromatosis. Br Med J 1969;3:218-9.*

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